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EXPLORING RESILIENCE AND ADAPTATION IN ADOLESCENTS WITH SICKLE CELL
DISEASE

by

LAURA A. COUSINS

Under the Direction of Lindsey L. Cohen (PhD)

ABSTRACT

Living with sickle cell disease (SCD) can be a significant adversity due to disease-related symptoms and complications. Compounding these challenges, SCD predominantly affects ethnic minority populations already vulnerable to societal stigmatization, discrimination, and health disparities. It is important to recognize the negative impact of this chronic illness on psychosocial functioning; however, there is value in utilizing a strengths-based approach to determine how to promote adaptation to a challenging life-long disease. The current study explored the association among pain characteristics, adolescent, caregiver, and family protective factors, and functioning outcomes. Another primary aim of this study was to apply the protective factor model of resilience based in resilience theory to pediatric SCD by evaluating the

moderating effect of adolescent, caregiver, and family protective factors on the relation between SCD pain burden and functioning outcomes. 93 12- to 18-year-olds with SCD and their caregivers were recruited from a large Southeastern children's hospital. Adolescents completed measures assessing pain intensity and frequency, general and pain-specific protective factors, and functional outcomes. Caregivers completed measures assessing demographic and disease variables, psychological flexibility, and family functioning. Correlation analyses revealed that the majority of variables were related in expected directions and supported previous research. Adolescent protective factors were generally associated with one another and increased functional ability and quality of life. With the exception of family functioning, caregiver and family variables were not related to primary outcomes. After controlling for demographic, pain, and disease variables, moderation analyses showed that adolescent pain acceptance buffered the relation between SCD pain burden and quality of life. Contrary to hypotheses, moderating effects were not significant for the remaining adolescent, caregiver, and family protective factors. Findings highlight the importance of continuing to identify individual, caregiver, family, and broader environmental protective factors and evaluate resilience mechanisms among adolescents with SCD. Pain acceptance may also be a critical variable to target in future pain-focused interventions. Utilizing a strengths-based approach might lead to novel clinical avenues to empower youth to positively adapt to a chronic illness characterized by pain.

INDEX WORDS: Resilience, Adolescents, Sickle cell disease, Pain management

EXPLORING RESILIENCE AND ADAPATATION IN ADOLESCENTS WITH SICKLE
CELL DISEASE

by

LAURA A. COUSINS

A Dissertation Submitted in Partial Fulfillment of the Requirements for the Degree of

Doctor of Philosophy

in the College of Arts and Sciences

Georgia State University

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Laura Ann Cousins
2017

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DISEASE

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DEDICATION

This work is dedicated to all youth with sickle cell disease and their families for demonstrating remarkable strength.

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TABLE OF CONTENTS

ACKNOWLEDGEMENTS	v
LIST OF TABLES	ix
LIST OF FIGURES	x
1 INTRODUCTION.....	1
1.1 Overview of Pediatric Sickle Cell Disease.....	1
1.2 Pediatric Sickle Cell Pain	1
1.3 Resilience Theory	3
1.4 Resilience in Pediatric Sickle Cell Disease	4
1.5 Ecological Resilience-Risk Model	7
<i>1.5.1 Individual Protective Factors</i>	<i>7</i>
<i>1.5.2 Caregiver and Family Protective Factors</i>	<i>15</i>
<i>1.5.3 Resilience Outcomes</i>	<i>18</i>
1.6 Current Study.....	20
1.7 Primary Aims and Hypotheses	21
2 METHOD	23
2.1 Participants.....	23
2.2 Measures	24
<i>2.2.1 Covariates.....</i>	<i>24</i>
<i>2.2.2 Adolescent Pain-Specific Risk Factor.....</i>	<i>25</i>

2.2.3	<i>Adolescent Protective Factors</i>	26
2.2.4	<i>Caregiver and Family Protective Factors</i>	29
2.2.5	<i>Functioning Outcomes</i>	30
2.3	Procedures.....	31
3	DATA ANALYSES.....	33
3.1	Preliminary Analyses	33
3.2	Primary Analyses	33
4	RESULTS	34
4.1	Preliminary and Descriptive Analyses	34
4.2	Primary Analyses	35
4.2.1	<i>Primary Aim 1</i>	35
4.2.2	<i>Primary Aim 2</i>	37
4.2.1.1	<i>Adolescent Protective Factors as Moderators</i>	37
4.2.1.2	<i>Caregiver and Family Protective Factors as Moderators</i>	39
5	DISCUSSION	48
5.1	Overview	48
5.2	Preliminary Analyses	49
5.3	Primary Aim 1	51
5.4	Primary Aim 2	54
5.5	Limitations and Future Directions	55

5.6 Summary	59
REFERENCES.....	61
APPENDICES	85
Appendix A. Background Information Form.....	85
Appendix B. Pain Intensity and Frequency	88

LIST OF TABLES

Table 1: Participant Demographic Information (N = 93)	41
Table 2: Descriptives of Pain and Outcome Study Variables.....	42
Table 3: Intercorrelations Among Study Variables	43

LIST OF FIGURES

<i>Figure 1.</i> Protective Factor Resilience Model	44
<i>Figure 2.</i> Ecological Resilience-Risk Model in Pediatric Pain	45
<i>Figure 3.</i> Resilience Pathways Tested in Current Study	46
<i>Figure 4.</i> The Moderating Effect of Adolescent Pain Acceptance on the Relation Between Pain Burden and Quality of Life	47

1 INTRODUCTION

1.1 Overview of Pediatric Sickle Cell Disease

Sickle Cell Disease (SCD) is an inherited genetic blood disorder that predominantly affects individuals whose families originate from Africa, South or Central America, Caribbean islands, India, Saudi Arabia, and countries in the Mediterranean. It is estimated that 70,000-100,000 individuals in the United States have SCD and that the disease occurs in approximately 1 out of every 500 African American births and 1 out of every 36,000 Hispanic American births (NHLBI, 2012). SCD is characterized by a mutation in the hemoglobin gene, which causes the red blood cells to assume a sickle shape (NHLBI, 2012). Sickle cells are less flexible and restrict blood flow, producing vascular occlusions that can lead to acute and chronic complications including pain episodes, cerebro-vascular attacks, chronic anemia, acute chest syndrome, growth retardation, progressive deterioration of major organs, leg ulcers, and aseptic necrosis of bone (Lemanek, Buckloh, Woods, & Butler, 1995). Currently, there is no widely available cure for SCD, however bone marrow and stem cell transplants provide a potential cure for a limited number of those living with the disease. Treatments are primarily used to manage complications, assist with symptom relief, such as pain, and prevent the occurrence of infections, organ damage, and strokes (NHLBI, 2012).

1.2 Pediatric Sickle Cell Pain

Pain is the hallmark feature of SCD and it is reported as the most frequent, unpredictable, and debilitating symptom among youth with SCD (Fuggle, Shand, Gill, & Davis, 1996). Acute pain episodes result from vaso-occlusive crises (VOCs; Shapiro & Ballas, 1994) caused by blocked blood flow, while chronic pain episodes can develop from VOCs due to damage from repeated episodes and tissue ischemia (Franck, Treadwell, Jacob, & Vichinsky, 2002). Such

VOCs produce unpredictable and distressing pain episodes that vary in frequency, severity, duration, and location based on age, disease genotype, and disease severity (Shapiro, 1993; Shapiro & Ballas, 1994). Furthermore, VOCs account for approximately 25% of emergency room visits and hospital admissions among youth with SCD (Frush, Ware, & Kinney, 1995; Rees et al., 2003). VOCs are routinely treated using nonsteroidal anti-inflammatory drugs, opioids, and adjuvant medications that promote analgesic effects and minimize side effects (Benjamin et al., 1999); however, transfusion therapy may be utilized in patients with recurrent, chronic, or severe pain (Styles & Vichinsky, 1994). A recent study that classified youth with SCD into three groups based on pain duration and frequency (chronic, episodic, no SCD pain in the past month) revealed that youth with chronic sickle cell pain endorsed more disability, depressive symptoms, and inpatient hospital admissions compared to the other two pain groups (Sil, Cohen, & Dampier, 2016). However, both the chronic and episodic pain groups reported similar pain intensity, pain catastrophizing, and health-related quality of life (Sil et al., 2016). Given that SCD can be impacted by both physiological and psychosocial factors, multidisciplinary treatments often involve physical strategies (e.g., heating pads, massages, fluid intake) and psychosocial interventions (e.g., cognitive-behavioral therapy, biofeedback, hypnosis, relaxation techniques) in addition to analgesic treatment. Research suggests that analgesic treatments may not successfully provide complete pain relief, and 60-90% of painful episodes are treated at home (e.g., Dampier, Ely, Brodecki, & O'Neal, 2002; Fuggle, Shand, Gill, & Davies, 1996). Thus, SCD pain management remains challenging for healthcare providers, patients, and caregivers given its inconsistent response to treatment and potential severity (Gil et al., 1997). As the mortality rates of youth with SCD have sharply declined over recent decades due to medical

advances (Yanni et al., 2009), research has shifted its focus on examining the psychosocial ramifications of the disease to promoting psychosocial adaptation.

1.3 Resilience Theory

Resilience is defined as a person's ability to respond effectively to risk or adversity (Masten, 2001). Resilience is a dynamic and multi-systemic process that originates within the individual and is enhanced through developmental, social, cultural, and environmental factors (Masten, 2001). Through this process, both risk and promotive factors must be present to elicit a positive outcome or mitigate or entirely eliminate a negative outcome (Fergus & Zimmerman, 2005). For example, an adolescent that attains positive outcomes in the presence of low risk follows a pathway consistent with normative development. However, an adolescent presented with high risk who still obtains positive outcomes follows a resilient pathway. According to resilience theory, promotive factors that buffer the impact of risks are classified as assets or resources (Beauvais & Oetting, 1999). While assets represent positive factors inherent in an individual (e.g., coping skills, self-efficacy), resources are external positive factors within the individual's environment (e.g., social support, community resources) (Sandler et al., 2003).

Three theoretical models of resilience illustrate the various ways promotive factors may influence and change risk pathways (i.e., the likelihood that a risk factor will lead to a negative outcome) (Garmezy, Masten, & Tellegen, 1984; Rutter, 1985; Zimmerman & Arunkumar, 1994). The *compensatory model* of resilience proposes that a promotive factor has an independent direct effect on the outcome, countering the impact of a risk factor (Zimmerman & Arunkumar, 1994). Statistically, this model is often tested through multiple regression analysis or structural equation modeling. The *protective factor* model suggests that assets or resources serve as moderators that weaken the effect of a risk factor on a negative outcome. This model is evaluated by creating an

interaction term using multiple regression or through structural equation modeling. Within the protective factor model, researchers have differentiated two sub-types, *protective-stabilizing* and *protective-reactive* (Luthar, Cicchetti, & Becker, 2000). The protective-stabilizing model represents situations where a protective factor completely eliminates the negative impact of a risk factor (i.e., no relation between the risk factor and outcome exists in the presence of the protective factor). In contrast, the protective-reactive model illustrates situations where a protective factor minimizes the association between a risk factor and an outcome. The third resilience model is the *challenge model* (Garmezy et al., 1984), which depicts a curvilinear relation between a risk factor and an outcome. More specifically, this model proposes that both low and high levels of a risk factor contribute to negative outcomes, while moderate levels of a risk factor contribute to better outcomes (Luthar & Zelazo, 2003). In the challenge model, risk and promotive factors are equivalent and depend on the level of exposure. Challenge models are evaluated statistically using polynomial terms in multiple regression. When the challenge model is placed within a longitudinal context, it resembles the inoculation model (Masten, 1999; Rutter, 1987; Zimmerman & Arunkumar, 1994). This model theorizes that repeated exposure to low levels of risk over time enables youth to more effectively confront adversity.

1.4 Resilience in Pediatric Sickle Cell Disease

Unpredictable SCD pain episodes and disease complications can interfere with youths' daily functioning, including academic achievement, participation in activities, peer relations, and family functioning (Fuggle, Shand, Gill, & Davis, 1996). Considerable pediatric research has focused on risk factors and maladjustment; in contrast, there has been a dearth of attention on variables that promote resilience and adaptation (Barakat, Lash, Lutz, & Colette Nicolaou, 2006). Utilizing a strengths-based approach that captures resilience is particularly salient in the

context of the African American culture. African Americans have developed a culture built on resilience, community support, and spirituality to confront a history of oppression, discrimination, and hardships (Caldwell-Colbert, Parks, & Eshun, 2009). Karlson et al. (2012) examined psychosocial risk in a pediatric SCD sample over one year. Interestingly, the majority of families in the sample reported low-risk scores and in fact risk for psychosocial distress generally decreased over the course of the year. However, results revealed that older child age, lower caregiver educational attainment, caregiver divorce, family member composition (fewer adults and more children in the home), and financial challenges contributed to the highest risk for psychosocial distress among families with SCD. Indeed, youth with SCD are at risk for poorer health-related quality of life, psychological distress (symptoms of anxiety and depression), and social difficulties (Benton et al., 2007; Gold et al., 2008; Hijmans et al., 2010; Trzepacz et al., 2004). Given the prevalence of mental and physical health disparities, it is crucial for future research to explore factors that promote strength and resilience in the SCD population.

Within the context of pediatric SCD, resilience can be conceptualized as the individual resources and effective responding that protect from dysfunction, lead to adaptation, or result in well-being and growth. The introduction for the special issue on resilience in the *Journal of Pediatric Psychology* defines resilience in pediatric psychology as “the demonstration of emotional, behavioral, or health outcomes that match or surpass normative developmental milestones, behavioral functioning, or emotional well-being, despite exposure to the substantial challenges of living with and managing a medical or developmental condition” (Hilliard, McQuaid, Nabors, & Hood, 2015).

Resilience models have been applied to several pediatric chronic illness populations, such as asthma (Koinis-Mitchell et al., 2013) and diabetes (Hilliard, Harris, & Weissberg-Benchell,

2012), and prior research has applied Wallander et al.'s (1989) risk-resistance adaptation model to assess adaptation and adjustment in children with SCD and their caregivers (Brown, Doepke, & Kaslow, 1993; Brown et al., 2000). Protective factors included within the risk-resistance adaptation model include intrapersonal factors (e.g., social and academic competence), social-ecological factors (e.g., family environment), and stress processing factors (e.g., cognitive appraisal) (Wallander et al., 1989). Among children with SCD and their caregivers, caregiver coping strategies were associated with caregiver adjustment and internal health locus of control was the best predictor of children's adaptation, however this study exclusively relied on caregiver report for both caregiver and child factors (Brown et al., 2000). Notably, another study found that psychosocial factors (i.e., intrapersonal, stress-processing, and social ecological) selected from the transactional stress and coping model (Thompson & Gustafson, 1996) and the Disability-Stress-Coping Model (Wallander & Varni, 1992) were better predictors of adaptation compared to biomedical risk factors (e.g., disease severity) (Burlew, Telfair, Colangelo, & Wright, 2000). Despite the previous application of Wallander et al.'s risk-resistance adaptation model and stress and coping models to the pediatric SCD population, a resilience model specific to pediatric SCD-related pain and adjustment has not been tested.

In the context of pain, Sturgeon and Zautra (2013) developed the predominant risk-resilience model for adults with chronic pain. Within this model, resilience resources are stable individual trait characteristics (e.g., optimism, mindfulness) or social situations (e.g., positive family relationships) that promote effective adaptation to adversity by influencing resilience mechanisms. Resilience mechanisms are modifiable, dynamic processes (i.e., cognitions, affect, behaviors) that enhance adaptive coping in response to pain, which promote resilience outcomes, such as sustainability and growth.

1.5 Ecological Resilience-Risk Model

An ecological resilience-risk model for pediatric pain was recently proposed (Cousins et al., 2015) that maintains model pathways within Sturgeon and Zautra's adult chronic pain risk-resilience model, but adapts this model to pediatric populations by (a) integrating individual and social/environmental variables that have been previously identified or received empirical support in the pediatric pain literature and (b) providing an ecological context (Figure 2). Ultimately, the model serves as a framework for testing the applicability of protective factors and potential resilience pathways that contribute to pain adaptation and improved pain/disease management. Given that semi-structured interviews have revealed that adolescents with SCD identify pain and pain management as their primary complaint related to living with SCD (Ware et al., 2014), the ecological resilience-risk model for pain might be a viable framework to better understand responses to pain in this population. For the purposes of this study, only prominent constructs within the individual and family/social environment levels will be further discussed.

1.5.1 Individual Protective Factors

The most recognized and studied resilience constructs within pain populations include optimism, mindfulness, and pain acceptance. Optimism, defined as generalized favorable expectancies for the future (Scheier & Carver, 1985), predicts superior physical and psychological outcomes (Rasmussen et al., 2009) as well as pain-related adjustment and adaptation among adults and youth with chronic pain (Cousins, Cohen, & Venable, 2015; Goodin & Bulls, 2013). Despite optimism's health benefits, it has rarely been examined in populations with SCD. In adults with SCD who were recruited during outpatient sickle cell-related clinic visits, when controlling for age and pain intensity, optimism predicted increased positive affect and spirituality and decreased perceived stress (Bediako & Neblett Jr., 2011).

Bediako and Neblett Jr. (2011) highlighted the need to examine mechanisms that explain optimism's impact on positive adjustment and re-conceptualize adjustment utilizing a strengths-based approach in SCD research. Among adolescents with SCD who completed daily diaries over a 3-month period, Pence et al. (2007) found that optimism moderated the relation between pain intensity and medication use such that adolescents with moderate to high levels of optimism used medications more frequently in accordance with their reported pain severity. Additionally, negative thinking, a maladaptive pain coping strategy, mediated the relation between pain intensity and depression and pain interference in the context of daily activities and anxiety in a sample of adolescents with SCD (Barakat, Schwartz, Simon, & Radcliffe, 2007). Given the detrimental impact of negative thinking on SCD pain adaptation, it is important to examine the potential protective role of optimism in the context of SCD-related pain. Similarly, another study highlighted the importance of examining the effect of stress processing factors (i.e., appraisals and coping) on resilience and adaptation among adolescents with SCD and suggested that interventions targeting optimism may contribute to enhanced resilience (Ziadni, Patterson, Pulgarón, Robinson, & Barakat, 2011).

Mindfulness, the nonjudgmental focus on and acceptance of present moment experiences (Kabat-Zinn, 1996), has not only been operationalized as a cognitive process refined through meditative practice, but also represents an individual disposition or trait (Brown & Ryan, 2004; Brown et al., 2007). In adults, mindfulness is associated with increased psychosocial functioning, specifically greater life satisfaction, self-esteem, positive affect, empathy, and optimism (Bowlin and Baer 2011; Brown and Ryan 2003; Dekeyser et al. 2008; Rasmussen & Pidgeon 2011; Thompson & Waltz 2007). Although mindfulness research in child and adolescent populations is limited, studies have illustrated that mindfulness is correlated with the reduced likelihood of

engagement in risky health behaviors among adolescents with poor decision-making (Black et al., 2012a), moderates the relation between adolescent stress and dysphoric mood (Ciesla et al., 2012), and predicts reduced negative affect and smoking behaviors (Black et al., 2012b). Additionally, a recent study found that mindfulness was associated with executive function processes, specifically working memory and inhibitory control, among a diverse sample of adolescents (Riggs, Black, & Ritt-Olson, 2014). These findings suggest that mindfulness may be related to high-order cognitive processes that promote self-regulation and pursuit of goal-oriented thoughts and behaviors.

In the context of pain, numerous studies have provided empirical support for the role of mindfulness in the reduction of pain perception and pain management enhancement (Brown et al., 2007; Grant, Courtemarche, Duerden, Duncan, & Rainville, 2010; Grant & Rainville, 2009; Zeidan et al., 2011). Among college students, mindfulness mediated the relations between pain severity and catastrophizing and pain severity and pain-related impairment (Mun, Okun, & Karoly, 2014). Substantial research evidence supports the negative relation between mindfulness and maladaptive cognitive processing that exacerbates pain-related impairment, such as pain catastrophizing. Pain catastrophizing is an exaggerated adverse and fearful appraisal of both present and anticipated pain, comprised of rumination, magnification, and helplessness (Sullivan, Bishop, & Pivik, 1995; Sullivan et al., 2001). The influence of mindfulness on attentional processes may specifically account for these beneficial effects. Among adults with chronic pain, mindfulness was associated with lower reported pain, higher pain management self-efficacy, and increased emotional intelligence (Wright & Schutte, 2014). Furthermore, both emotional intelligence and pain management self-efficacy mediated the relation between mindfulness and pain, supporting the notion that mindfulness reduces pain by contributing to enhanced positive

emotional functioning and behavioral regulation (Keng et al., 2011; Wright & Schutte, 2014). Research has suggested that mindfulness may improve metacognitive awareness, attentional control, and engagement with valued behaviors (Keng et al., 2011).

Within an adolescent community sample, in the context of daily pain, mindfulness has been shown to be a unique predictor of decreased pain interference, partially mediated by pain catastrophizing (Petter, Chambers, McGrath, & Dick, 2013). Furthermore, in the context of an experimental cold pressor pain task, mindfulness was indirectly related to pain intensity and pain tolerance, mediated by state pain catastrophizing. This study illustrates that one primary mechanism through which mindfulness exerts its beneficial effects (i.e., decrease pain interference and pain intensity and increase pain tolerance) is by reducing catastrophic thoughts. Given the empirical support for mindfulness as a resilience resource in the context of pain and the dearth of literature examining mindfulness in the sickle cell population, it is imperative that studies begin to assess this construct and its relation to pediatric sickle cell adaptation.

The psychological flexibility model (McCracken & Morley, 2014) has recently been applied to research and treatment for pain. Psychological flexibility is the ability to be present-focused and act effectively and consistently with personal values in the presence of interfering thoughts, emotions, and bodily sensations. Acceptance and Commitment Therapy (ACT; Hayes, Strosahl, & Wilson, 1999) is a recent cognitive-behavioral treatment approach based on the psychological flexibility model that aims to increase psychological flexibility and related processes. One of the interrelated processes underlying psychological flexibility is pain acceptance, an individual's willingness to live life with pain without efforts to control or avoid it in order to pursue a life consistent with personal values.

Notably, using multilevel structural equation modeling, pain-related acceptance was

recently shown to mediate changes in pain-related outcomes (i.e., pain interference, pain intensity, and depression) over time among adults participating in a 5-week cognitive-behavioral therapy (CBT)-based multidisciplinary pain treatment program (Åkerblom, Perrin, Rivano Fischer, & McCracken, 2015). Furthermore, pain-related acceptance was the strongest mediator across outcomes when compared to three variables proposed to serve as potential mediators in CBT treatment (i.e., life control, affective distress, and social support). These results suggest that pain-related acceptance may represent a common mechanism of change responsible for improvements in functioning following CBT-based treatments (Åkerblom et al., 2015).

Prior research has shown that increased pain-related acceptance is predictive of better functioning and reduced distress among adolescents with chronic pain (McCracken, Gauntlett-Gilbert, & Eccleston, 2010). Several studies (i.e., case report, open trial, randomized controlled trial) have utilized an ACT-based treatment approach for youth with chronic pain (Wicksell, Dahl, Magnusson, & Olsson, 2005; Wicksell, Melin, Lekander, & Olsson, 2009; Wicksell, Melin, & Olsson, 2007). Wicksell et al. (2009) found that a 10-session ACT intervention significantly improved functioning when compared to a multidisciplinary approach incorporating medication. With regard to processes of change in ACT treatment for pediatric chronic pain, one study found that variables consistent with the psychological flexibility framework (i.e., pain impairment beliefs, pain reactivity) were responsible for treatment improvements and mediated effects of treatment on follow-up outcomes (i.e., pain interference, depression) (Wicksell, Olsson, & Hayes, 2011). Gauntlett-Gilbert et al. (2013) demonstrated that an uncontrolled trial of 3-week residential group interdisciplinary intensive ACT treatment among a group of severely disabled adolescents with chronic pain improved functioning (both self-reported and objective physical performance), anxiety and pain catastrophizing, school attendance, and health care

utilization at 3-month follow-up. Additionally, improvements in outcomes from pre-treatment assessment until 3-month follow-up were associated with increased acceptance during treatment (Gauntlett-Gilbert et al., 2013).

A case study examining an eight-session ACT program for an adolescent with SCD and his parents found that adolescent psychological flexibility and parent acceptance likely explained improvements in functioning and quality of life that persisted at 3-month follow-up (Masuda, Cohen, Wicksell, Kemani, & Johnson, 2011). In sum, prior research has supported the value of utilizing an ACT approach and assessing acceptance among youth with medical conditions, specifically those with pain as a primary component (Wicksell, Kanstrup, Kemani, Holmström, & Olsson, 2015), however additional research is needed.

Benefit finding or post-traumatic growth refers to the positive changes or gains (e.g., sense of purpose, deepened relationships) associated with a significant adversity (Sears, Stanton, & Dandoff-Burg, 2003). This construct has primarily been studied in adult cancer populations (Stanton, Bower, & Low, 2006). A meta-analysis examined the relation between benefit finding and psychological and physical health and identified moderators of these relations exclusively among studies with adult samples (Helgeson, Reynolds, & Tomich, 2006). This meta-analysis revealed that lower depression, higher positive well-being, and heightened intrusive and avoidant thoughts about the stressor were all associated with higher benefit finding. Higher levels of benefit finding were also positively correlated with disease severity and subjective perceptions of stress, optimism, and religiosity. Moderators of the relation between benefit finding and health outcomes included time elapsed since the stressor or trauma and race/ethnicity, with longer time elapsed related to more positive outcomes. Interestingly, benefit finding had a stronger correlation with positive mental health outcomes when participant samples primarily included

racial/ethnic minorities and these participants engaged in higher levels of benefit finding. In terms of demographic correlates, marital status and socioeconomic status were not related to benefit finding, however women, racial/ethnic minorities, and younger participants were found to engage in the most benefit finding. Although results from this meta-analysis are limited given the predominance of cross-sectional studies examined, the authors highlight that benefit finding is a construct that should be further explored in child and adolescent populations and may be a construct that is more adaptive and relevant for racial/ethnic minorities (Helgeson et al., 2006).

The construct of benefit finding has recently been applied to pediatric populations, primarily pediatric cancer (Phipps, Long, & Ogden, 2007). Phipps et al. (2007) demonstrated that higher optimism and self-esteem and lower trait anxiety are associated with higher benefit finding among youth with cancer. With regard to demographic variables, benefit finding did not differ based on age, gender, or socioeconomic status; however, in terms of race/ethnicity, African American youth reported greater benefit finding. Additionally, older age at diagnosis and shorter time elapsed since diagnosis was related to higher levels of benefit finding. Interestingly, benefit finding was unrelated to post-traumatic stress symptoms or other domains of health-related quality of life (Phipps et al., 2007). Similarly, Barakat et al. (2006) found that a majority of adolescent cancer survivors and their families reported post-traumatic growth at least one year following cancer treatment. In a study examining pediatric cancer survivors, the authors found that leukemia diagnosis, increased optimism, and perceiving enduring effects of having cancer on daily living were related to increased benefit finding (Michel, Taylor, Absolom, & Eiser, 2010). Interestingly, adolescents' benefit finding and parents' post-traumatic growth were unrelated. A recent study also showed that among young adult survivors of childhood cancer recruited from three pediatric oncology medical treatment centers, females, nonwhite survivors,

and individuals diagnosed at an older age reported higher levels of post-traumatic growth (Yi, Zebrack, Kim, & Cousino, 2015). Furthermore, after controlling for demographic and cancer-related factors, optimism and social support remained associated with increased post-traumatic growth. In a qualitative study investigating the adolescent cancer experience among adolescent and young adult cancer survivors, benefit finding represented one of the emerging themes and perceived benefits included improved personal attributes, strengthened interpersonal relationships, and material gains (Wicks & Mitchell, 2010).

In addition to pediatric cancer, benefit finding has been shown to mitigate the maladaptive impact of negative affect and emotions to stress on type 1 diabetes management among adolescents (Tran, Wiebe, Fortenberry, Butler, & Berg, 2011). Findings revealed that reduced depressive symptoms, greater perceived coping effectiveness, better adherence, and higher positive and negative affect in response to diabetes-related stress were associated with higher benefit finding. Consistent with a stress-buffering process, benefit finding moderated the relation between negative affective reactions to diabetes-related stress and depressive symptoms and metabolic control. More specifically, negative affective reactions to stress were unrelated or less strongly associated with poor adjustment among adolescents with high benefit finding (Tran et al., 2011).

Surprisingly, benefit finding has been minimally studied in the SCD population. A recent study revealed that discussing pain episodes with siblings, intimate partners, or close friends was associated with increased benefit finding among adults with SCD (Derlega, Janda, Miranda, Chen, Goodman, & Smith, 2014). Furthermore, a number of adolescents with SCD identified benefits and positive experiences as a result of living with SCD including feeling “special,” “stronger and better,” and gaining a different perspective on life compared to their peers in a

qualitative study (Ware et al., 2014). Despite the adversity and challenges associated with living with SCD, the construct of benefit finding seems particularly relevant to this population given the generally low levels of maladjustment and cultural context.

1.5.2 Caregiver and Family Protective Factors

When applying resilience theory to pediatric psychology, it is also critical to consider an adolescent's social-ecological context. Palermo and Chambers (2005) created a multi-level theoretical model illustrating important parent and family factors to consider in the context of pediatric pain. Indeed, parents who have a child with persistent pain endorse elevated levels of parent role stress, anxiety and depressive symptoms, and also experience its considerable social impact (Palermo & Eccleston, 2009). Due to this high likelihood of familial strain, consideration of the family is critical in better understanding adolescents' pain trajectory and experience. While there has been substantial literature identifying parent and family emotions, cognitions, and behaviors that negatively impact an adolescent's pain experience, caregiver and family protective factors have received less empirical attention (Palermo, Valrie, & Karlson, 2014). Given this neglected area of research, it is important to evaluate parent and family constructs that promote pain-related coping, pain management, and disease adaptation.

One protective factor that has gained increasing attention and support in pediatric pain is parent psychological flexibility, comprised of values-based action, pain acceptance, emotional acceptance, and pain willingness (Wallace, McCracken, Weiss, & Harbeck-Weber, 2015). In other words, parent psychological flexibility assesses the ability to accept uncomfortable and distressing experiences related to witnessing your child in pain in order to persevere with valued behaviors. Cross-sectional studies have shown that parent psychological flexibility is negatively associated with parent protective pain responses, adolescent anxiety and depression, and

avoidance of social activities and school (McCracken & Gauntlett-Gilbert, 2011; Simons, Sieberg, & Kaczynski, 2011; Wallace et al., 2015). A recent pilot study developed an 8-week ACT-based group intervention for parents of adolescents with chronic pain to target parent psychological flexibility (Wallace, Woodford, & Connelly, 2016). Results revealed that this intervention improved parent psychological flexibility throughout the intervention and at 6-month follow-up, and contributed to declines in protective parenting responses and adolescent reported pain interference at follow-up. Findings suggest that modifying parent psychological flexibility, a cognitive factor, may be essential in order to subsequently change parent responses to pain and adolescents' perceived pain interference (Wallace et al., 2016).

At a broader level, family functioning is an important construct that has been examined in pediatric chronic pain and sickle cell populations. Families are conceptualized as organized systems striving to achieve balance and order through communication and designated roles (Fiese, Spagnola, & Everhart, 2008; Kazak, Rourke, & Crump, 2003). Family functioning encompasses the comprehensive social and structural properties contained within the family environment, such as the degree of conflict, cohesion (e.g., involvement and closeness), adaptability, organization (e.g., roles, leadership, and alliance formation), and communication quality (e.g., clarity of expression and directness) present in familial interactions and relationships (Alderfer et al., 2008). Family cohesion refers to both the emotional bonding and level of independence between family members, while family adaptability reflects the capacity to which families are able to exhibit flexibility when confronted with stressors (Patterson, 2002; Walsh, 2006). Such adaptability has been associated with parenting style, problem-solving approaches, and the transmission of beliefs and values within the family. Family relationships and interactions play a substantial role in promoting positive social and emotional development

and youth functioning. Clear communication, well-defined roles, cohesion, and affect regulation all contribute to healthy or adaptive family functioning, while increased conflict, disorganization, role rigidity, and poor affective and behavioral control predict poor or maladaptive family functioning (Alderfer et al., 2008).

Irrefutably, families that have a child with a chronic health condition are susceptible to significant stress and distress, impacting intrafamilial relationships, dynamics, and family structure as well as family influences on the child's health status (Alderfer & Kazak, 2006). Given the persistent, unpredictable nature of SCD and SCD-related pain, families may experience minimal relief from the ongoing demands of the condition and endure continual adaptation and adjustment of family roles. Overall, research has suggested that youth with SCD are more likely to experience positive adaptation within families who exhibit more cohesion, flexibility, and organization (Brown et al., 2000; Casey, Brown, & Bakeman, 2000; Thompson et al., 1999). Among adolescents with SCD, poorer family functioning was related to increased disease severity and healthcare utilization (Barakat et al., 2007). However, Schlenz et al. (2016) found that better general family functioning was unrelated to pain-specific healthcare utilization, but was negatively associated with child negative thinking and caregiver passive coping. Similarly, another study found that positive family functioning was unrelated to healthcare utilization (number of hospitalizations, emergency room visits, days of pain), but was associated with positive patient coping (Mitchell et al., 2007). Better family functioning is also correlated with higher parental internal locus of control, but was not associated with parent-reported child quality of life among parents who had a child admitted to the hospital for pain or fever (Barakat et al., 2005). Family problem solving and behavior control subscale measures of family

functioning are correlated with treatment adherence among children with SCD (Barakat, Smith-Whitley, & Ohene-Frempong, 2002).

A systematic review (Lewandowski, Palermo, Stinson, Handley, & Chambers, 2010) found significant differences in family functioning between families of children with chronic pain and healthy controls. Specifically, chronic pain families had less family cohesion and organization, higher conflict, and greater psychological distress. Poorer family functioning was also more consistently associated with increased child pain-related disability relative to child pain. However, it is worth noting that poor family functioning is not present in all families and only seems to characterize a specific subgroup of children with chronic pain (Scharff et al., 2005).

1.5.3 Resilience Outcomes

Resilience outcomes are classified into three primary domains: recovery (i.e., resumed functioning), sustainability (i.e., perseverance with valued activities), and growth (i.e., realization and better understanding of one's capabilities) (Reich, Zautra, & Hall, 2010). One of the primary limitations of the pediatric pain and chronic illness literature is the lack of measures developed to assess and capture outcomes consistent with resilience theory. However, two gold standard pediatric pain measures that fall within the domain of recovery and sustainability that will be referred to as "functioning outcomes" include functional ability and valued living/quality of life.

Measures that assess functional ability evaluate participation in daily or physical activities despite pain (Walker & Greene, 1991). Within pediatric SCD samples, research has shown that higher socioeconomic status and lower neighborhood economic distress predict better functional outcomes (Palermo, Riley, & Mitchell, 2008). Another study found that based on youth and parent report, youth with SCD endorsed lowest functional ability with physical

activities (e.g., running or walking the length of a football field, gym activities/playing sports) (Oliver-Carpenter, Barach, Crosby, Valenzuela, & Mitchell, 2011). Furthermore, in the context of disease management, both youth and parent report reflected that youth with higher functional ability (i.e., better functioning) experience decreased parent involvement with disease-management tasks (Oliver-Carpenter et al., 2011). Within this study, functional ability scores were comparable to those reported in other pediatric SCD samples (Palermo et al., 2008) and youth with chronic abdominal pain (Claar & Walker, 2006). Studies that have examined adolescent adaptation to SCD have neglected to assess functional ability as an outcome (e.g., Burlew et al., 2000) despite its significant impact on living with SCD and have instead evaluated negatively framed outcomes, such as anxiety and depression.

Health-related quality of life (HRQOL) is an important outcome to assess in pediatric populations because it assesses how a specific health condition impacts youth with regard to physical, emotional, and social functioning and detects changes in health status over time. Furthermore, measuring HRQOL allows researchers to determine which interventions may be effective in increasing HRQOL and whether HRQOL is responsive to change in improving patient's well-being. Research has consistently revealed that youth with SCD typically endorse impaired HRQOL given the impact of pain and disease complications on various physical and psychosocial domains of functioning (Barakat et al., 2006; Fuggle et al., 1996; Panepinto et al., 2005), which is even more pronounced in adolescence and young adulthood (Palermo, Schwartz, Drotar, & McGowan, 2002; Thomas & Taylor, 2002). Frequency of pain episodes between two time points has been illustrated as a predictor of decreased caregiver-reported HRQOL in children with SCD after controlling for time interval, demographic factors, and disease or medical factors (Schlenz, Schatz, McClellan, & Roberts, 2012). Barriers to treatment adherence

and pain crisis frequency have been shown to be some of the most robust predictors of poorer HRQOL in pediatric SCD (Fisak, Belkin, von Lehe, & Bansal, 2012). Additional research has also found that pain catastrophizing, anxiety, depression, and disease-related parenting stress are also associated with lower HRQOL in children and adolescents with SCD (Barakat et al., 2008; Lukombo et al., 2013). A recent study examined generic discrepancy QOL using the Generic Children's Quality of Life Measure, which assesses QOL in the general population as well as with children with health conditions or social difficulties, in a sample of youth with SCD in the United Kingdom and found that these youth did not report reduced QOL relative to their healthy peers and demographic and disease severity markers were minimally related to QOL (Constantinou, Payne, & Inusa, 2015). This study further highlights the need to understand how psychosocial factors may contribute to unexpected high QOL in youth with SCD.

1.6 Current Study

Living with SCD is often conceptualized as a significant adversity due to disease-related symptoms (e.g., pain, fatigue) and complications (e.g., stroke, organ damage) predominantly affecting ethnic minority populations already vulnerable to societal stigmatization, discrimination, and health and healthcare disparities. Although it is important to consider and recognize the negative impact of this chronic illness on psychosocial functioning, it may be valuable to utilize a strengths-based approach in order to determine how to promote adaptation in the context of experiencing a life-long disease. To support this strengths-based approach, many children and adolescents with SCD have demonstrated optimal functioning, adaptation, and resilience (Barakat, Lash, Lutz, & Nicolaou, 2006). Additionally, there has been a recent theoretical shift from primarily examining risk and vulnerability to assessing resilience and adaptation in pediatric psychology (Cousins et al., 2015; Hilliard et al., 2012; Koinis-Mitchell et

al., 2013). It seems particularly important to adopt a strengths-based approach when studying pediatric psychology populations, especially one that is predominately comprised of ethnic minorities. Given that African Americans gain substantial benefits from engagement in interventions that focus on empowerment and identification of strengths (Caldwell-Colbert, Parks, & Eshun, 2009), it follows that applying resilience theory and an ecological resilience-risk model is optimal for understanding protective factors among adolescents with SCD.

Although several prior studies have assessed risk and resistance variables in pediatric SCD, the current study uniquely applies the protective factor model, specifically the protective-reactive model (Luthar, Cicchetti, & Becker, 2000), within resilience theory in consideration of the ecological resilience-risk model (Cousins et al., 2015) to examine protective factors in the context of pain and disease management. The protective-reactive model was selected as it is anticipated that protective factors will reduce, but not completely eliminate the strength of the relation between pain-related risk and functioning outcomes in youth with SCD. Given the dearth of literature applying resilience theory to pediatric pain, research that provides empirical support for protective factors, particularly in understudied pain populations such as pediatric SCD, is needed. Utilizing a strengths-based approach to examine adolescents with SCD might lead to novel clinical avenues to empower youth to positively adapt to life with a chronic illness. Ultimately, focusing attention on constructs that promote pain-related resilience has tremendous implications for improving quality of life, growth, and functioning within a chronic illness group that is highly stigmatized yet demonstrates impressive strengths.

1.7 Primary Aims and Hypotheses

Primary Aim 1. To explore associations among pain variables, adolescent protective factors (mindfulness, optimism, pain acceptance, benefit finding), caregiver and family

protective factors (caregiver psychological flexibility, family functioning), and functioning outcomes (functional ability, quality of life) in adolescents with SCD.

Hypothesis 1. It is hypothesized that adolescent protective factors will be positively associated with caregiver and family protective factors and functioning outcomes (higher functional ability and quality of life), and negatively associated with pain variables.

Primary Aim 2. To investigate whether each adolescent, caregiver, and/or family protective factor independently moderates the relation between pain burden and functioning outcomes (functional ability, quality of life), consistent with the protective-reactive model of resilience (Figure 1).

Hypothesis 2. It is hypothesized that each adolescent, caregiver, and family protective factors will independently moderate the relation between pain burden and functioning outcomes (Figure 3). More specifically, consistent with the protective-reactive model within resilience theory, each adolescent, caregiver, and family protective factor will separately attenuate the relation between pain burden and functioning outcomes at higher levels of these variables.

2 METHOD

2.1 Participants

A power analysis was conducted using G*Power 3.1.3 (Faul, Erdfelder, Buchner, & Lang, 2009) and revealed that 67 participants would provide 80% power to detect a low to moderate correlation. With regard to moderation analyses, more than 200 participants are needed to detect medium interaction effects with measures that have reliabilities of .70 (Aiken, West, & Reno, 1991).

Inclusion criteria were that the patient was between 12 and 18 years of age and diagnosed with SCD. In addition, patients had to exhibit proficiency in English and were able and willing to provide verbal consent and comply with the requirements of the study protocol. Exclusion criteria included documented severe developmental or cognitive delays that prevented the participant from understanding study procedures and completing questionnaires. Both inclusion and exclusion criteria were confirmed through a review of the patient's medical record.

Participants were recruited through two recruitment methods. In the first recruitment method, participants were recruited from outpatient clinics at an urban children's hospital in the southeastern United States. In the second recruitment method, a list of patients from a cohort of approximately 100 families who participated in a previous SCD study and provided permission to be contacted about future research opportunities was generated and contacted. A total of 102 families were verbally consented and enrolled, however 9 (8.8%) of these families did not initiate study measures. Fifteen families declined participating in the current study. Based on the two recruitment methods, 65 families (63.7%) were recruited through the first recruitment method and 37 families (36.3%) were recruited using the second recruitment method. Of the total

sample, 67 families (72%) completed paper questionnaires and 26 families (28%) completed questionnaires online.

The final sample included 93 adolescents between the ages of 12 and 18 ($M = 15.23$ years, $SD = 1.97$ years) diagnosed with SCD and their caregivers (Table 1). Forty-nine (52.7%) youth were female and 44 (47.3%) were male. In terms of race, 92 (98.9%) participants were “Black or African American” and 1 (1.1%) participant was “Multiracial.” With regard to ethnicity, all participants identified as “Not Hispanic or Latino.” The majority of caregivers completing questionnaires were mothers ($n = 80$, 86%) and the remaining caregivers included fathers ($n = 11$, 11.8%) and step-fathers ($n = 1$, 1.1%). The most prevalent SCD genotype was HbSS ($n = 64$, 68.8%) followed by HbSC ($n = 17$, 18.3%), HbSB0 thalassemia ($n = 8$, 8.6%), and HbSB+ thalassemia ($n = 4$, 4.3%). Caregivers endorsed that 77 (82.8%) adolescents did not have a diagnosis of a psychological disorder, while 12 (12.9%) did have a diagnosis. Of these 12 adolescents, 5 had diagnoses of “anxiety,” two “ADHD,” two “depression,” and two multiple psychological diagnoses. With regard to annual family income, 5 (5.4%) caregivers reported an annual income at or below \$10,000.00, 14 (15.1%) ranged between \$10,001.00 and \$20,000.00, 15 (16.1%) between \$30,001.00 and \$40,000.00, 9 (9.7%) between \$40,001.00 and \$50,000.00, 3 (3.2%) between \$50,001.00 and \$60,000.00, 6 (6.5%) between \$60,001.00 and \$70,000.00, 4 (4.3%) between \$70,001.00 and \$80,000.00, 9 (9.7%) between \$80,001.00 and \$90,000.00, and 16 (17.2%) at or exceeded \$90,000.00. Twelve (12.9%) caregivers did not report income.

2.2 Measures

2.2.1 Covariates

Background Information. Caregivers completed the Background Information Form. This form includes questions about the parent (e.g., relation to child, gender, age, ethnicity, race,

education, occupation, family income, and health status) and the adolescent (e.g., gender, age, ethnicity, race, and health status). Caregivers also provided information about the adolescent's SCD (e.g., number of vaso-occlusive pain crises in the past year, history of stroke or other neuropsychological impairments, SCD-related complications experienced, days of school/work missed due to SCD in the past year) (Appendix A).

Pain Intensity and Frequency. To assess pain intensity, adolescents reported typical and worst intensity over the past month as well as current pain intensity using an 11-point numerical rating scale (NRS-11) from 0 (*no pain*) to 10 (*worst possible pain*). Numeric rating scales are well-validated measures in assessing self-reported pain intensity among youth (von Baeyer et al., 2009). Adolescents also responded to both open-ended and close-ended questions about their pain frequency (e.g., “How often do you have pain?” “How many days have you had pain in the past month?” “Was this a typical month of pain for you?”). Caregivers reported the number of days their child had experienced pain in the past month. Pain intensity composite scores were used in analyses and computed by averaging typical pain and worst pain intensity ratings. For pain frequency, adolescent report and parent proxy report of number of pain days in the past month were averaged and used in analyses (Appendix B).

2.2.2 Adolescent Pain-Specific Risk Factor

Pain Burden. Adolescents completed the Sickle Cell Disease Pain Burden Interview-Youth (SCPBI-Y; Zempsky et al., 2013), a 7-item disease-specific measure that evaluates the impact of sickle cell pain on physical, social/community, and emotional domains of daily functioning (e.g., “How many days have you been unable to do things you enjoy because of pain?”). This multidimensional interview was developed through collaboration with experts, patients, and caregivers. Responses are rated on a Likert scale (“none” = 0, “a few” = 1, “some”

= 2, “many” = 3, “every” = 4) and scores range from 0 (no pain) to 28 (severe pain burden). This measure has been validated among youth with SCD between the ages of 7 and 21 across four urban children’s hospitals in both inpatient and outpatient settings. The SCPBI-Y demonstrated strong internal consistency, cross-informant agreement between youth and their caregivers, and test-retest reliability. Furthermore, this measure exhibited moderate to strong construct validity and discriminant validity when compared to validated measures of mood, functional ability, pain, and quality of life. Finally, the SCPBI-Y accurately differentiated youth based on clinical setting (inpatient versus outpatient) and severity of SCD symptoms (i.e., youth in inpatient settings with higher disease severity endorsed elevated pain burden) (Zempsky et al., 2013) (Appendix C). In the current sample, Cronbach’s alpha was .91 indicating good reliability.

2.2.3 Adolescent Protective Factors

Mindfulness. The Child and Adolescent Mindfulness Measure (CAMM; Greco, Baer, & Smith, 2011) is a 10-item measure that assesses mindfulness skills (observing, acting with awareness, and accepting without judgment) among children and adolescents (e.g., “I keep myself busy so I don’t notice my thoughts or feelings”). The CAMM asked respondents to rate how often each item is true for them using a 5-point Likert-type scale (0 = *never true*; 4 = *always true*) and items are reverse scored. Scores range from 0 to 40, with higher scores indicating higher levels of mindfulness. The CAMM has been shown to be a developmentally appropriate, valid measure and exhibited good internal consistency ($\alpha = .846$; Greco et al., 2011) (Appendix D). Internal consistency in the current sample was good with a Cronbach’s alpha of .87.

Optimism. The Youth Life Orientation Test (YLOT; Ey et al., 2005) is a developmentally appropriate measure of optimism in youth, created as an analogue of the Life Orientation Test (Scheier & Carver, 1985), a well-established measure of optimism in adults. Adolescents rated

their agreement on seven optimism items (e.g., “I usually expect to have a good day”), seven pessimism items (e.g., “If something nice happens, chances are it won’t be to me”), and two filler items (e.g., “I like to be active”) using a 4-point Likert scale format (3-*true for me*, 2-*sort of true for me*, 1-*sort of not true for me*, 0-*not true for me*). Both a total score (i.e., global optimism) and subscale scores for optimism and pessimism can be calculated. The test-retest reliability of the YLOT ranged from .68 to .70 over a 1-month period and intraclass correlations of .65 to .75 across 7 months have been found (Ey et al., 2005). The two-factor structure and validity of the YLOT has been demonstrated with children with cancer and healthy controls between the ages of 7 and 18 years (Williams et al., 2010). Associations between the YLOT and measures of competency, hope, and psychological adjustment have also provided support for the YLOT’s convergent and discriminant validity (Ey et al., 2005). In this study, the optimism subscale was used as prior studies have recommended optimism and pessimism be examined separately given their differential impact on outcomes (Williams et al., 2010) (Appendix E). In the current sample, Cronbach’s alpha was .86 indicating good reliability for the optimism subscale.

Pain Acceptance. Acceptance of pain was measured using the adolescent version of the Chronic Pain Acceptance Questionnaire (CPAQ-A; McCracken, Gauntlett-Gilbert, & Eccleston, 2010). This measure contains 20 items (e.g., “When my pain increases, I can still do things I have to do”) rated on a 5-point scale ranging from 0 (*never true*) to 4 (*always true*). The total score is comprised of two subscales: activity engagement (11 items) and pain willingness (9 items). Activity engagement includes items that assess the extent to which adolescents attempt to participate in regular activities despite their pain and pain willingness evaluates the extent to which adolescents indicate that controlling or reducing pain is less important compared to other goals. CPAQ-A total scores range from 0 to 80, with higher scores reflecting greater acceptance

of pain. Items on the pain willingness subscale are reverse-scored. Psychological acceptance is conceptualized as the inverse of avoidance and cognitive fusion (Hayes et al., 2006). The CPAQ-A has demonstrated strong internal consistency and validity in adolescent samples with chronic pain (McCracken et al., 2010; Wallace, Harbeck-Weber, Whiteside, & Harrison, 2011). The total score was used in analyses (Appendix F). Internal consistency in the current sample was good with a Cronbach's alpha of .80.

Benefit Finding. Adolescents completed the Benefit Finding and Burden Scale for Children (BFBS-C; Currier, Hermes, & Phipps, 2009), a 20-item measure detailing the potential benefits of an illness (e.g., "Having had my sickle cell disease...has helped me become a stronger person") and illness-related burden (e.g., "Having had my sickle cell disease...has made me less hopeful about my life"). This measure is a revision of the Benefit Finding Scale for Children (BFSC; Phipps et al., 2007) and includes the same benefit finding items. However, the addition of burden items minimizes the likelihood that youth will report in a socially desirable way. This measure has two 10-item subscales, a benefit finding subscale and burden subscale. Items are rated on a 5-point Likert scale (1 = *not at all* to 5 = *very much*), with higher scores indicating more benefit finding or illness-related burden. These subscales have been shown to remain uncorrelated with one another and demonstrated good internal reliability ($\alpha = .85$ for benefit items and $\alpha = .80$ for burden items) in a pediatric cancer sample (Currier et al., 2009). This measure can be readily adapted for various pediatric chronic illness populations and the benefit subscale was used in the current study (Appendix G). In the current sample, Cronbach's alpha was .91 indicating good reliability for the benefit subscale.

2.2.4 Caregiver and Family Protective Factors

Parent Psychological Flexibility. The Parent Psychological Flexibility Questionnaire (PPFQ; McCracken & Gauntlett-Gilbert, 2011; Wallace, McCracken, Weiss, & Harbeck-Weber, 2015) is a 17-item measure assessing parents' capacity to accept their distress pertaining to their adolescent's pain, maintain present-moment awareness, and pursue values-based goals (e.g., "Despite my child's pain, we are able to pursue activities that are important to our family"). The PPFQ is comprised of four subscales including Values-based Action, Pain Acceptance, Emotional Acceptance, and Pain Willingness. Caregivers responded to each item using a 7-point scale ranging from 0 (*never true*) to 6 (*always true*) where higher scores are indicative of greater psychological flexibility. PPFQ responses were significantly correlated with adolescent pain acceptance, functional disability, and depression among parents of adolescents attending a pediatric chronic pain clinic appointment. This measure demonstrated good internal consistency among mothers ($\alpha = .87$) and fathers ($\alpha = .88$) (Wallace et al., 2015) (Appendix H). Internal consistency in the current sample was good with a Cronbach's alpha of .85.

Family Functioning. Caregivers completed the Family Assessment Device (FAD; Epstein, Baldwin, & Bishop, 1983) that assesses family functioning based on the McMaster Model of Family Functioning. The FAD is a 60-item measure that examines 7 dimensions of family functioning: Problem Solving (e.g., "We try to think of different ways to solve problems"), Communication (e.g., "When we don't like what someone has done, we tell them"), Roles (e.g., "We make sure members meet their family responsibilities"), Affective Responses (e.g., "We cry openly"), Affective Involvement (e.g., "We get involved with each other only when something interests us"), Behavioral Control (e.g., "There are rules about dangerous situations"), and General Functioning (e.g., "We confide in each other"). Items are rated on a 4-point Likert scale,

with lower scores indicating better family functioning. The FAD has been used in several studies examining family functioning in pediatric SCD (Barakat, Lutz, Nicolaou, & Lash, 2005; Mitchell et al., 2007) and has been deemed as a “well-established” family measure in pediatric psychology (Alderfer et al., 2008) (Appendix I). In this study, the General Functioning subscale was used, which consists of 12 items and has demonstrated good psychometric properties in pediatric SCD samples (e.g., Alderfer et al., 2008). General Functioning scores range from 1 to 4 as they are averaged across the 12 items, with lower scores indicative of better general family functioning. For the General Functioning subscale, internal consistency in the current sample was good with a Cronbach’s alpha of .83.

2.2.5 Functioning Outcomes

Functional Ability. The Functional Disability Inventory (FDI; Walker & Greene, 1991) is a 15-item self-report measure that assesses children’s perceived physical ability and psychosocial functioning in the context of their physical health (e.g., “Doing chores at home”). Adolescents were asked to rate their perceptions of ability to engage in various activities during the past 2 weeks on a 5-point scale ranging from 0 (*no trouble*) to 4 (*impossible*). The total score ranges from 0-60 with lower scores indicating greater physical ability. The FDI has demonstrated reliability and validity in children and adolescents (Palermo et al., 2008) and has been shown to be internally consistent with a population of adolescents with chronic pain and their parents ($\alpha = .85-.93$) (Cohen, Vowles, & Eccleston, 2010) (Appendix J). In the current study, Cronbach’s alpha was .94 indicating good reliability.

Quality of Life. Adolescents completed the Pediatric Quality of Life Inventory (PedsQL 4.0 Generic Core Scale; Varni, Seid, & Kurtin, 2001), which has four subscales assessing physical, emotional, social, and school functioning that have been validated with children and

adolescents between the ages of 5 and 18 (e.g., “I cannot do things that other teens my age can do”). Respondents report the extent to which each item has been problematic over the past month using a Likert 5-point scale (0 = never a problem, 1 = almost never a problem, 2 = sometimes a problem, 3 = often a problem, 4 = almost always a problem). Items are reversed scored and transformed into standard scores ranging from 0 to 100, with higher scores indicative of better health-related quality of life. Subscale scores are computed as the sum of the items divided by the number of items answered to account for missing data. However, subscale scores are not computed if more than 50% of the items in the scale are missing. The Total Scale Score is computed as the sum of the items divided by the number of items answered. The PedsQL has been shown to be reliable and valid in youth with SCD (McClellan, Schartz, Sanchez, & Roberts, 2008; Panepinto, Pajewski, Foerster, & Hoffmann, 2008) and responsive to change following recovery from acute pain episodes (Brandow, Brousseau, Pajewski, & Panepinto, 2010) (Appendix K). For this study, the Total Scale Score was used in analyses. Internal consistency in the current sample was good with a Cronbach’s alpha of .93.

2.3 Procedures

As this research involved no more than minimal risk of harm and did not necessitate a procedure for which written consent is typically required outside of a research context, adolescents and caregivers provided verbal consent to participate in this research study. Families who participated in a previous SCD study and provided permission to be contacted about future research were initially contacted via mail. These families received a letter mailed to their home address inquiring about their interest participating in this study. After two weeks, families were subsequently contacted via phone if they had not contacted the research team regarding their interest in participating. A trained research assistant read a telephone script over the phone to

receive verbal consent from families who were interested in participating in this study. These participants were then provided the option to receive a link via email to complete questionnaires electronically via REDCap (Research Electronic Data Capture) or receive paper-and-pencil questionnaires in the mail with pre-paid postage to return completed to the mailing address provided.

Families were also recruited from outpatient clinics at an urban children's hospital in the southeastern United States identified as having the largest volume of SCD patients per week. The student PI or a trained research assistant approached eligible families after they completed the check-in process in the waiting area. The student PI or trained research assistant explained the nature of the current study in greater detail and verbally obtained caregiver and adolescent consent if the family expressed interest in participating. Given the previously identified low response rate with allowing families to complete questionnaires online using REDCap, families were provided paper-and-pencil questionnaires and encouraged to complete these measures by the end of their clinic visit. Given that the average length of a clinic visit ranged from 60-90 minutes in duration, all families were able to complete these questionnaires and return them at the end of their clinic visit.

Caregivers completed the Background Information Form and filled out measures evaluating their psychological flexibility in the context of their adolescent's pain and family functioning. Adolescents answered questions about their SCD pain burden, pain frequency and intensity, mindfulness, general and pain-specific expectancies, benefit finding, well-being, and functional ability. Study measures took approximately 30-45 minutes to complete. Families received \$5 Target gift cards for study participation and questionnaire completion. Questionnaire data were entered into REDCap, a secure web application for building and managing online

research surveys and databases. Information regarding adolescents' SCD, medical history, and additional medical or psychological diagnoses were confirmed through chart review. All procedures were reviewed and approved by Children's Healthcare of Atlanta and Georgia State University institutional review boards.

3 DATA ANALYSES

3.1 Preliminary Analyses

Data from REDCap were imported into Excel and subsequently transferred into SPSS (Version 22) to conduct preliminary and primary analyses.

Descriptive statistics, including means, standard deviations, ranges, and frequencies were performed to characterize the sample and primary variables. Data were tested for normality and statistical assumptions were inspected, including regression diagnostics to confirm that all regression assumptions were met (Field, 2009). Correlations, *t* tests, and one-way analyses of variance (ANOVAs) were conducted to identify potential covariates based on associations between demographic, disease factors, and outcome variables, and examine associations among study variables. Finally, one-way ANOVAs were performed to ensure that no differences in demographic or outcome variables emerged based on method of data completion (i.e., paper-based versus online).

3.2 Primary Analyses

The following covariates were controlled for in analyses given their relation with outcome variables: adolescent age, SCD genotype, pain composite, and average pain frequency. Correlation analyses were conducted to determine associations among pain variables, adolescent protective factors, caregiver and family protective factors, and functioning outcomes (Primary Aim 1). To investigate whether each adolescent (mindfulness, optimism, pain acceptance, benefit

finding), caregiver (psychological flexibility), and family (family functioning) protective factor moderated the relation between adolescent SCD pain burden and functioning outcomes (functional ability, quality of life), the SPSS PROCESS macro (Hayes, 2013; model 1) was used (Primary Aim 2). The PROCESS macro uses bootstrapping, a nonparametric resampling technique (5,000 samples) to assess effects (Preacher & Hayes, 2004). A bootstrapping approach is recommended for smaller sample sizes that may not be normally distributed, rectifying the impact of asymmetrical sampling distributions on statistical power (Preacher & Hayes, 2004, 2008). As recommended when testing and interpreting interactions, all predictor variables were mean centered to minimize multicollinearity (Aiken, West, & Reno, 1991). Effects were considered significantly different from zero at $p < .05$ when zero did not fall within the 95% bias-corrected confidence interval (Preacher & Hayes, 2004). A total of 12 separate moderation analyses were conducted. To graph statistically significant interactions, standard output from the PROCESS macro was used for conditional effects of the predictor at low (one standard deviation below the mean), average (sample mean), and high (one standard deviation above the mean) values of the moderator.

4 RESULTS

4.1 Preliminary and Descriptive Analyses

Initially, the participant sample was characterized by running descriptive statistics, including means, standard deviations, and frequencies (Table 1). Next, means, standard deviations, and ranges of primary study variables were obtained (Table 2). Pearson's correlations (Table 3) revealed that the pain composite (average of typical and worst pain intensity) positively correlated with days of pain reported over the past month, SCD pain burden, and functional disability; and the pain composite negatively correlated with quality of life. Adolescent age was

positively associated with days of pain reported over the past month and functional disability, and negatively associated with optimism. An independent-samples *t* test revealed no differences in outcome variables (functional disability and quality of life) between adolescent males and females. A one-way ANOVA revealed that there were no statistically significant differences between adolescent SCD genotypes for quality of life [$F(3,77) = 1.797, p = .16$], however there were statistically significant differences for functional disability [$F(3,82) = 5.146, p = .003$]. Specifically, adolescents with the SC genotype had greater functional disability compared to those with the SS ($p = .007$) and SB (0) thalassemia ($p = .024$) genotypes. Interestingly, there were also no statistically significant differences for functional disability and quality of life based on family income level. Finally, study variables did not differ based on participant method of data completion (online versus paper questionnaires). Based on these analyses, adolescent age, SCD pain genotype, pain composite, and average pain frequency were entered as covariates in primary analyses involving regression and moderation.

Regression diagnostics indicated that all regression assumptions were met to perform moderation analyses. Assumptions of normality, linearity, and homoscedasticity were confirmed by running scatter plots of residuals. Variance inflation factors (VIFs) ranged from 1 to 1.1. Prior literature has advised that VIF values approaching or exceeding 10 indicate severe multicollinearity requiring correction (Hair, Anderson, Tatham, & Black, 1995).

4.2 Primary Analyses

4.2.1 Primary Aim 1

The first primary goal of this study was to explore associations among pain variables, adolescent protective factors, caregiver and family protective factors, and functioning outcomes. To examine this aim, correlation analyses were performed. The pain composite and number of

pain days over the past month were positively correlated with one another ($r = .61, p < .001$).

Pain composite was positively correlated with SCD pain burden ($r = .74, p < .001$) and functional disability ($r = .59, p < .001$), and negatively correlated with adolescent mindfulness ($r = -.36, p = .001$), pain acceptance ($r = -.24, p = .024$), and quality of life ($r = -.59, p < .001$).

Similarly, number of pain days over the past month was positively correlated with SCD pain burden ($r = .67, p < .001$) and functional disability ($r = .60, p < .001$), and negatively correlated with adolescent mindfulness ($r = -.23, p = .046$), pain acceptance ($r = -.29, p = .013$) and quality of life ($r = -.50, p < .001$). Number of pain days over the past month was also negatively correlated with adolescent optimism ($r = -.23, p = .045$). In addition to the pain variables, SCD pain burden was positively associated with functional disability ($r = .64, p < .001$) and poorer family functioning ($r = .28, p = .014$), and negatively associated with mindfulness ($r = -.33, p = .002$), pain acceptance ($r = -.24, p = .023$), optimism ($r = -.24, p = .028$), and quality of life ($r = -.64, p < .001$).

With regard to protective factors, adolescent mindfulness was positively correlated with optimism ($r = .48, p < .001$), quality of life ($r = .58, p < .001$), and caregiver psychological flexibility ($r = .26, p = .015$), and negatively correlated with functional disability ($r = -.39, p < .001$). Adolescent pain acceptance was only positively associated with caregiver psychological flexibility ($r = .27, p = .012$) and negatively associated with functional disability ($r = -.24, p = .028$). In addition to mindfulness, optimism was positively correlated with benefit finding ($r = .33, p = .003$) and quality of life ($r = .55, p < .001$), and negatively correlated with functional disability ($r = -.40, p < .001$). Benefit finding was also positively related to quality of life ($r = .24, p = .035$) and negatively correlated with poorer family functioning ($r = -.33, p = .003$). Finally, poorer family functioning was also negatively associated with quality of life ($r = -.25, p$

= .028).

4.2.2. Primary Aim 2

The second primary goal of this study was to investigate whether adolescent, caregiver, and family protective factors moderated the relation between adolescent SCD pain burden and functioning outcomes. Using the PROCESS macro, predictors and interaction terms were centered and adolescent age, SCD genotype, pain composite scores, and average pain frequency were entered as covariates in analyses.

4.2.1.1 Adolescent Protective Factors as Moderators

When examining mindfulness as a moderator of the relation between pain burden and quality of life, the overall model was significant, $F(6, 72) = 20.12, p < .001$, and accounted for 59% of the variance in quality of life. Pain burden, $\beta = -1.19, t(72) = -2.80, p = .007$, and mindfulness, $\beta = .81, t(72) = 4.59, p < .001$, were both significantly related to quality of life, however the interaction term was not significant, $\beta = -.01, t(72) = -.28, p = .78$.

When considering pain acceptance as a moderator, the overall model was significant, $F(6, 72) = 14.24, p < .001$, and accounted for 52% of the variance in quality of life. While pain burden predicted quality of life, $\beta = -.96, t(72) = -2.32, p = .02$, pain acceptance was not a predictor of quality of life, $\beta = .16, t(72) = .96, p = .34$. However, the interaction term was significant, $\beta = .06, t(72) = 2.42, p = .02$. More specifically, there is a significant relation between pain burden and quality of life at low levels of pain acceptance, $\beta = -1.75, t(72) = -3.98, p < .001$, and average levels of pain acceptance, $\beta = -.96, t(72) = -2.32, p = .02$, but not at high levels of pain acceptance, $\beta = -.18, t(72) = -.29, p = .77$ (Figure 4). When probing the moderation effect further to examine zones of significance using the Johnson-Neyman Technique, pain burden and quality of life were negatively related when pain acceptance scores

were at or below 44.59, $\beta = -.86$, $t(72) = -1.99$, $p = .05$. As pain acceptance scores decreased further, the relation between pain burden and quality of life became more negative (e.g., lowest pain acceptance score of 19, $\beta = -2.50$, $t(72) = -3.91$, $p < .001$). In other words, higher pain acceptance scores buffer the impact of pain burden on quality of life by weakening the negative relation between these variables.

When entering optimism as a moderator of the relation between pain burden and quality of life, the overall model was significant, $F(6, 71) = 16.76$, $p < .001$, and accounted for 62% of the variance in quality of life. Pain burden, $\beta = -1.04$, $t(71) = -2.65$, $p = .01$, and optimism, $\beta = 2.05$, $t(71) = 4.91$, $p < .001$, both individually predicted quality of life, however the interaction term was not significant, $\beta = -.03$, $t(71) = -.36$, $p = .72$. When considering benefit finding as a moderator of the relation between pain burden and quality of life, the overall model was significant, $F(6, 70) = 10.40$, $p < .001$, and accounted for 50% of the variance in quality of life. Pain burden predicted quality of life, $\beta = -1.16$, $t(70) = -2.64$, $p = .01$, however neither benefit finding, $\beta = .31$, $t(70) = 1.86$, $p = .07$, nor the interaction term, $\beta = -.0001$, $t(70) = -.0031$, $p = .998$, were significant. When examining caregiver psychological flexibility as a moderator of the relation between pain burden and quality of life, the overall model was significant, $F(6, 71) = 10.81$, $p < .001$, and accounted for 48% of the variance in quality of life. Pain burden predicted quality of life, $\beta = -1.20$, $t(71) = -2.80$, $p = .007$, however caregiver psychological flexibility, $\beta = .13$, $t(71) = 1.25$, $p = .21$, and the interaction term, $\beta = -.0015$, $t(71) = -.08$, $p = .94$, were not significant predictors. When entering family functioning as a moderator of the relation between pain burden and quality of life, the overall model was significant, $F(6, 66) = 11.05$, $p < .001$, and accounted for 51% of the variance in quality of life. Pain burden predicted quality of life, $\beta = -1.09$, $t(66) = -2.50$, $p = .02$, however family functioning, $\beta = -2.93$, $t(66) = -.69$, $p = .49$, and

the interaction term, $\beta = -1.10$, $t(66) = -1.44$, $p = .15$, did not predict quality of life.

When exploring mindfulness as a moderator of the relation between pain burden and functional disability, the overall model was significant, $F(6, 78) = 10.56$, $p < .001$, and accounted for 56% of the variance in functional disability. Pain burden predicted functional disability, $\beta = .70$, $t(78) = 2.86$, $p = .006$, however neither mindfulness, $\beta = -.21$, $t(78) = -1.52$, $p = .13$, nor the interaction term, $\beta = -.03$, $t(78) = -.89$, $p = .38$, were significant predictors of functional disability. When selecting pain acceptance as a moderator of the relation between pain burden and functional disability, the overall model was significant, $F(6, 78) = 9.77$, $p < .001$, and accounted for 57% of the variance in functional disability. Pain burden predicted functional disability, $\beta = .68$, $t(78) = 2.87$, $p = .005$, however pain acceptance, $\beta = -.12$, $t(78) = -1.31$, $p = .19$, and the interaction term, $\beta = -.03$, $t(78) = -1.25$, $p = .21$, were not predictors of functional disability. The overall model was significant when optimism was entered as a moderator of the relation between pain burden and functional disability, $F(6, 77) = 10.78$, $p < .001$, and accounted for 58% of the variance in functional disability. Both pain burden, $\beta = .71$, $t(77) = 2.70$, $p = .009$, and optimism, $\beta = -.58$, $t(77) = -2.49$, $p = .01$, predicted functional disability, however the interaction term remained non-significant, $\beta = -.03$, $t(77) = -.85$, $p = .40$. When examining benefit finding as the moderator of the relation between pain burden and functional disability, the overall model was significant, $F(6, 75) = 8.87$, $p < .001$, and accounted for 57% of the variance in functional disability. Pain burden was a significant predictor of functional disability, $\beta = .73$, $t(75) = 2.92$, $p = .005$. Neither benefit finding, $\beta = -.18$, $t(75) = -1.64$, $p = .11$, nor the interaction term, $\beta = -.02$, $t(75) = -.69$, $p = .49$, predicted functional disability.

4.2.1.2 Caregiver and Family Protective Factors as Moderators

With regard to caregiver and family protective factors, when caregiver flexibility was

entered as a moderator of the relation between pain burden and functional disability, the overall model was significant, $F(6, 77) = 8.78, p < .001$, and accounted for 54% of the variance in functional disability. Pain burden predicted functional disability, $\beta = .84, t(77) = 2.92, p = .005$, however caregiver flexibility, $\beta = .006, t(77) = .09, p = .93$, and the interaction term, $\beta = .008, t(77) = .57, p = .57$, were not significant predictors. Finally, when family functioning served as a moderator of the relation between pain burden and functional disability, the overall model was significant, $F(6, 70) = 7.87, p < .001$, and accounted for 55% of the variance in functional disability. While pain burden was a significant predictor of functional disability, $\beta = .70, t(70) = 2.78, p = .007$, neither family functioning, $\beta = 1.90, t(70) = .65, p = .52$, nor the interaction term, $\beta = .31, t(70) = .48, p = .63$, were significant predictors. In summary, only adolescent pain acceptance moderated the relation between pain burden and quality of life, supporting the protective factor model of resilience.

Table 1: Participant Demographic Information (N = 93)

	<i>M (SD)</i>
Age	15.23 (1.97)
	<i>N (%)</i>
Gender	
Male	44 (47.3)
Female	49 (52.7)
Ethnicity	
Hispanic/Latino	0 (0)
Not Hispanic/Latino	93 (100)
Race	
Black or African American	92 (98.9)
Multi-racial	1 (1.1)
SCD genotype	
SS	64 (68.8)
SB (0) thalassemia	8 (8.6)
SB (+) thalassemia	4 (4.3)
SC	17 (18.3)
Psychological disorder	
Diagnosis	12 (12.9)
Anxiety	5 (5.4)
Depression	2 (2.2)
Attention-deficit/hyperactivity disorder	2 (2.2)
Multiple diagnoses	2 (2.2)
None	77 (82.8)
Missing	4 (4.3)
Caregiver relation to child	80 (86)
Mother	11 (11.8)
Father	1 (1.1)
Step-father	1 (1.1)
Missing	
Approximate annual family income	
Up to \$10,000	5 (5.4)
\$10,001-\$20,000	14 (15.1)
\$30,001-\$40,000	15 (16.1)
\$40,001-\$50,000	9 (9.7)
\$50,001-\$60,000	3 (3.2)
\$60,001-\$70,000	6 (6.5)
\$70,001-\$80,000	4 (4.3)
\$80,001-\$90,000	9 (9.7)
\$90,000 and above	16 (17.2)
Missing	12 (12.9)

Table 2: Descriptives of Pain and Outcome Study Variables

Variables (Measures)	<i>M (SD)</i>	Minimum	Maximum
Pain composite	4.62 (3.07)	0	10
Number of pain days over the past month	7.41 (8.86)	0	30
SCD Pain Burden (SCPBI-Y) ^a	6.90 (6.20)	0	22
Mindfulness (CAMM) ^b	28.50 (8.38)	1	40
Pain Acceptance (CPAQ-A) ^c	42.97 (11.97)	19	78
Optimism (YLOT) ^d	13.64 (3.87)	2	18
The Benefit Finding and Burden Scale for Children (BFBSC) Benefit Finding subscale ^e	33.39 (10.61)	10	50
Caregiver Psychological Flexibility (PPFQ) ^f	40.38 (16.76)	0	83
Family Functioning (FAD GF subscale) ^g	1.63 (.37)	1	2.58
Functional disability (FDI) ^h	10.79 (12.10)	0	47
Quality of life (PedsQL) ⁱ	72.67 (18.43)	25	100

Note. ^a SCPBI-Y scores range from 0 to 28, with higher scores indicative of more pain burden. ^b CAMM scores range from 0 to 40, with higher scores indicative of greater mindfulness. ^c CPAQ-A scores range from 0-80, with higher scores indicative of more pain acceptance. ^d YLOT optimism subscale scores range from 0 to 18 [healthy sample mean (standard deviation) = 14.40 (3.59)], with higher scores indicative of greater optimism. ^e BFBSC Benefit Finding subscale scores range from 0 to 50, with higher scores indicative of greater benefit finding. ^f PPFQ scores range from 0 to 102, with higher scores indicative of more caregiver psychological flexibility in the context of adolescent pain. ^g FAD GF subscale scores range from 1 to 4, with lower scores indicative of better family functioning. ^h FDI scores range from 0 to 60, with higher scores indicative of worse daily functioning. ⁱ PedsQL total scores range from 0 to 100, with higher scores indicative of greater overall quality of life.

Table 3: Intercorrelations Among Study Variables

Variable	1	2	3	4	5	6	7	8	9	10
1. Pain composite	-									
2. Days pain in last month	.61**	-								
3. SCD Pain Burden	.74**	.67**	-							
4. Mindfulness	-.36*	-.23*	-.33*	-						
5. Pain Acceptance	-.24*	-.29*	-.24*	.11	-					
6. Optimism	-.17	-.23*	-.24*	.48**	.18	-				
7. Benefit Finding	.06	-.02	-.11	.10	.06	.33*	-			
8. Functional Disability	.59**	.60**	.64**	-.39**	-.24*	-.40**	-.16	-		
9. Quality of Life	-.59**	-.50**	-.64**	.58**	.20	.55**	.24*	-.72**	-	
10. Caregiver Flexibility	-.17	.02	-.14	.26*	.27*	.07	-.07	-.05	.15	-
11. Family Functioning	.08	.03	.28*	-.17	-.17	-.21	-.33*	.22	-.25*	-.04

Note. * $p < .05$. ** $p < .001$.

Variables negatively correlated with family functioning are associated with better family functioning.

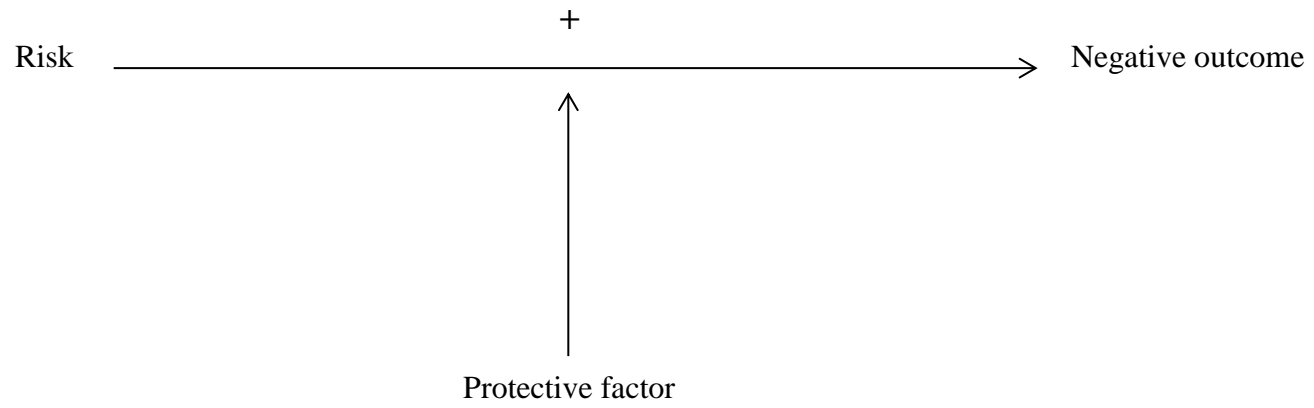


Figure 1. Protective Factor Resilience Model

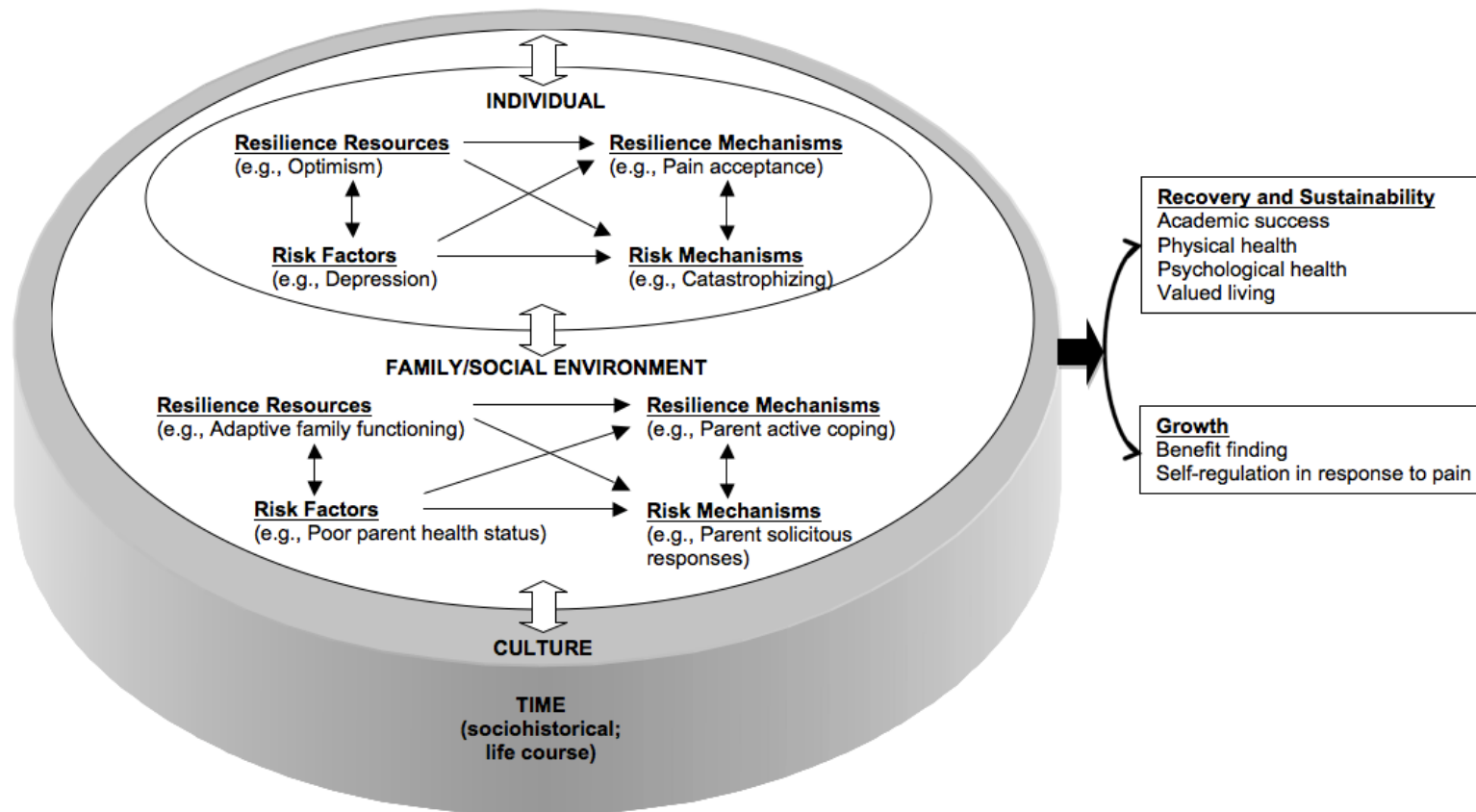


Figure 2. Ecological Resilience-Risk Model in Pediatric Pain

Note. Within this model, resilience resources promote adaptive outcomes by enhancing resilience mechanisms and minimizing risk factors and mechanisms. Conversely, risk factors interfere with resilience and contribute to poor pain adaptation by enhancing risk mechanisms and minimizing resilience factors and mechanisms. These risk and resilience pathways occur within the context of the individual, the family/social environment, culture, and time, which mutually interact with one another.

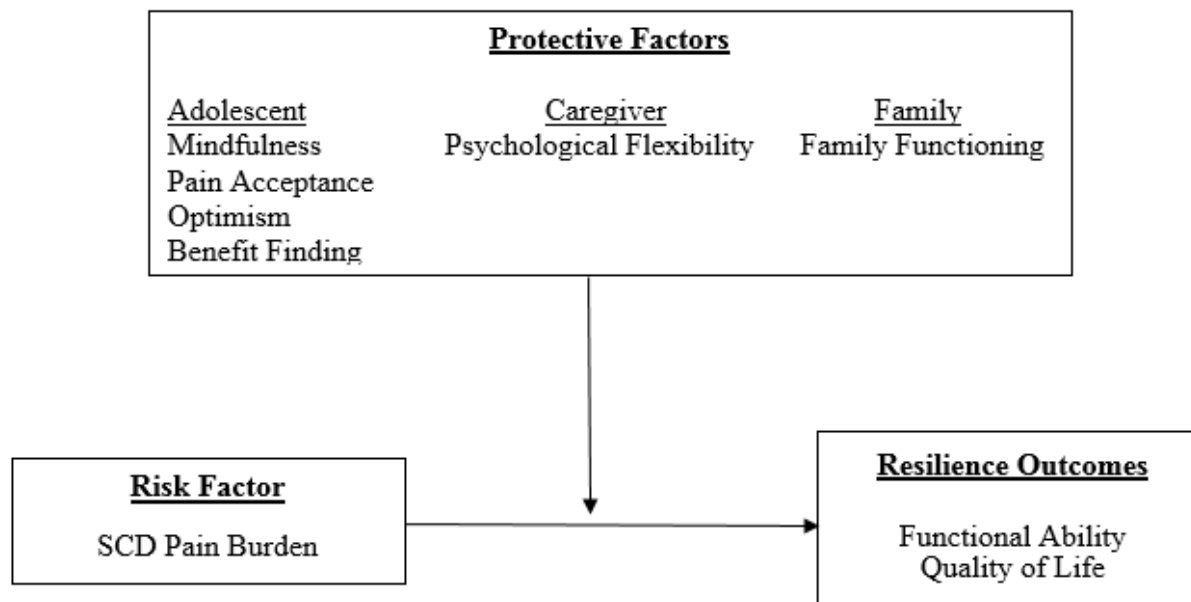


Figure 3. Resilience Pathways Tested in Current Study

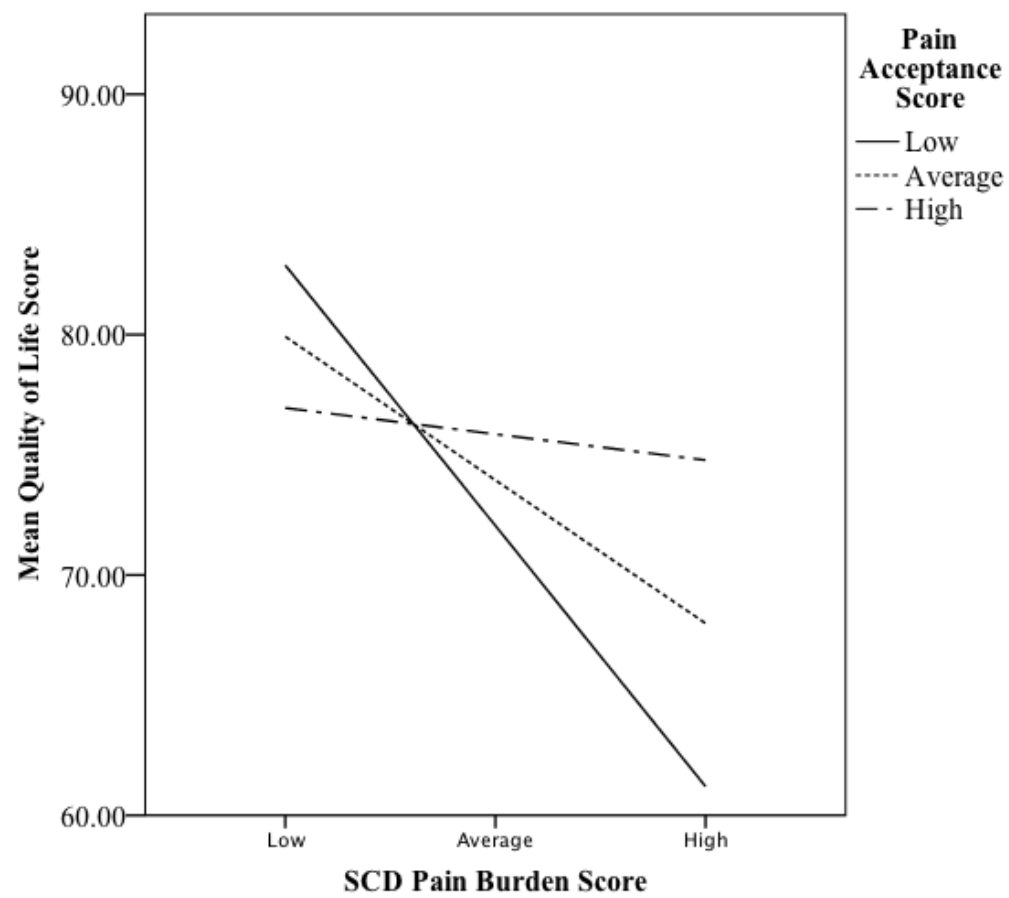


Figure 4. The Moderating Effect of Adolescent Pain Acceptance on the Relation Between Pain Burden and Quality of Life

5 DISCUSSION

5.1 Overview

Pain in adolescents with SCD remains an understudied research area, which is especially problematic given the high stigmatization and range of medical needs in this population. Prior literature has utilized risk and resistance models to identify and examine relevant factors in pediatric SCD (Brown et al., 1993; 2000); however, no studies to date have applied resilience theory from developmental psychology to assess the role of protective factors in pediatric SCD pain and disease management. Given the adversity of living with a chronic medical condition with high pain, it is particularly important to determine how to promote positive adjustment and adaptation to a life-long illness and alleviate disease burden. As pain relief is rarely achieved through pharmacological treatments alone, it is critical to bolster strengths that foster positive coping skills and effective pain and disease self-management. Furthermore, applying resilience theory and examining mechanisms of resilience in pediatric psychology populations remains a relatively unexplored yet potentially important and promising area of study (Hilliard et al., 2015).

This study had several aims. First, I explored associations among pain characteristics and adolescent, caregiver, and family protective factors previously identified in the ecological resilience-risk model for pediatric pain (Cousins et al., 2015). Relatedly, I examined pain characteristics and functioning outcomes (functional ability, quality of life). Second, I applied the protective-reactive model of resilience to investigate moderating effects of adolescent, caregiver, and family protective factors on the relation between pain burden and functioning outcomes (functional ability, quality of life).

5.2 Preliminary Analyses

With regard to the sample, the number of male and female adolescents was relatively equivalent. Consistent with prior SCD research, the most prevalent SCD genotypes were HbSS and HbSC (Sil et al., 2016). In terms of family income, there were more families recruited with higher annual incomes when compared to those with incomes below the national average and previous studies (e.g., Sil et al., 2016); however, a large percentage of caregivers did not disclose their annual income. When conducting further analyses, family income was found to be associated with some of the protective factors examined in this study including adolescent benefit finding, caregiver psychological flexibility, and family functioning. Thus, the higher socioeconomic status among families in the current study may also reflect more optimal functioning in the context of SCD-related pain. Additionally, participants endorsed a moderate level of pain intensity according to their pain composite scores, which is consistent with prior literature (Dampier et al., 2002; Gil et al., 2000; Sil et al., 2016); however, their average number of days experiencing pain within the past month was lower than what has been found in prior studies (Soumitri et al., 2016).

When considering primary variables of interest, all measures included in the current study demonstrated good psychometrics. SCD pain burden scores were consistent with a previous pediatric SCD outpatient sample (Zempsky et al., 2013). Adolescent mindfulness was higher in this sample compared to community samples of youth (Greco et al., 2011) and adolescents (Petter et al., 2013). This is particularly interesting given that Greco et al. (2011) found that African American youth had lower mindfulness scores relative to Non-Hispanic White and multiracial youth; however, these youth represented a small percentage of the total sample. Adolescent optimism in the current study was only slightly lower than in a sample of

healthy children and higher than in a sample of youth with chronic pain (Cousins et al., 2015). With regard to adolescent pain acceptance, participants in the current sample had higher total scores relative to adolescents from an outpatient pediatric pain clinic (Wallace et al., 2011) and a severely disabled group of adolescents with chronic pain who participated in a 3-week residential ACT treatment program (Gauntlett-Gilbert et al., 2013). Notably, pain acceptance scores in the current study more closely approximated those reported by the severely disabled adolescents post-treatment (Gauntlett-Gilbert et al., 2013). Benefit finding was slightly lower relative to the pediatric cancer population (Currier et al., 2009; Phipps et al., 2007), but more closely approximated scores endorsed by childhood cancer survivors (Michel et al., 2010). Of note, African American children diagnosed with cancer demonstrate higher levels of benefit finding compared to Non-Hispanic White children diagnosed with cancer and the adolescent participants in the current study (Phipps et al., 2007). Based on these measures, the current sample demonstrated a similar degree of SCD-related pain burden compared to other samples of youth with SCD and equivalent benefit finding relative to childhood cancer survivors, but interestingly endorsed higher mindfulness, optimism, and acceptance compared to community samples of adolescents and youth with non-SCD chronic pain.

Caregiver psychological flexibility in the context of adolescent pain was substantially lower compared to previous research with primarily Non-Hispanic White caregivers of youth with chronic pain (Wallace, Woodford, & Connelly, 2016) and Swedish parents of youth with chronic pain (Wiwe Lipsker et al., 2016). The general family functioning subscale of the FAD closely approximated previously reported scores among caregivers of children with SCD (Schlenz et al., 2016). Adolescents in this study reported disability scores that fell in the minimal disability range, which suggests better functioning than prior samples of youth with SCD (Sil et

al., 2016) or youth with chronic pain (Kashikar-Zuck et al., 2011). Adolescents also reported slightly higher quality of life compared to prior studies with youth with SCD (Panepinto et al., 2008; Zempky et al., 2013). Thus, although parents are reporting low flexibility, the adolescents in this sample are reporting relatively high functioning.

5.3 Primary Aim 1

Consistent with prior literature, adolescent age, SCD genotype, and pain composite scores were associated with functional ability (Dampier et al., 2016; Sil et al., 2016). In line with hypotheses, correlation analyses supported associations among variables in the expected directions and consistent with the ecological resilience-risk model (Cousins et al., 2015). Pain characteristics were associated with greater SCD pain burden and disability, and negatively related to protective factors and quality of life.

Exploring associations among protective factors provided support for their influence on one another. For example, adolescent mindfulness was positively associated with adolescent optimism. Previous literature has supported this finding among university students and adults (e.g., Brown & Ryan, 2003). Consistent with this relation, in a 10-week school-based randomized-controlled trial with pre- and early-adolescents, Schonert-Reichl and Lawlor (2010) found that mindfulness training resulted in increases in optimism. Although mindfulness and optimism have rarely been examined simultaneously in pediatric pain research, findings provide initial support that these constructs are important to continue investigating among youth with pain. In this study, it is possible that adolescents with higher daily mindfulness were more likely to be optimistic as mindfulness fosters enhanced psychological flexibility and a better ability to separate from negative emotions and thoughts. In contrast, it might be that adolescents with an

optimistic outlook might engage in more mindfulness. Given the cross-sectional nature of the study, there might be other explanations for the correlation between these constructs.

Caregiver psychological flexibility correlated with both adolescent mindfulness and adolescent pain acceptance. These relations support the notion that core processes pertinent to psychological flexibility are modeled by caregivers (Wallace et al., 2015), and these findings are consistent with the application of the psychological flexibility model to pediatric chronic pain (McCracken & Morley, 2014). Adolescent optimism was positively correlated with benefit finding, which mirrors prior findings with a pediatric oncology sample (Phipps et al., 2007). It is understandable that adolescents who exhibit more positive expectancies would have a greater ability to identify positive aspects of living with a chronic illness. In youth with SCD, this is particularly important to highlight given the unique health-related stigma and discrimination this population faces (Wesley, Zhao, Carroll, & Porter, 2016). Finally, adolescent benefit finding was associated with more adaptive general family functioning. This is the first study to examine these constructs together so it is unclear exactly how these protective factors are related; however, one hypothesis is that adolescents who are in a home environment that promotes effective communication and problem-solving may have had more opportunities to learn how to view situations adaptively. Another hypothesis is that these adolescents also perceive their families as more supportive, which makes them more likely to focus on ways having SCD has unified the family.

Although most protective factors were positively related to one another, some were not. Adolescent benefit finding was not associated with adolescent mindfulness, adolescent pain acceptance, or caregiver psychological flexibility. Family functioning remained unrelated to adolescent mindfulness, adolescent pain acceptance, adolescent optimism, and caregiver

psychological flexibility. Surprisingly, adolescent mindfulness and pain acceptance were also not related. These findings suggest that some aspects of the psychological flexibility model may not translate precisely to pediatric sickle cell populations. It is also important to consider that the mindfulness measure assessed general non-pain specific observations of daily mindfulness whereas the acceptance and psychological flexibility measures were developed in the context of pain. Furthermore, it is likely that benefit finding and family functioning were not correlated with many of the pain-specific protective factors examined in this study, as they are more global non pain-specific measures.

With regard to protective factors and functioning outcomes, consistent with a previous study using a mixed pediatric chronic pain sample (Cousins et al., 2015), optimism was related to both functioning outcomes. Similarly, mindfulness was related to both functioning outcomes, however pain acceptance was only related to the FDI, and benefit finding and general family functioning were only related to the PedsQL. A previous randomized controlled trial demonstrated that an ACT-oriented intervention based on exposure and acceptance strategies for youth with chronic pain showed higher quality of life in children and adolescents post-treatment in addition to perceived functional ability compared to a multidisciplinary treatment approach with amitriptyline (Wicksell et al., 2009). Notably, this study assessed quality of life with a different measure compared to the current study, which may explain discrepant findings. Wicksell et al. (2009) utilized the SF-36 to measure quality of life, which exclusively contains physical and mental subscales, whereas the PedsQL captures physical, emotional, social, and school functioning domains. Benefit finding and family functioning may have only been related to quality of life as they are not pain-specific measures and the PedsQL targets broader domains of functioning associated with well-being. The only protective factor that remained unrelated to

both the FDI and PedsQL was caregiver psychological flexibility. This contrasts with previous studies that have found significant associations between caregiver psychological flexibility and adolescent functioning measured by the FDI and the Bath Adolescent Pain Questionnaire (McCracken & Gauntlett-Gilbert, 2011; Wallace et al., 2015). The null finding in the current study might suggest that caregiver psychological flexibility does not influence physical functioning or well-being among adolescents with SCD. This construct may not be as pertinent to caregivers of adolescents with SCD given the nature of SCD-related pain (acute, chronic, vaso-occlusive pain crises). For instance, caregivers may benefit from having more structured and circumscribed beliefs about and reactions towards pain as the disease process triggers pain, which can require immediate attention due to adverse health complications. Furthermore, given the genetic component of SCD, it is likely that many caregivers who participated in this study also manage SCD themselves, which may have impacted their responses to questions assessing their attitudes, thoughts, and feelings about their adolescent's sickle-cell related pain.

5.4 Primary Aim 2

Consistent with the protective-reactive resilience model (Luthar, Cicchetti, & Becker, 2000), adolescent pain acceptance buffered the relation between SCD pain burden and quality of life. As expected, higher levels of adolescent pain acceptance mitigated the impact of SCD pain burden on quality of life. This significant moderation is also consistent with the tenets of ACT as increased acceptance reduces activity avoidance and associated distress, which in turn enhances engagement in valued activities and well-being (Dahl et al., 2005). As ACT-oriented approaches have been shown to be particularly helpful for youth with SCD (Masuda et al., 2011) as well as chronic pain (e.g., Wicksell et al., 2015), it is not surprising that pain acceptance serves as a protective factor in the context of pain.

Findings did not support the other adolescent, caregiver, and family protective factors as moderators. It is possible that these constructs may not be applicable moderators in the context of SCD-related pain. Another explanation is that these constructs may be applicable, but the measures were not sufficiently sensitive for youth with SCD. In fact, the majority of the measures were developed for non-SCD chronic pain populations. It could also be that these constructs serve different functions (e.g., mediators). On the other hand, functioning might not have been an appropriate outcome. Specifically, although disability is commonly used in pediatric pain research, it is not considered a “resilience outcome.” Additionally, it is important to highlight that covariates and SCD pain burden accounted for 47.6 to 54.5 percent of the variance in outcomes entered in the regression models, limiting the amount of remaining variance protective factors could account for. A final explanation for the null findings is that the small sample size might have been insufficient to identify significant small effects. In fact, a post-hoc power analysis indicated that a sample of at least 368 participants would have been necessary to identify the small effect (.03) found when optimism was entered as a moderator of the relation between pain burden and quality of life.

5.5 Limitations and Future Directions

Despite the novel contributions this research adds to the study of resilience in pediatric SCD, there are limitations to note and areas for future directions. First and foremost, the field of resilience has been criticized for having diverse and inconsistent terminology, which has complicated interpretation of study findings and implications. For example, it was initially assumed that resilience referred to a global, stable individual trait (Tarter & Vanyukov, 1999); however, researchers refuted this generalization by emphasizing that the construct of resilience is fluid and determined by the socio-ecological context and specific risk, protective, and outcome

factors (Kaplan, 1999). This led to researchers arguing that the state-like term “resilience” should be used rather than the trait-like term “resiliency” (Luthar et al., 2000). Furthermore, researchers have stressed that resilience should be coined as a profile or trajectory descriptor instead of a person-specific descriptor that inadvertently faults individuals who are unable to overcome the adversity of risk (Luthar & Zelazo, 2003). As highlighted by Fergus and Zimmerman (2005), trait-like constructs not only disregard the importance of environmental and contextual factors, but also infer that they are not modifiable or able to be targeted by resilience promotion interventions.

Another criticism of resilience research is that it is challenging to generalize findings when resilience is conceptualized as being specific to a population of individuals, the socio-environmental context, and influential risk and protective factors (Fergus & Zimmerman, 2005). Given this caveat in the resilience literature, it is critical to acknowledge the limited generalizability of the current study findings to other pediatric pain populations that do not embody the unique characteristics of this sample.

The cross-sectional design prevents the ability to determine causal relations or the direction of influence among selected risk, protective, and outcome variables. However, given the infancy of this research area in the context of pediatric SCD-related pain, future studies should continue to develop and identify relevant risk, protective, and outcome variables before investigating these variables in longitudinal or experimental models. It will also be important for future research to utilize more advanced statistical methods (e.g., structural equation modeling) to test the overall model with multiple moderators using one single analysis. Longitudinal studies will capture resilience processes chronologically and be more statistically adept at illustrating how risk and protective factors mutually influence one another at various time points. In

addition, important foundational studies will help support interventions aimed at manipulating resilience factors to improve outcomes.

In terms of study measures, when available, this study did calculate average scores based on adolescent and caregiver report to better assess pain characteristics. Given the limited development of measures targeting caregiver and family protective factors, this study only included a few contextual measures that captured caregiver pain attitudes and the family environment. It will be important for future research to develop more caregiver- and family-focused resilience measures that are applicable to pediatric pain populations. Future studies would also benefit from more extensively measuring key contextual factors (e.g., adolescent-caregiver dynamics, school environment, community support). A specific area of growth regarding measurement is developing assessments of resilience outcomes. One of the most well-established, gold standard outcome measures in pediatric pain is the Functional Disability Inventory (Walker & Greene, 1991), which captures functional disability, but does not serve as an appropriate resilience measure for this population. In other words, low disability does not necessarily indicate high ability or resilience. The FDI was included in the current study to provide comparisons with the extant literature and due to the lack of measures that capture resilience in pediatric pain. Although study measures had acceptable psychometric properties, the majority of assessments were developed and validated in mixed pediatric chronic pain samples or other pediatric chronic illness populations. Thus, it is possible that these instruments did not appropriately capture the characteristics and nature of SCD-related pain. Additionally, some of the specific protective factors that were examined in this study may be less applicable or operate through different mechanisms for the pediatric SCD population. From this standpoint, it

will be important for future research to validate these measures with larger sample sizes to determine their utility.

The sample included adolescents with lower pain frequency and reported levels of pain intensity. It is likely that the youth most disabled by SCD-related pain may not have participated in the study due to lack of interest or poor follow-through. Furthermore, it is also possible that recruitment methods did not target families with poorer access to resources that may exhibit suboptimal adherence to clinic visits given numerous environmental barriers (e.g., limited transportation, high caregiver demands). However, recruitment methods attempted to prevent this by offering various formats of completing questionnaires (mailing paper questionnaires with return postage, providing online survey link, administering questionnaires during clinic visit). To more accurately capture the pediatric SCD-related pain experience, future research should exclusively target specific pain characteristics or enroll a larger sample size to classify subgroups of patients based on pain frequency and duration (e.g., Sil, Cohen, & Dampier, 2016). These larger sample sizes and stringent inclusion criteria can be better achieved by recruiting across multiple pediatric healthcare sites. Despite this, it is important to note that adolescents who did not endorse experiencing pain over the past month still encounter other adversities related to their chronic health condition (e.g., fatigue, dizziness, delayed growth, activity restrictions).

The sample size of the current study also imposed statistical limitations that warrant further discussion. There are challenges in detecting interactions with small samples (e.g., McClelland & Judd, 1993; Whisman & McClelland, 2005). For instance, according to Cohen's power tables and recommendations, 392 participants would be required to achieve adequate power (.80) to detect small effects for variables that demonstrate no measurement error. The estimated sample size fluctuates depending on the reliability of the measures included in the

interaction term. Aiken et al. (1991) suggest that the optimal sample size more than doubles when measure reliabilities drop from 1.0 to .80 and more than triples when reliabilities decrease from 1.0 to .70. Additionally, interaction term variables that are correlated with the outcome variable also require larger sample sizes. In order to maintain adequate power to detect medium interaction effects with measures that have reliabilities of .70, more than 200 participants should be included in a study, and more than 1,000 participants would be needed to detect small interaction effects (Aiken, West, & Reno, 1991). Whisman and McClelland (2005) recommend selecting measures with high reliability, augmenting statistical power, oversampling extreme scores, and including well-established variables to increase the likelihood of detecting moderation effects.

5.6 Summary

Findings generally support the importance of applying resilience theory to the pediatric SCD population. Results suggest that adolescent pain acceptance may be a critical modifiable variable to target in future interventions explicitly designed for SCD-related pain. ACT-based treatment approaches may need to be tailored for this population, as some primary ACT intervention components (i.e., mindfulness) were not found to impact outcomes; however, it is also possible that adolescents and caregivers would benefit from acceptance-based interventions that promote psychological flexibility. Given the unpredictable nature of SCD, treatments such as ACT that encourage adaptability and resilience might be especially beneficial. However, it is important that caregivers and patients appreciate that there are times when SCD-related pain is an important alert for a potential issue that requires medical care. Thus, it is likely that adolescents with SCD-related pain would benefit from different pain management interventions depending on the nature and chronicity of their pain.

Adolescents in this study endorsed higher scores on measures of resilience (i.e., mindfulness, optimism, pain, acceptance) compared to community samples and youth with chronic pain, which is notable given the immense psychosocial ramifications of their disease and compromised health status. It is time that scientists and practitioners recognize that optimal healthcare requires more than focusing on the elimination of problems; to help our patients thrive, we must strive to not only identify and minimize their struggles, but also explore and enhance their strengths.

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APPENDICES

Appendix A. Background Information Form

Questions about your family

1. Your Relation to Child: ___Mother ___Father ___Grandparent If other, describe:

2. Your Gender: ___Male ___Female ___Other (please specify:
_____)
3. Your Age: _____
4. Your Ethnicity: ___Hispanic or Latino ___Not Hispanic or Latino
5. Your Race: ___American Indian or Alaska Native ___Asian ___Black or African
American ___Native Hawaiian or Other Pacific Islander ___White
6. The highest education level you completed (Please write a number. For example, 8 =
completed middle school, 10 = completed sophomore year of high school, 12 = graduated
high school, 13 = completed freshman year of college, 16 = graduated college): _____
7. Did you complete graduate or professional school? ___Yes ___No
8. Please describe your occupation:

9. Your Marital Status: ___Single ___Married/Partnered ___Separated ___Divorced
___Widowed
If other, please describe: _____
10. The highest education level your spouse/partner completed (Please write a number. For
example, 10 = completed sophomore year of high school, 12 = graduated high school, 13 =
completed freshman year of college, 16 = graduated college): _____
11. Did your spouse/partner complete graduate or professional school? ___Yes ___No
12. Please describe your spouse/partner's occupation:

13. Please circle your approximate total family income per year:

a. Up to \$10,000	f. \$50,001 – 60,000
b. \$10,001 – 20,000	g. \$60,001 – 70,000
c. \$20,001 – 30,000	h. \$70,001 – 80,000
d. \$30,001 – 40,000	i. \$80,001 – 90,000

e. \$40,001 – 50,000

j. \$90,000 and above

14. Do you have a chronic medical condition (e.g., asthma, SCD, diabetes, etc.)? YES
NO
If so, what kind(s) _____
15. Does your spouse/partner have a chronic medical condition? YES NO
If so, what kind(s) _____
16. Have you been diagnosed with a psychological disorder (i.e., anxiety, depression, etc.)?
YES NO
If so, what _____
17. Has your spouse/partner been diagnosed with a psychological disorder? YES NO
If so, what _____

*Questions about **your child***

18. Child's Gender: ___ Male ___ Female ___ Other (please specify:
_____)
19. Child's Age: ___ yrs. ___ mos.
20. Child's Ethnicity: ___ Hispanic or Latino ___ Not Hispanic or Latino
21. Child's Race: ___ American Indian or Alaska Native ___ Asian ___ Black or African
American ___ Native Hawaiian or Other Pacific Islander ___ White
22. How many *other children* live in the home? ___ What are their ages? _____
How many children in the home have SCD? _____ How many do not have SCD?

23. How many *other adults* live in the home? _____ What are their ages? _____
24. What type of SCD does your child have? _____
25. Does your child have a chronic illness or medical condition besides SCD (e.g., asthma,
diabetes)?
YES NO If so, what? _____
26. Has your child been diagnosed with a psychological disorder (i.e., anxiety, depression,
etc.)?
YES NO If so, what _____
27. What medication(s) is your child prescribed (please also include medication doses)?

28. Who is responsible for making sure your child takes their medication (i.e., you, child)?

29. When was your child's last SCD related clinic visit? _____
30. When was your child's last SCD related hospitalization? _____
31. How many SCD related pain crises does your child usually experience in one year?

32. How many days has your child had pain in the past month? _____
33. What major complications has your child experienced related to SCD (i.e., strokes, etc.)?

34. How many days of school has your child missed due to SCD symptoms in the past school year? _____
35. How many days of work have you missed due to your child's SCD symptoms in the past year? _____
36. Would you be willing to allow us to keep you and your child's contact information for follow-up or future research projects? YES NO

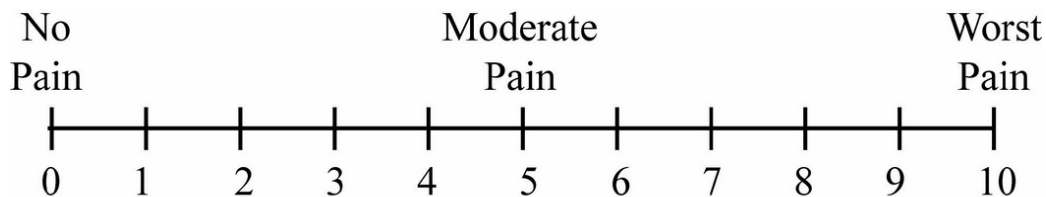
If YES, please provide your contact information below:

Your Name: _____ Phone #: _____

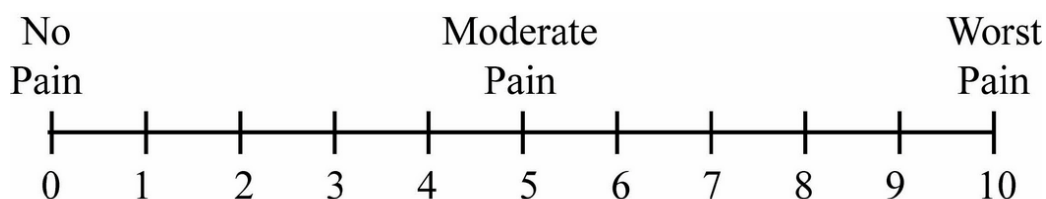
Address: _____

Appendix B. Pain Intensity and Frequency

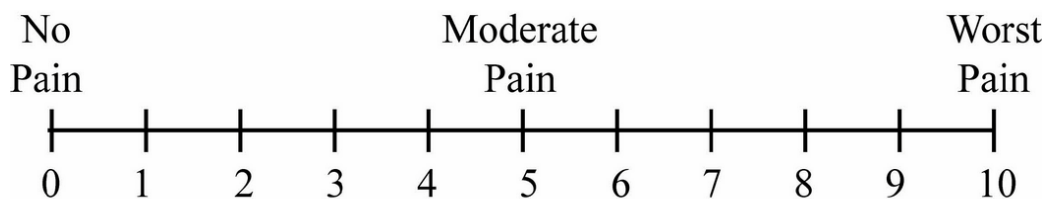
Over the past month, what was your **typical** pain intensity? Please circle.



Over the past month, what was your **worst** pain intensity? Please circle.



What is your **current** pain intensity? Please circle.



How often do you have pain?

- Everyday
- 5-6 days per week
- 3-4 days per week
- 1-2 days per week
- A few days per month
- Never
- Other (explain): _____

How many days have you had pain in the past month? _____

Was this a typical month of pain for you? Yes No (explain)
