Fall 2022

A Longitudinal Study of Pain in Youth and Young Adults with Spina Bifida: Three Studies Based on the Bio-Neuropsychosocial Model

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A LONGITUDINAL STUDY OF PAIN IN YOUTH AND YOUNG ADULTS WITH SPINA BIFIDA: THREE STUDIES BASED ON THE BIO-NEUROPSYCHOSOCIAL MODEL

A DISSERTATION SUBMITTED TO

THE FACULTY OF THE GRADUATE SCHOOL

IN CANDIDACY FOR THE DEGREE OF

DOCTOR OF PHILOSOPHY

PROGRAM IN CLINICAL PSYCHOLOGY

BY

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CHICAGO, IL

DECEMBER 2021

ACKNOWLEDGEMENTS

I would like to express my sincere gratitude to my mentor and committee chair, Dr. Grayson Holmbeck. This project would not have been possible without your guidance and support. You have taught me how to be an impassioned scholar that always views research as a way to serve the children and families with whom we work. You also taught me how to reframe mistakes as points of growth and how to bring humor into the dissertation process. Your mentorship has made me a better researcher and clinician. Should I supervise students in the future, I hope I can be half the mentor you were to me throughout these six years. I would also like to thank my committee members for their patience and excellent input throughout this process. I would like to express my gratitude to my cohort whose support has been unfailing. Thank you for making this journey full of laughter and joy. To Aram, thank you for patiently listening and brainstorming with me over the past year. To my family, I would like to say that I simply would not have become a clinical psychologist without your support. Specifically, I would like to thank my Mom and Dad, who knew I was going to be a psychologist before I did. Your love made all things possible. Finally, I would like to thank the CHATS study families, whose strength cannot be adequately measured with statistics.

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ABSTRACT

The overarching goal of this project was to provide a comprehensive examination of pain in youth and young adults with spina bifida (SB). Using the bio-neuropsychosocial model, this collection of studies explored how pain interacts with multiple domains of functioning in SB. The first study examined biological and psychological factors relevant to the experience of pain. Specifically, we described pain characteristics (e.g., intensity, frequency, etc.) and coping responses, examined relations between pain symptoms and biological factors (e.g., shunt status), and explored longitudinal associations between pain and youth psychological adjustment. This study found that a significant minority of youth with SB experience chronic pain and that pain symptoms are generally not related to biological factors. Further, youth with SB tend to respond to their pain with condition-specific methods of pain relief (e.g., medication), and internalizing symptoms tend to precede pain symptoms. The second study examined longitudinal relations between pain and social factors in SB (parent mental health and family functioning). Results revealed several bi-directional associations between family functioning (i.e., cohesion, conflict) and youth chronic pain. No associations between parent mental health and youth chronic pain were found. The final study explored pain in relation to neuropsychological and psychological factors in young adults with SB. This study found that about 25% of our study sample experienced elevated levels of pain severity and pain catastrophizing, and pain catastrophizing was associated with increased pain severity/interference. Further, while executive dysfunction did not moderate the relationship between pain catastrophizing and pain severity/interference,

significant associations between executive dysfunction and pain catastrophizing were found. Overall, this collection of research can inform the development of pain interventions tailored to both SB and to the individual needs of youth and families with SB.

CHAPTER ONE

INTRODUCTION

Spina bifida (SB) is a common congenital birth defect, which results from the failed closure of the embryonic neural tube during the first month of pregnancy. SB is associated with difficulties across physical, neurocognitive, psychological, and social domains; complications can include hydrocephalus, seizures, neurogenic bowel and bladder, orthopedic complications, and varying degrees of paralysis (Copp et al., 2015). Individuals with SB are often required to follow a complex medical regimen to manage these complications (Zukerman et al., 2011). Further, individuals with SB often struggle with executive functioning deficits, learning problems, increased internalizing symptoms, social skills deficits, and frequent pain (Brown et al., 2008; Rose & Holmbeck, 2007; Holmbeck et al., 2003; Clancy et al., 2005).

The Bio-Neuropsychosocial Model of Adjustment in Spina Bifida

As such, individuals with SB and their families contend with numerous potential stressors. To determine how to best support this population, it is necessary to adopt a theoretical framework that addresses all of these factors and interactions among them. This can be accomplished by utilizing a developmentally oriented bio-neuropsychosocial model. This model, introduced by Holmbeck and Devine (2010), is an adaptation of the more commonly known biopsychosocial model (Engel, 1977). This model purports that biological, neuropsychological, social, and contextual factors interact and influence the adjustment of individuals with SB. Psihogios and colleagues (2017) used this model to aid in understanding medical adherence and

responsibility in SB, an important area of adjustment for this population. This research demonstrated that biological, neuropsychological, family, and peer factors were *all* related to levels of medical autonomy and adherence (Psihogios et al., 2017). Given the complexity of this condition, this comprehensive model will serve as a useful guide when examining other areas of adjustment in SB, such as adjustment to pain.

Pediatric Pain

Pain is defined as "an unpleasant sensory and emotional experience associated with actual or potential tissue damage, or described in terms of such damage" (IASP, 2019), and is typically considered to be either acute or chronic. Acute pain is often thought to be related to a specific disease state or injury, and is limited in duration (Grichnik & Ferrante, 1991). Conversely, chronic pain is defined as pain that persists past the expected time of healing (typically past three months; King et al., 2011). Prevalence rates for pediatric chronic pain range from 20% to 35% (King et al., 2011; Stanford et al., 2008). Headache is the most common form of pediatric pain; other common pain conditions include musculoskeletal pain, headaches, and abdominal pain (King et al., 2011). With regard to demographics, chronic pain tends to increase with age and is most commonly seen in females (King et al., 2011).

Biopsychosocial Model of Pediatric Pain

In line with the model described above, chronic pain has also been thought to be the result of interactions among many different factors, including neurosensory, cognitive, behavioral, social, cultural, and affective factors (Liossi & Howard, 2016). As such, adopting a biopsychosocial model is common in both pediatric and adult pain research (Liossi & Howard, 2016; Gatchel et al., 2007). Indeed, the aforementioned definition of pain, while limited in scope,

recognizes that pain is *both* a sensory and emotional experience and recognizes that it is difficult to establish a single origin of pain.

Pain in Spina Bifida

Recent research has drawn attention to the importance of understanding the incidence and nature of pain in SB. Existing studies have demonstrated that pain is a widespread phenomenon in this population, with prevalence estimates ranging from 25-56% (Ohanian et al., 2020; Clancy et al., 2005). Further, research has found pain to exert a considerable impact on the wellbeing of youth with SB; increased pain symptoms have been associated with elevated internalizing symptoms (Oddson et al., 2006; Ohanian et al., 2020), reduced quality of life (Bellin et al. 2013), and reduced social activity and social competence (Essner et al., 2014). Studies examining the origin of pain in this population have pointed both to condition-related factors (e.g., shunt status and muscle overuse due to ambulation method, Stellman-Ward et al., 1997; Marge, 1994) as well as psychosocial stressors commonly associated with pain symptoms (e.g., internalizing symptoms; Ohanian et al., 2020). The current limited literature highlights the need for further study of how pain emerges, is maintained, and affects individuals with SB.

Bio-Neuropsychosocial Model: Psychosocial Functioning

We will now review components of the bio-neuropsychosocial model relevant to one's adjustment to both SB and pain. The first component of our model is psychosocial functioning. The relationship between pain and psychosocial functioning is well recognized. Indeed, pain is associated with youth and parent psychological adjustment (Noel et al., 2016; Palermo & Eccleston, 2009), family functioning (Lewandowski et al., 2010; Palermo et al., 2014), and peer relations (Forgeron et al., 2010).

Psychological Adjustment

Research has demonstrated bi-directional relations between pain and youth internalizing symptoms (Noel et al., 2016). Pain symptoms are thought to both be a stressor that makes one vulnerable to the emergence of internalizing symptoms, and to be a response to increased psychological stress (Liossi & Howard, 2016). Similar to youth with chronic pain, youth with SB are also at higher risk for internalizing symptoms (e.g., anxiety and depressive symptoms; Holmbeck et al., 2010) than their typically developing (TD) peers. Multiple factors have been associated with internalizing symptoms in SB, including condition severity (Cate et al., 2002), parenting behaviors (Holmbeck et al., 2002), and pain (Oddson et al., 2006).

Parent/Family Functioning

Parent and family functioning also represent important elements of healthy adjustment in pediatric populations. Indeed, similar to individual psychological adjustment to pain, research has also demonstrated bi-directional relations between parent mental health and pediatric pain outcomes (Palermo et al., 2014). Parents of youth with SB also often experience increased psychological distress and caregiver stress (Holmbeck et al., 1997; Vermaes et al., 2005). Indeed, managing the care of a child with increased medical needs can disrupt normative parent and family functioning. Therefore, the current literature has also highlighted the importance of examining broader level family factors in pediatric populations. Research has reported mixed findings with regard to family functioning for both families with a child with chronic pain and families with a child with SB (Lewandowski et al., 2010; Holmbeck & Devine, 2010). Pediatric pain has been associated with both maladaptive family functioning and positive family functioning (Lewandowski et al., 2010). Further, research has found that compared to families of

TD children, families of youth with SB demonstrate less cohesion (Holmbeck et al., 2002) *but also* demonstrate lower levels of conflict (Holmbeck & Devine, 2010). As such, parent/family functioning may represent a risk factor or an area of resilience for both of these populations. Understanding how these associations may function in families of youth with SB *and* chronic pain is critically important, as youth with SB tend more socially isolated than their TD peers (Holmbeck & Devine, 2010), thereby underscoring the unique importance of parent and family relations.

Pain Coping and Pain Catastrophizing

The manner in which one copes with pain is another important element of healthy adjustment. Maladaptive coping has been consistently associated with poor pain outcomes, such as increased pain intensity and disability (Claar et al., 2008). Conversely, adaptive coping strategies have been shown to mitigate the effects of chronic pain (Gauntlett-Gilbert et al., 2013). One of the most impactful and maladaptive forms of coping is pain catastrophizing, or the tendency to magnify and ruminate on the threat of pain (Sullivan et al., 2001). It should be noted that there is debate as to whether catastrophizing is best understood as a coping mechanism vs. a stress response (Connor-Smith et al., 2000; Compas et al., 2012). Many coping frameworks consider it a form of passive coping (Walker et al., 1997), which is also often subsumed under the larger umbrella of emotion-focused coping (i.e., attempting to change one's emotional response vs. attempting to alter the circumstances, often referred to as problem-focused coping or active coping; Lazarus & Folkman, 1984; Walker et al., 1997). While catastrophizing is thought to be distinct from anxiety (Tran et al., 2015) it is still a related construct, and insights regarding the function of worry drawn from the larger anxiety literature may aid our

understanding of catastrophizing as a coping mechanism. Research examining generalized anxiety disorder (GAD) has found that individuals often employ worry as an attempt to gain more control over a problem by anticipating negative outcomes (Newman et al., 2013). Therefore, we might conceptualize pain catastrophizing as a form of pain worry, wherein the individual engaging in this behavior is attempting to gain control over their experience of pain.

Overall, it is critical to examine this coping construct, as it appears to be uniquely and negatively impactful—more so than other commonly cited predictors of pain (e.g., negative affectivity; Vervoort et al., 2006). Relatively little is known about how youth with SB cope with pain; in fact, only one study has examined how youth with SB cope with pain, finding that youth with SB predominately use condition-specific methods (e.g., taking off braces, taking pain-relievers; Ohanian et al. 2020). Further, no studies have directly studied pain catastrophizing in SB.

Bio-Neuropsychosocial Model: The Role of Neuropsychological Functioning

Neuropsychological functioning is associated with individual adjustment to pediatric conditions, such as SB and chronic pain. Individuals with SB commonly demonstrate deficits in executive functioning (EF), or high order attentional, organizational, and goal-directed skills. The cognitive profile of SB often includes reduced working memory, cognitive flexibility, abstract reasoning, planning and organizing, and self-monitoring (Brown et al., 2008; Rose & Holmbeck, 2007; Zabel et al., 2011). These deficits have been linked to multiple CNS factors such as reduced cortical surface area, reduced cerebral white matter, and shunted hydrocephalus (Juranek & Salman, 2010; Ware et al., 2016; Brown et al., 2008). EF deficits have also been found in TD youth and young adults with chronic pain, including problems with inhibitory

control, working memory, and shifting (Berryman et al., 2013; Diamond, 2013; Moriarty et al., 2011). Moreover, studies have also found that individuals tend to select maladaptive pain coping strategies when they have reduced EF, resulting in poorer outcomes (Compas & Boyer, 2001; Bell et al., 2018). In sum, given the prevalence of EF deficits in SB, and research demonstrating associations between pain and EF deficits and associations between poor pain coping and reduced EF, it will be important to examine the role that EF may play in adjustment to pain in SB.

Developmental Context

Finally, when examining pain and SB, we must consider the developmental context. Both pain and SB interrupt typical developmental processes (Palermo et al., 2014). Specifically, during adolescence youth start to separate themselves from their parents and young adults are expected to have achieved autonomy from their families. Both pain and SB limit social interactions with peers (Forgeron et al., 2010; Holmbeck et al., 2010) and increase dependency on parents (Palermo et al., 2014; Zukerman et al., 2011). Further, both pain and internalizing symptoms tend to peak in adolescence (Stanford et al., 2008, Hyde et al., 2008), making an adolescent with SB who experiences chronic pain particularly vulnerable to poor outcomes. Therefore, it is important examine pain in SB during adolescence and young adulthood, as they are extremely impactful periods for TD youth, youth with SB, and youth with chronic pain.

Furthermore, developmental context is equally critical to consider when examining the role of neuropsychological functioning in relation to pain. EF abilities mature throughout childhood and become significantly more developed during adolescence and young adulthood (Best & Miller, 2010). Indeed, many studies have suggested that Miyake and colleagues (2000)

three-factor structure of EF (i.e., updating, shifting, inhibition) is a more useful framework to use when conceptualizing EF in adolescents vs. younger children (Lehto et al., 2010; Wiebe et al., 2008; Lee et al., 2013). Some studies have suggested that these three EF factors are not clearly differentiated from each other during early childhood (i.e., there is significant overlap amongst these skills; Lee et al., 2013; Shing et al., 2010). Further, the neural structures that support EF, such as the prefrontal structures, continue to develop into young adulthood (Tamnes et al., 2010). As noted above, individuals with SB often struggle with cognitive deficits, including executive dysfunction; therefore, it may be particularly informative to examine the contribution of EF to the experience of pain in the context of young adulthood when these executive functions are more likely to be differentiated and further developed.

Overview of Current Studies

This collection of research sought to expand the limited research regarding pain in youth and young adults with SB. The first study, "Longitudinal Associations Between Pain and Psychosocial Adjustment in Youth With Spina Bifida," published in the *Journal of Pediatric Psychology* (Ohanian et al., 2020), focused on associations between pain and individual *biological* and *psychological* factors. This study described the nature of pain in SB (e.g., location, duration, frequency, and intensity) and examined associations between pain and disease and demographic variables. Further, to understand relevant psychological factors, this paper described common coping responses and examined bi-directional longitudinal relations between internalizing symptoms and pain. To address methodological gaps in the literature, this study used two times points and a multi-informant (e.g., child, parent[s], teacher) design. Results suggested that while few condition factors (e.g., shunt status, shunt revisions, lesions level) were

related to pain symptoms, condition-related coping was the most common response to pain. Further, in youth with SB, internalizing symptoms were found to precede, rather than follow pain symptoms.

The second study, "Bi-Directional Associations between Chronic Pain and Parent and Family Functioning in Youth with Spina Bifida," addressed *social* factors relevant to the experience of pain. This study addressed a significant gap in the current literature by examining relations between pain and parent mental health, and relations between pain and family functioning; to our knowledge no studies had yet examined these relations in youth with SB. This study also adopted a bi-directional longitudinal design (two time points) to examine directionality between these variables. This allowed us to gain a better understanding of the role that pain plays with regard to parent and family adjustment, as well as how pain manifests in the face of increased social stressors, such as reduced parent mental health and problematic family functioning. Additionally, this study used a multi-method (e.g., observational and questionnaire) and multi-informant design (parent[s], child).

Finally, the third study, "Pain Catastrophizing in Association with Pain Severity and Pain Interference in Young Adults with Spina Bifida: Executive Functioning as a Moderator," examined both *psychological* and *neuropsychological* factors that may impact pain symptoms in SB. Catastrophizing, a maladaptive coping mechanism, has increasingly been considered one of the most important predictors of adjustment to pain. As such, it is extremely important to understand its relationship to pain and pain interference in SB. Further, given the unique neuropsychological profile of SB, to understand how pain functions in this condition *specifically*, we also examined relations between executive functioning and pain catastrophizing, as well as

between executive functioning, and pain severity and interference. Finally, we examined the potential additive and interactive effect of poor neuropsychological functioning with regard to the relationship between pain catastrophizing, and pain severity and interference.

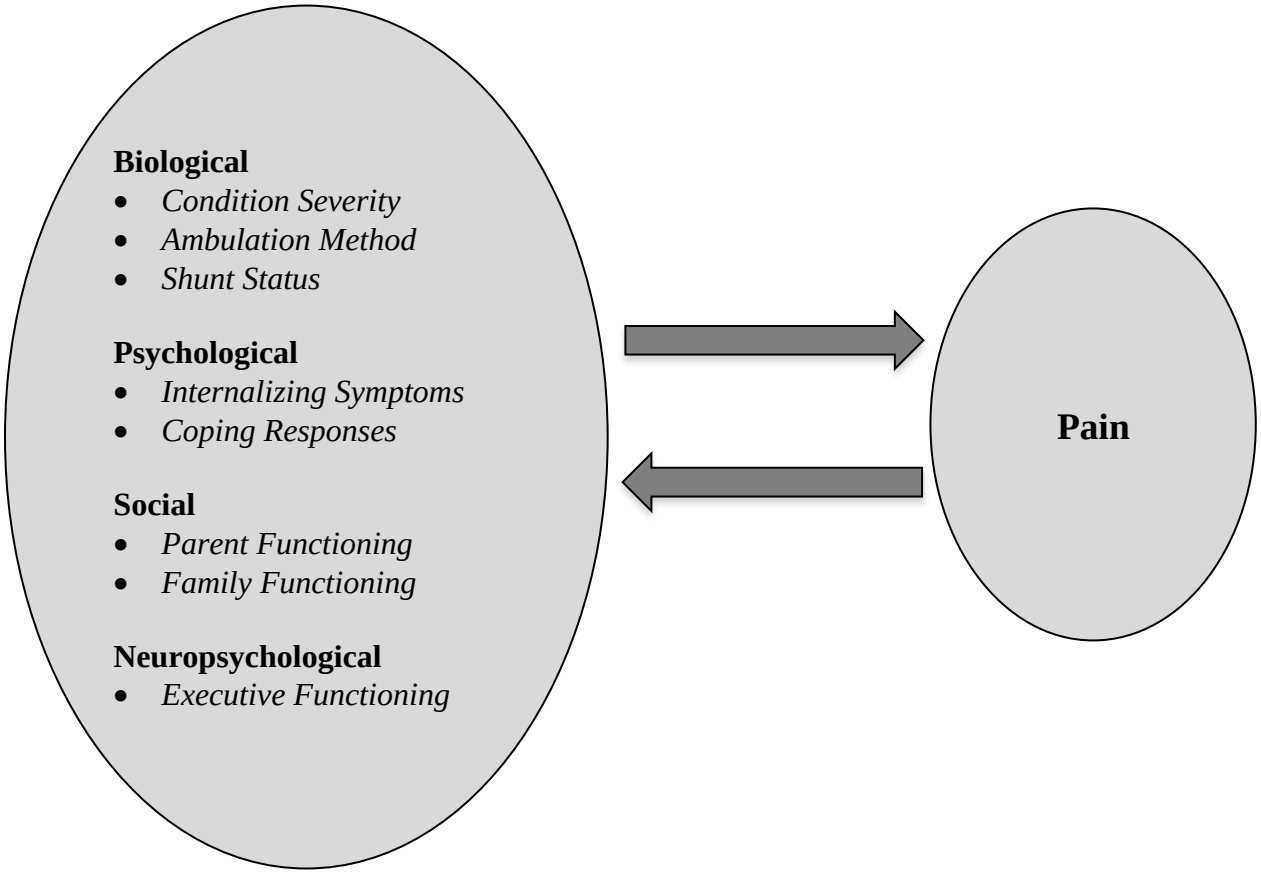


Figure 1. Bio-Neuropsychosocial Model of Pain in Spina Bifida

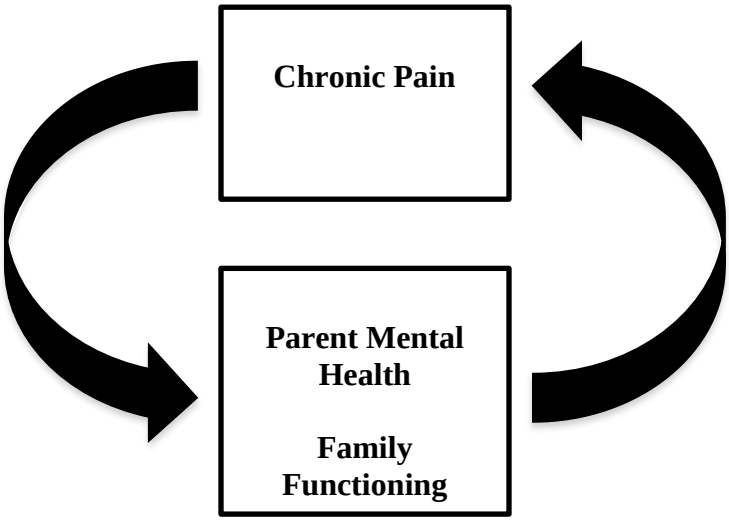


Figure 2. Bi-Directional Relations between Chronic Pain and Parent and Family Functioning

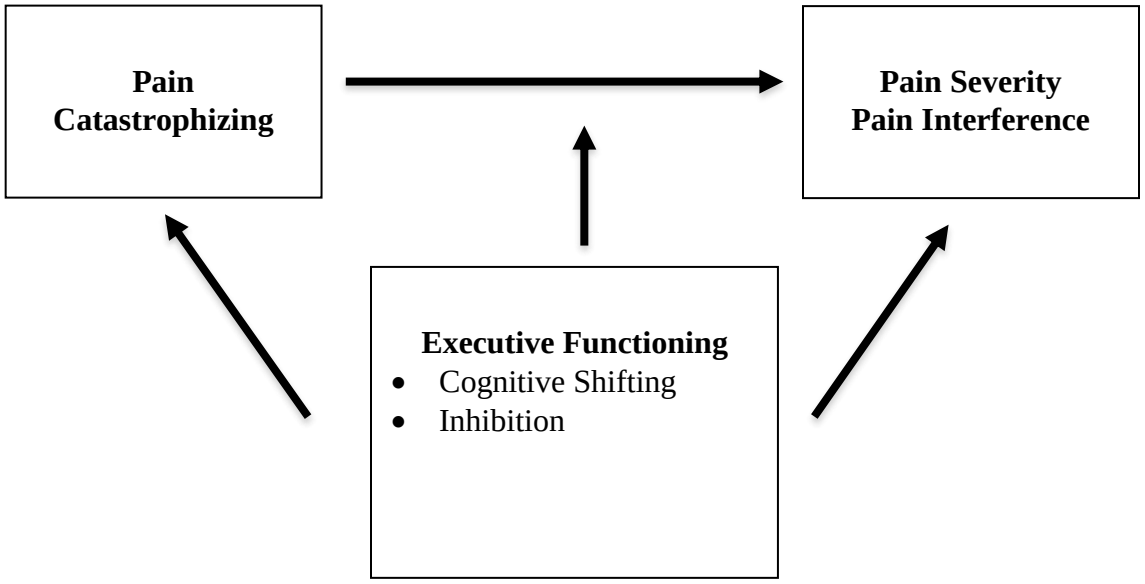


Figure 3. Moderating Role of Executive Functioning on the Relationship between Pain Catastrophizing and Pain Severity/Interference

CHAPTER TWO

LONGITUDINAL ASSOCIATIONS BETWEEN PAIN AND PSYCHOSOCIAL ADJUSTMENT IN YOUTH WITH SPINA BIFIDA

Introduction

Spina bifida (SB) is a relatively common congenital birth defect that occurs in roughly three out of every 10,000 births in the United States (Centers for Disease Control and Prevention [CDC], 2011). SB is associated with a number of complications including bowel and bladder incontinence, varying degrees of paralysis of the lower extremities, orthopedic conditions, hydrocephalus, and neurocognitive issues, all of which require youth to follow a demanding medical regimen (Zukerman et al., 2011). Due to the medical sequelae related to this condition, youth with SB are also at risk for secondary physical conditions, including chronic pain (Clancy et al., 2005; Oddson et al., 2006).

The negative impact of chronic pain on the physical and psychosocial well-being of children and their families is well-established (Liossi & Howard, 2016); nevertheless, research investigating the nature, prevalence, and development of pain in youth with SB remains limited. Of the few studies conducted to date, research suggests that pain is both prevalent and impactful in this population. One study of children and adolescents with SB (ages 8 to 19 years) found that over half of their sample experienced pain at least once per week (Clancy et al., 2005). Moreover, youth with SB may be at risk for untreated pain because parents often underestimate their child's pain (Clancy et al., 2005).

Prior research has begun to report on the type and location of pain in youth with SB, the etiology of which may in part be driven by extensive medical sequelae related to this condition. This research has found that youth with SB most commonly experience headache due to shunt infection or malfunction (Stellman-Ward et al., 1997), and musculoskeletal pain and joint pain due to ambulation method (i.e., overuse of certain muscles necessary for using wheelchairs or crutches; Marge, 1994). However, previous studies have not examined pain characteristics longitudinally wherein one could identify whether pain is consistent across time and affects the same subset of individuals, or changes over time either in its nature or with whom it affects. As such, the first objective of this study was to describe several pain characteristics in this population, including the frequency, intensity, duration, quality, and location of pain.

Given the prevalence of pain in SB, it is also important to consider how youth with SB cope with pain as this can have implications for long-term adjustment. Research has consistently demonstrated associations between less adaptive forms of pain coping and adverse outcomes, such as increased pain intensity, functional disability, and poorer psychosocial functioning in pediatric pain populations (e.g., chronic migraine, chronic musculoskeletal pain, chronic abdominal pain; Claar et al., 2008). However, to our knowledge, no study to date has examined how youth with SB typically cope with pain. Many studies have used the Pain Response Inventory (PRI; Walker et al., 1997), a well-established measure that assesses broad domains of coping (e.g., active, passive, accommodative). Therefore, the second objective of this study was to examine coping responses to pain among youth with SB, which were coded using the PRI framework.

Finally, we examined longitudinal and bidirectional associations between pain and internalizing symptoms in youth with SB. In the larger pediatric literature, pain is strongly linked to reduced psychosocial functioning, particularly increased internalizing symptoms (Noel et al., 2016). Research examining the temporal relationship between chronic pain and internalizing symptoms has found evidence of bi-directional relations between the two constructs; some studies have found pain to precede the onset of internalizing symptoms (Noel et al., 2016), others have found it to follow the onset of internalizing symptoms (Shelby et al., 2013). Much of the literature on temporal relations between these constructs has also reported methodological limitations when making directionality claims (e.g., retrospective self-report). Therefore, the current study sought to explore directionality with multi-informant longitudinal data. Determining the temporal relationship between these two constructs can inform clinical interventions in pediatric SB to prevent the development or maintenance of chronic pain and psychosocial difficulties. In line with trends in recent research, our hypothesis was that internalizing symptoms would predict later pain symptoms and that pain symptoms would predict later internalizing symptoms in youth with SB.

Methods

Participants

Participants were recruited from a larger ongoing longitudinal study examining family and peer relationships, neuropsychological functioning, and psychological adjustment in youth with spina bifida (e.g., Psihogios et al., 2017). The current study represents an analysis of psychosocial functioning and pain symptoms during the first two time points, with each time point spaced two years apart. Families of youth with SB were recruited from four hospitals and a

statewide SB association in the Midwest. Families were recruited in person at regularly scheduled clinic visits or through recruitment letters. Interested families were screened by phone or in person by a trained member of the research team to determine if their child met the following inclusion criteria: (1) a diagnosis of SB (types included myelomeningocele, lipomeningocele, and myelocystocele); (2) age 8-15 years; (3) proficiency in English or Spanish; (4) involvement of at least one primary caregiver; and (5) residence within 300 miles of the laboratory (to allow for data collection at participants' homes). During recruitment a total of 246 families were approached, of which 163 agreed to participate. However, 21 of the 163 families could not be contacted or later declined to participate, and two families did not meet inclusion criteria. The final sample included 140 families of children with SB at Time 1 (53.6% female, M_{age} = 11.40; see Table 1). Youth of families who declined to participate did not differ from participants with respect to type of SB (myelomeningocele or other), χ^2 (1) = .0002, $p > .05$, shunt status, χ^2 (1) = .003, $p > .05$, or occurrence of shunt infections, χ^2 (1) = 1.08, $p > .05$.

Data were collected at Time 2 for 111 (79%) of the original 140 participants. Reasons for attrition at Time 2 (N=29) were as follows: 16 participants declined to participate, 12 participants were unable to be contacted, and 1 participant was deceased. Youth of families who did not participate at Time 2 did not differ from participants with respect to sex, χ^2 (1)=0.28, $p>.05$, SES, $t(128)=-1.86$, $p>.05$, type of SB (myelomeningocele or other), χ^2 (1)=1.19, $p>.05$, lesion level, χ^2 (1)=0.72, $p>.05$, or shunt status, χ^2 (1)=2.73, $p>.05$.

Table 1. Youth Demographic and Spina Bifida Information at Time 1

	Total M (SD) or N (%)
Participants	140 (100%)
Age	11.43 (2.46)
Gender: female	75 (53.6%)
Primary Language: Spanish	28 (20.0%)
Race	
Caucasian	74 (52.9%)
African American/Black	19 (13.6%)
Hispanic/Latino	39 (27.9%)
Asian	2 (1.4%)
Bi-racial	6 (4.3%)
Spina bifida type	
Myelomeningocele	122 (87.1%)
Non-myelomeningocele	17 (12.1%)
Unknown/not reported	1 (0.7%)
Lesion level	
Thoracic	23 (16.4%)
Lumbar	69 (49.3%)
Sacral	41 (29.3%)
Unknown/not reported	7 (5.0%)
Shunt: present	109 (77.9%)
IQ	85.75 (19.54)
Family SES	39.12 (16.09)
T1 Anxious/Depression (P)	54.97 (6.08)
T1 Withdrawn/Depression (P)	56.18 (6.56)
T2 Anxious/Depression (P)	54.11 (5.16)
T2 Withdrawn/Depression (P)	55.30 (5.48)
T1 Anxious/Depression (T)	56.22 (6.89)
T1 Withdrawn/Depression (T)	57.34 (7.89)
T2 Anxious/Depression (T)	55.03 (6.31)
T2 Withdrawn/Depression (T)	57.04 (8.54)
T1 Depressive symptoms (C)	1.31 (.23)
T2 Depressive symptoms (C)	1.27 (.19)

Note. M = Mean; SD (Standard Deviation); T1=Time 1, T2=Time 2. Anxious/Depressive and Withdrawn/Depressive subscales are parent and teacher report on the Child Behavior Checklist (CBCL); Child-reported depressive symptoms are youth report on the Child Depression Inventory (CDI). P=Parent, T=Teacher, C=Child.

Procedure

The current study was approved by university and hospital Institutional Review Boards. Trained undergraduate and graduate student research assistants collected data during home visits that lasted approximately three hours. At Time 1, two separate three-hour home visits were conducted. At Time 2 only one three-hour home visit was conducted. At home visits with families who primarily spoke Spanish (N=28), at least one research assistant who was fluent in Spanish was present. Prior to each home visit, informed consent from parents and assent from children were obtained. At Time 1, 26 of the 140 participants were estimated to have an IQ below 70. These youth were assented and filled out the questionnaires when possible. Youth who were nonverbal did not fill out the questionnaires. Research assistants were trained to judge the validity of their responses based on the consistency of their answers. If their answers were not considered valid, they were not included in analyses. This resulted in a total of 4 participants at Time 1 being excluded from analyses. Parents also completed releases of information to obtain data from medical charts, health professionals, and teachers. After obtaining releases, questionnaires were sent to participant's current teachers. During data collection, family members completed questionnaires independently. Questionnaires were offered in both English and Spanish. When questionnaires were only available in English, members of the research team, who were native Spanish speakers, translated and back translated the questionnaires. The questionnaires were counterbalanced to avoid order effects. Research assistants read questionnaires aloud to participants when requested or when the youth appeared to be having reading difficulties. The current study used youth-, parent-, and teacher -reported questionnaire

data. Families received $150 and small gifts (e.g., logo t-shirts, pens, water bottles) as compensation for participation at each time point.

Measures

Demographics. At Time 1, parents reported on family and youth demographic information, including age, gender, race/ethnicity, income, education, and employment. The Hollingshead Four Factor Index of Socioeconomic Status (SES) was computed using parent's education and occupation, with higher scores indicating higher SES (Hollingshead, 1975).

Illness severity. At Time 1, parents completed the Medical History Questionnaire (MHQ; Holmbeck et al., 2003), which included questions about youth's disease-specific medical information including bowel and bladder functioning, ambulation method (i.e., ankle-foot orthoses [AFOs], knee-ankle-foot orthoses [KAFOs], hip-knee-ankle-foot orthoses [HKAFOs], wheelchair, or no assistance), medications, frequency of medical care, and surgery history. In addition to the MHQ, data were collected from medical charts to assess type of SB (i.e., lipomeningocele, meningocele, or myelomeningocele), shunt status, and lesion level (i.e., sacral, lumbar, or thoracic). These variables were used to compute an illness severity index based on membership in specific groups: shunt status (no = 1, yes = 2), myelomeningocele (no = 1, yes = 2), lesion level (sacral = 1, lumbar = 2, thoracic = 3), and ambulation status (no assistance/AFOs = 1, KAFOs/HKAFOs = 2, wheelchair = 3). Illness severity scores ranged from 4 to 10, with higher scores indicating higher levels of severity (Hommeyer et al., 1999).

Youth IQ. At Time 1, youth were administered the Vocabulary and Matrix Reasoning subtests of the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999). These subtests were used to estimate a Full-Scale Intelligence Quotient (IQ) score. The vocabulary

subtest consists of 42 items, which measure an individual's expressive vocabulary, verbal knowledge, and fund of information. The matrix reasoning subtests consists of 35 items which measure a child's nonverbal fluid reasoning and general intellectual ability. These subtests have demonstrated high levels of internal consistency for youth 6-16 years old (α = .89 for Vocabulary, α = .92 for Matrix Reasoning; Wechsler, 1999).

Internalizing symptoms. Internalizing symptoms were measured using youth-, mother-, father-, and teacher- report. At Times 1 and 2, youth reported on 27 depressive symptoms on the Child Depression Inventory, a well-validated measure of depression (CDI; Kovacs, 1992); higher scores indicated greater symptomatology. The CDI demonstrated acceptable levels of internal consistency and Times 1 and 2 (α=.82; α= .78). Parents completed the Child Behavior Checklist (CBCL) and teachers completed the Teacher Report Form (TRF; Achenbach & Rescorla, 2001), well-validated broad-based measures of youth psychosocial functioning. Parents whose primary language was Spanish completed the Spanish version of the CBCL; this questionnaire has demonstrated strong validity and reliability in Spanish and is commonly used in contemporary research (Rescorla et al., 2007). This study used mother, father, and teacher report on two internalizing subscales from the CBCL: Anxious/Depressed and Withdrawn/Depressed at Times 1 and 2. The anxious/depressed scale demonstrated acceptable reliability across reporters and at both time points: mother (T1: α=.76, T2: α=.70), father (T1: α=.68, T2=.70), and teacher (T1: α=.65, T2: α=.74). Further, the withdrawn/depressed scale also demonstrated acceptable reliability across reporters and at both time points: mother (T1: α=.72, T2: α=.69), father (T1: α=.76, T2: α=.70), and teacher (T1: α=.84, T2: α=.87).

Pain characteristics. We examined a variety of youth-reported pain characteristics, including intensity, frequency, duration, location, and quality over the last three months. *Pain intensity* was measured via youth report on a Visual Analogue Scale; youth marked a point along a 10-cm line ranging from "no pain" to "worst pain ever" (see Palermo, Zebracki et al., 2004). Pain intensity was categorized according to severity (e.g., 0.0-0.9: none/minimal, 1.0-3.9: mild, 4.0-6.9: moderate, 7.0-10.0: severe; Breivik et al., 2008). *Pain frequency* was measured on a 6-point Likert-type scale, ranging from "0-less than once a month" to "5-daily" (Palermo, Valenzuela et al., 2004). Youth were considered to have chronic pain if they reported experiencing pain one or more times per week over the past three months (i.e., corresponding to a rating of a 2 on the 0-5 scale; King et al., 2011). *Pain duration* was measured on a 4-point Likert-type scale, ranging from "0- less than 1 hour" to "3 -all day" (Palermo, Valenzuela et al., 2004). Youth also marked *pain location*, or where they experienced pain on a validated body outline, which showed an anterior and posterior view of the body (Savedra et al., 1989); up to four responses were coded for each participant. Finally, *pain quality* was measured by youth checking off which descriptor(s) (e.g., sharp, aching, stinging, etc.) best captured the nature of their pain.

Coping behaviors in response to pain. Youth were also asked to list "what helps you feel better" when experiencing pain due to SB. This question resulted in brief written responses such as "watch TV," "drink cranberry juice," or "take medicine." Up to three of these responses were written per participant (though many people wrote fewer than three; range=1-3). Initially, these responses were categorized into 152 very specific behavior categories (e.g., drink cranberry juice, watch TV).

Next, we categorized these 152 specific behaviors based on the factors of the Pain Response Inventory (PRI; Walker et al., 1997), a frequently used measure for assessing child and adolescent coping behaviors in response to pain. Although the PRI has not yet been used to code free-written responses in this manner, our team sought to draw from an established, validated, and widely used measure to guide our coding of youths' coping responses to pain. Two raters (a clinical psychology graduate student and a clinical psychology professor) independently coded each of the 152 behaviors into one of the first-order factors (e.g., social support, massage, distract/ignore) based on a comparison of the behavior to the specific items on the PRI and their respective factors. Interrater kappa values ranged from .87 to .99 for each first-order factor, indicating a very high level of agreement between raters. In several instances (N=45) raters coded a behavior into more than one coping category (e.g., "mom rubbing tummy" was coded as both massage and social support). When there was disagreement, both reviewers discussed their response and came to a consensus. While the coders categorized all of the responses from the entire sample, descriptive analyses are limited to the chronic pain subsample because the PRI was intended for use with pediatric chronic pain samples.

Statistical Treatment

Prior to hypothesis testing, psychometric properties were evaluated and composite scores were created when possible to reduce the number of analyses and decrease the possibility of shared method variance.[1] Associations between measures in which there were two reporters (e.g., mother-report, father-report) were calculated using Pearson correlation coefficients; associations

[1]Both parent and teacher report on the CBCL anxious/depression and withdrawn/depression scales, Children's Depression Inventory total scores and pain intensity were mildly skewed at both time points. Analyses were run first with untransformed variables and then with square root transformations. Because findings were the same results presented here are with the untransformed variables.

between three or more informants were calculated using Cronbach's alpha coefficients (e.g., youth, mother, father, teacher). Criteria of $r \geq .40$ and $\alpha > .60$ were used, respectively, to determine which measures could be collapsed across reporters (Holmbeck et al., 2002). Parent, youth, and teacher report of depressive symptoms (both anxious/depressed and withdrawn/ depressed) were not highly associated. However, mother and father report of anxious/depressed and withdrawn/depressed symptoms were adequately correlated ($r > .40$) with each other at both time points and, therefore, were combined to create composite "parent-report" internalizing scores at Times 1 and 2. Thus, parent, youth, and teacher report of internalizing symptoms were examined separately in the analyses.

Descriptive statistics including mean, standard deviation, and percentage frequency were used to address the first two aims of this study which involved describing the nature and prevalence of pain (Aim 1) and common behavioral coping responses (Aim 2). This study included descriptives at Time 1 and Time 2 for pain chronicity, intensity, and duration; all other descriptive statistics (location, quality, and coping responses) represent responses from Time 1 only. To address Aim 3, a series of hierarchical multiple linear and logistic regression analyses were conducted to examine associations between (1) internalizing symptoms at Time 1 and pain symptoms (intensity and chronicity) at Time 2, and (2) pain symptoms at Time 1 and internalizing symptoms at Time 2. The "pain chronicity" variable was created by dichotomizing the pain frequency variable into those who had chronic pain and those who did not have chronic pain (see above for criteria for determining those who have "chronic pain.") Logistic regressions were used when examining pain chronicity as an outcome. Several covariates were chosen due to their potential influence on increased risk of pain and internalizing symptoms, including older

child age and female gender (Hyde et al., 2008; King et al., 2011), and lower SES (Dorner et al., 2012; Schreier & Chen, 2013). In addition, previous research has revealed associations between illness severity and psychosocial and physical functioning in this population (Hommeyer et al., 1999), thus we controlled for illness severity in analyses. Variables were entered in the following order: (Step 1) internalizing or pain symptoms at Time 1; (Step 2) covariates: illness severity, age, SES, gender, illness severity; and (Step 3) individual predictor (pain symptom [child report] or internalizing symptoms [teacher and parent report of anxious and withdrawn depressive symptoms, and child report of depressive symptoms]). Separate regressions were conducted for each combination of predictor and outcome variables. Missing data was handled with listwise deletion.

Results

Description of Study Sample

The total sample included 140 children and adolescents with SB ages 8-15, (53.6 % female; 52.9% Caucasian), with the majority having myelomeningocele SB as well as a shunt. About half of the sample had a lumbar level lesion, the mean IQ was 85.68 (Low Average), and mean family SES was 39.44. Associations between Time 1 pain symptoms and demographic variables revealed that lower IQ was associated with greater pain chronicity (r= -.22, p<.05). Further, gender was significantly associated with pain intensity at Time 1 (r=.27, p<.01), such that females with SB reported greater pain intensity than males with SB. No significant bivariate associations were found between age, socioeconomic status, or race/ethnicity and Time 1 pain symptoms. With regard to illness-related variables, no significant bivariate associations were found between shunt status, lesion level, or illness severity and Time 1 pain symptoms.

Objective 1: Describing the nature of pain in spina bifida. For the full sample at Time 1, 29.8% (N=34) of youth reported moderate or severe levels of pain. At Time 1, about 25% (N=31) of the sample reported chronic pain (e.g., pain occurring at least once per week over the past 3 months), with over 10% of youth reporting daily pain (N=13). Moreover, about 8% (N=10) of the sample reported chronic pain at both Time 1 and Time 2 (see Table 2). Pain frequency and intensity were similar at Time 2. Moreover, pain was predominately described as aching (42.4%, N=50), and lasting less than an hour (55.8%, N=43). The most common pain locations included back, extremities (e.g., hands, arms, legs), and abdomen, and head (see Table 2 for a description of pain characteristics for the full sample).

For the chronic pain subsample, pain intensity was moderate to severe for 62.5% of the sample at Time 1. Moreover, a sizeable portion of the chronic pain subsample reported pain symptoms ranging in duration from a few hours to half of the day (40.8%, N=11), with 14.8% of this subsample (N=4) reporting experiencing pain all day. Pain was typically located in the extremities (30.8%, N=20), back (26.2%, N=17), and head (18.5%, N=12), and was overwhelmingly characterized as aching (73.3%, N=22).

Table 2. Descriptive Data for Pain Frequency, Intensity, Duration, and Quality

Variable	Time 1		Time 2	
Intensity	N	%	N	%
None/Minimal	45	39.5	44	43.1
Mild	35	30.7	33	32.3
Moderate	17	14.9	17	16.7
Severe	17	14.9	7	6.9
Frequency	N	%	N	%
No pain	34	27.3	32	30.0
Less than once per month	38	30.8	36	33.6
1 to 3 times per month	20	16.3	12	11.2
1 time per week	6	4.9	7	6.5
2 to 3 times per week	8	6.5	6	5.6
3 to 5 times per week	4	3.3	6	5.6
Daily	13	10.6	8	7.5
Chronic Pain	31	24.8	27	25.2
Chronic Pain at Time 1 and Time 2			10	8
Duration	N	%	N	%
Less than 1 hour	43	55.8	38	52.1
A few hours	15	19.5	17	23.3
Half the day	13	16.9	10	13.7
All day	6	7.8	8	11.0
Pain Quality				
Aching	50	42.4		
Dull	21	17.8		
Sharp	19	16.1		
Throbbing	17	14.4		
Stinging	15	12.7		
Pounding	13	11.0		
Burning	10	8.5		
Hammering	7	5.9		
Cutting	6	5.1		

Note. M=Mean, SD=Standard Deviation, N=sample size. For pain quality, youth were able to check all descriptors that applied. Percentages reflect frequency of chosen descriptors. Pain intensity is presented according to severity categories (0.0-0.9: none/minimal, 1.0-3.9: mild, 4.0-6.9: moderate, 7.0-10: severe)

Objective 2: Youth response to pain. It was possible to code responses into seven of the 13 original PRI's primary subscales. These included condition-specific, rest, massage, problem

solving, distraction, social support, and behavioral disengagement. For the chronic pain subscale, it was most common for youth to report using condition-specific methods when they experienced pain (57.7%; N=15). Common condition-specific methods included over-the-counter pain medications (e.g., Tylenol and Advil) and drinking cranberry juice, which is frequently associated with preventing and treating urinary tract infections. Resting was the second most common method listed for ameliorating pain (30.8%, N=8). Responses also revealed that youth with SB frequently use massage (23.1%, N=6) and engage in problem solving (19.2%, N=5) to address pain. Some youth also reported using distraction (15.4%, N=4) or seeking social support (11.3%, N=3). Finally, only one participant provided a response consistent with behavioral disengagement (3.8%). No participants generated methods that could be coded as self-isolation, stoicism, catastrophizing, acceptance, minimizing, or self-encouragement.

Objective 3: Longitudinal, bidirectional associations between pain and psychosocial functioning (see Tables 3 and 4). Hierarchical logistic and linear regressions revealed significant longitudinal associations between Time 1 internalizing symptoms and Time 2 pain symptoms. Assumptions were met for both logistic and linear regressions; logistic regressions passed the Hosmer and Lemshow Test and linear regressions did not demonstrate multicollinearity. Regression analyses included five predictors and two outcomes, resulting in a total of 10 analyses. After entering the covariates, greater teacher reported anxious/depressed symptoms at Time 1 significantly predicted chronic pain group status at Time 2 (Exp(b)=1.11, Wald=5.14, p<.05; see Table 3), indicating that increased anxious/depressive symptoms increased the odds of being in the chronic pain group by a small but significant amount. Further, greater child-reported depressive symptoms at Time 1 significantly predicted higher pain intensity at Time 2 (β=.25,

$p<.05$, $\Delta R^2=.05$, Table 4), and accounted for about 5% of the variance, indicating again a small but significant effect. While Time 1 child-reported depressive symptoms were significantly correlated with Time 2 chronic pain, in the logistic regression with covariates, this association was no longer significant (see Table 3). Of note, while few demographic and no illness-related variables were significantly associated with pain in bivariate correlations, significant associations were seen in longitudinal regressions. Specifically, age was related to chronic pain group membership, such that older youth at Time 1 were more likely to be in the chronic pain group at Time 2 (see Table 3). Further, illness severity was significantly associated with change in pain intensity at Time 2 in longitudinal regression, such that lower illness severity at Time 1 was significantly associated with greater pain intensity at Time 2 (see Table 4). No significant associations were found between parent-reported depressive symptoms at Time 1 and chronic pain or pain intensity at Time 2 in longitudinal logistic and linear regressions, respectively (see Table 4). With regard to examining the alternative direction, or associations between Time 1 pain symptoms and Time 2 internalizing symptoms, results revealed no significant associations. This finding was consistent across pain symptoms (chronic pain and pain intensity) and across reporters (parent, teacher, child).

Table 3. Longitudinal Hierarchical Logistic Regression Analyses for Covariates and Teacher, Parent, and Child Depressive Symptoms at Time 1 Predicting Chronic Pain Group Membership at Time 2

Teacher-Report			
Predictor	b	Exp(b)	Wald
Chronic Pain (T1)	.17	1.18	.04
Youth Age	.35	1.42	6.08*
SES	.03	1.03	1.40
Illness Severity	-.30	.74	2.35
Gender	.44	1.60	.42
Teacher Reported Anxious/Depressed Symptoms (T1)	**.10**	**1.11**	**5.24***
Chronic Pain (T1)	.62	1.86	.75
Youth Age	.34	1.41	6.10*
SES	.03	1.03	1.37
Illness Severity	-.22	.80	1.46
Gender	.51	1.67	.59
Teacher Reported Withdrawn/Depressed Symptoms (T1)	.03	1.03	.55
Parent-Report			
Chronic Pain (T1)	.74	2.10	1.05
Youth Age	.38	1.45	7.26**
SES	.02	1.02	.91
Illness Severity	-.24	.79	1.73
Gender	.48	1.62	.57
Parent Reported Anxious/Depressed Symptoms (T1)	-.01	.99	.01
Chronic Pain (T1)	.74	2.10	1.10
Youth Age	.37	1.45	7.53**
SES	.02	1.02	.93
Illness Severity	-.24	.79	1.74
Gender	.49	1.63	.60
Parent Reported Withdrawn/Depressed Symptoms (T1)	-.01	.99	.03
Child-Report			
Chronic Pain (T1)	.85	2.34	1.69
Youth Age	.40	1.49	8.28**
SES	.03	1.03	1.66
Illness Severity	-.19	.83	1.25
Gender	.48	1.61	.59
Child Reported Depressive Symptoms (T1)	1.81	6.12	1.17

Note: All predictor variables were measured at Time 1. The covariates of SES, youth age, illness severity, and gender were entered in as a block. Chronic pain at Time 1 was entered at Step 1. *$p < .05$, ** $p < .01$

Table 4. Longitudinal Hierarchical Multiple Linear Regression Analyses for Covariates and Teacher, Parent, and Child reported Depressive Symptoms at Time 1 Predicting Pain Intensity at Time 2

Teacher-Report			
Predictor	b	β	ΔR^2
Pain Intensity (T1)	.35	.31	.10*
Youth Age	.06	.14	.14
SES	.01	.14	.14
Illness Severity	-.18	-.28	.14*
Gender	.28	.14	.14
Teacher Reported Anxious/Depressed Symptoms (T1)	.01	.06	.00
Pain Intensity (T1)	.35	.31	.10**
Youth Age	.05	.14	.14
SES	.01	.10	.14
Illness Severity	-.18	-.28	.14*
Gender	.28	.14	.14
Teacher Reported Withdrawn/Depressed Symptoms (T1)	-.01	-.07	.00
Parent-Report			
Pain Intensity (T1)	.35	.34	.11*
Youth Age	.06	.15	.13
SES	.01	.14	.13
Illness Severity	-.16	-.26	.13*
Gender	.45	.23	.13
Parent Reported Anxious/Depressed Symptoms (T1)	.03	.17	.02
Pain Intensity (T1)	.36	.34	.11**
Youth Age	.06	.15	.13
SES	.01	.14	.13
Illness Severity	-.16	-.26	.13*
Gender	.36	.19	.13
Parent Reported Withdrawn/Depressed Symptoms (T1)	.01	.05	.00
Child-Report			
Pain Intensity (T1)	.35	.34	.12**
Youth Age	.05	.11	.12
SES	.01	.19	.12
Illness Severity	-.14	-.23	.12
Gender	.36	.18	.12
Child Reported Depressive Symptoms (T1)	**1.22**	**.23**	**.05***

Note: All predictor variables were measured at Time 1. The covariates of SES, youth age, illness severity, and gender were entered in as a block. Pain intensity at Time 1 was entered at Step 1. *$p < .05$, ** $p < .01$

Discussion

To date, pain in youth with SB has been understudied and underreported. Thus, the purpose of the current study was to describe the nature of pain in SB, examine how youth cope with their pain, and examine longitudinal, bidirectional associations between pain and internalizing symptoms. Results revealed that a small but significant minority of youth with SB experience chronic pain. Those that experience chronic pain typically employ condition-specific methods to alleviate their pain. Further, this study demonstrated that internalizing symptoms often precede chronic pain and pain intensity, highlighting the need for interventions targeting youth psychosocial functioning and regular assessment of youth pain symptoms.

The current study examined a variety of pain characteristics in this population. The prevalence of chronic pain (25%) in this sample was similar to what has been found in the larger pediatric pain literature (King et al., 2011). Moreover, pain intensity predominately fell in the minimal to mild range, however a significant portion of the full sample and the majority of the chronic pain subsample reported experiencing moderate to severe pain. Location of pain resembled that found in previous studies, with youth reporting pain to be predominately located in the extremities, back, abdomen, and head (Clancy et al., 2005). Further, this study examined quality of pain, an underreported construct. Both the full sample and chronic pain subsample predominately described pain as aching. Pain literature has suggested that this information can be of use to better understand the etiology of pain as different sources of pain are often associated with a differential quality (Schilder et al., 2018). Indeed, aching pain may be related to muscle overuse or stress on joints related to ambulation methods. Given the limited understanding of

pain in SB, pain quality may help clinicians better understand the etiology of pain and develop an appropriate treatment plan.

Overall, these results suggest that youth with SB are vulnerable to the development of chronic pain. SB is a complex condition accompanied by a myriad of physical sequelae that require constant management. As such, pain may be under-identified and undertreated compared to other more pressing symptoms. Increased screening and education efforts including patient, family, and provider awareness of the risk for chronic pain in pediatric SB is needed. Moreover, given that 8% of the sample experienced continued, unresolved chronic pain two years later, there is likely a subgroup of youth with SB suffering from disabling, persistent chronic pain who require more intensive pain treatment. Clearly, there is a need to increase prevention and intervention efforts in this pediatric population.

Regarding the relationship between demographic variables and pain symptoms, our findings highlight that females, older children/adolescents, and those with cognitive impairments may be at higher risk for experiencing pain symptoms. Notably, lower intellectual functioning was associated with pain chronicity. There are several possible reasons for this finding. It is possible that those with a lower IQ over-endorsed or misunderstood this question. On the other hand, youth with cognitive impairments have been found to experience pain more frequently than their TD peers (Breau et al., 2003). Moreover, the current literature has also demonstrated associations between chronic pain and reduced attention and executive functioning (EF; Moriarty et al., 2011), both of which are critical components of cognitive functioning and are often reduced in youth with SB (Rose & Holmbeck, 2007). Overall, these findings indicate that clinicians should be aware of these demographic risk factors for pain. Teaching health care

professionals how to assess pain in populations with cognitive impairments such as SB may be particularly critical (e.g., with behavioral observation measures; Cohen et al., 2008).

This was also the first study to examine how youth with SB cope with chronic pain symptoms. Results revealed that condition-specific methods were used most frequently, followed by resting, massage, problem solving, and distraction. Methods were primarily active, which is not surprising given that the prompt asked youth to describe what makes their pain feel better, rather than asking them how they cope with pain. Active strategies tend to be more behavioral where the person is *doing something,* whereas accommodative and passive strategies are frequently cognitive in nature (Shirkey et al., 2011). However, the more frequent use of condition-specific methods may reflect a sort of pragmatism to coping with pain, which may be due to a perception of pain as *only* condition-specific in nature. Coupled with our findings indicating there were few significant relations between pain and condition-related characteristics *and* identifying internalizing symptoms as a predictor of pain intensity and frequency, these results suggest a greater need for children with SB and their families to be taught psychological-based strategies for pain management. Skills derived from cognitive-behavioral (e.g., deep breathing) and Acceptance and Commitment-based Therapy (ACT) skills (e.g., mindfulness, acceptance) are effective methods for the treatment of chronic pain (Gauntlett-Gilbert et al., 2013); however, it is possible that youth with SB and their families are not commonly taught these alternative pain management strategies due to the other rigorous demands of this medical condition. Indeed, biopsychosocial models of pain highlight the multifaceted, dynamic nature of pain (Liossi & Howard, 2016) and include condition specific, intraindividual (psychological), and family (parenting) factors that exacerbate pain. Such factors have not been examined in

depth in this population and deserve further attention in order to develop a comprehensive approach to treating pain in SB.

Our longitudinal examination of pain symptoms and internalizing symptoms revealed that internalizing symptoms predicted the chronicity and future levels of intensity of pain in this sample, over and above demographic and clinical factors. These results indicate that an unexplored etiology of pain in SB may be the presence of depressive or anxious symptomology. Moreover, internalizing symptoms may serve to amplify otherwise acute and mild condition-related pain. Notably, associations were only found between teacher-reported and self-reported youth internalizing symptoms and pain. Similar findings involving teacher-reported data have been demonstrated in other research examining youth psychological functioning in SB (e.g., Stern et al., 2018) possibly indicating that the connection between pain and psychosocial functioning is particularly salient in the school environment. In other words, health providers who rely heavily on parent-report of youth psychosocial functioning may be missing important information about youth wellbeing and critical areas for early intervention. Therefore, it is imperative that stress, coping, and adjustment in the context of school be assessed by providers and parents, and that interventions that focus on peer and school-based stress be developed for youth with SB.

This study had several strengths, including the use of multiple methods and reporters of internalizing symptoms, comprehensive assessment of pain characteristics, and a longitudinal design. However, there were several limitations that should be noted. First, pain was only measured with youth report. Guidelines for best practice have recommended the use of multiple reporters, particularly for younger children and children with cognitive deficits (Cohen et al.,

2008). Although research has consistently demonstrated strong associations between anxiety and pain (Shelby et al., 2013), the current study unfortunately did not include specific measures of anxiety. Finally, while the longitudinal design is a strength of the study, our findings did not capture the day-to-day temporal relations among the variables of interest; future research may use daily diaries to advance understanding of potential bidirectional relationships between internalizing symptoms and pain in pediatric SB. Further, it is important to recognize that most regression analyses were not significant; therefore, findings should be interpreted with caution. Moreover, while having multiple reporters and measures was a strength of this study, findings were based on numerous regression analyses, increasing the likelihood of Type 1 error. Finally, effect sizes were small, indicating that internalizing symptoms are likely only one of several clinically significant factors to address in the prevention and treatment of pain in SB.

There were also both strengths and limitations with our approach to measuring coping responses to pain. While we coded written responses using the PRI as a theoretical framework (Walker et al., 1997), this measure is intended to be used in questionnaire format. Moreover, since the question that was asked focused on what one *does* that is *helpful*, most responses may have naturally fell into the active coping second-order factor. Relatedly, passive coping (e.g., catastrophizing), is typically linked to poorer outcomes, and thus we are likely missing important information about the use of passive strategies that youth with SB may understand to be *unhelpful* but use nonetheless. These concerns notwithstanding, this was still the first study to examine pain coping in this population. Therefore, this initial examination of pain coping in youth with SB may serve as a launching point for future studies to more systematically examine pain coping in youth with SB.

Conclusions and Clinical Implications

The results of this study have important clinical implications for promoting psychological and physical well-being in youth with SB. About 25% of youth with SB experience chronic pain and 10% experience daily pain. Pain is often understudied and undertreated in youth with SB, and there is a critical need for increased research and clinical attention to ameliorate pain in this population. Further, similar to the greater pediatric pain literature, internalizing symptoms are related to pain in this population, and may be key drivers of the development and chronicity of pain symptoms. While pain in this population may be related to condition-specific factors, it also may be the result of poor psychological functioning, in line with biopsychosocial models of pain. Therefore, results from this study provide a critical first step in understanding the etiology of chronic pain in youth with SB and underscore the importance of treatment for internalizing symptoms to ameliorate ongoing pain and suffering in children as they grow older. Future research should continue to expand our knowledge of the multifaceted etiology of pain in this population in order to facilitate the development of effective pain interventions for youth with SB and their families.

CHAPTER THREE

BI-DIRECTIONAL ASSOCIATIONS BETWEEN CHRONIC PAIN AND PARENT AND FAMILY FUNCTIONING IN YOUTH WITH SPINA BIFIDA

Introduction

Spina bifida (SB) is a congenital birth defect, occurring early in pregnancy, which results from of the failed closure of the embryonic neural tube (Copp et al., 2015). SB is associated with numerous physical, neurocognitive, and psychosocial difficulties (Copp et al., 2015). Physical complications include bowel and bladder incontinence, potential paralysis of the lower extremities, orthopedic conditions, and hydrocephalus (Zukerman et al., 2011). Recent research has also recognized pain as another important secondary condition parameter in this population. Regarding prevalence, Clancy et al. (2005) found that 56% of their sample of youth with SB, ages 8-19, experienced pain at least once per week. Further, a more recent study examining youth with SB (ages 8-15) reported a lower, yet sizeable prevalence rate, with 25% of their sample reporting pain one or more times per week over the past three months (Ohanian et al., 2020). While limited, the current literature has also highlighted the impact of pain in this population; research has demonstrated associations between pain and several psychosocial variables, including social competence (Essner et al., 2014), quality of life (Bellin et al. 2013), and internalizing symptoms (Oddson et al., 2006; Ohanian et al., 2020). These studies have demonstrated the importance of examining and developing interventions for youth with SB and

chronic pain; nevertheless, other impactful and modifiable areas of psychosocial functioning have yet to be explored, such as relations between pain and parent/family functioning.

Current research has identified bi-directional associations between parent mental health, and youth physical and psychosocial functioning (Cousino & Hazen, 2013; Palermo & Eccleston, 2009). In line with family systems theory, it is thought that parent mental health is both *influenced* by youth physical and psychosocial functioning, and *influences* youth physical and psychosocial functioning. Parents of youth with chronic conditions face unique stressors, such as reduced social contact, managing complex treatments, and maintaining regular and lengthy medical appointments. Both parents of youth with chronic pain and parents of youth with SB regularly experience these and other emotionally taxing stressors (Palermo et al., 2014; Holmbeck et al., 1997), thereby increasing their risk for poor psychosocial adjustment (Cohn et al., 2020; Palermo & Eccleston, 2009). Indeed, parents of youth with chronic pain frequently report higher rates of depression, anxiety, somatic symptoms, and greater overall emotional distress (Palermo et al., 2014; Palermo & Eccleston, 2009). Similarly, research has also found that parents of youth with SB are at risk for internalizing symptoms and higher overall levels of psychological distress (Holmbeck et al., 1997; Vermaes et al., 2005).

Conversely, parent mental health also impacts youth physical and psychosocial outcomes. Indeed, reduced parent mental health has been associated with poor health outcomes in both TD youth and youth with chronic conditions; parent psychosocial adjustment is associated with poor glycemic control in youth with Type 1 Diabetes (Cunningham et al., 2011), increased BMI in TD youth (Hooper et al., 2010), and increased pain-related disability in youth with sickle cell disease (Sil et al., 2016). For youth with chronic pain, poor parent psychosocial adjustment has been

associated with higher pain intensity (Ross et al., 1993), greater pain-related disability (Logan & Scharff, 2005) and poor youth psychosocial adjustment (Lewandowski & Palermo, 2009). For youth with SB, reduced parent psychosocial adjustment has also been indirectly associated with reduced medical responsibility (Driscoll et al., 2020), and directly associated with poor youth psychological adjustment (Friedman et al., 2004). In spite of these bi-directional findings, no study to date has examined bi-directional, longitudinal associations between youth pain symptoms and parent mental health in SB within the same study.

It is also important to examine bi-directional associations between pediatric pain and broader family-level factors, such as family cohesion and conflict. Similar to parent mental health, family functioning is both thought to increase the risk of developing and exacerbating pediatric pain and is thought to be negatively impacted by the presence of pediatric pain (Palermo & Holley, 2013). Current research has typically examined group differences between youth with chronic pain and TD youth with regard to several domains of family functioning. Research has indicated that maladaptive family interaction styles and dysfunctional role assignment are common in families of youth with chronic pain (Palermo & Chambers, 2005; Palermo et al., 2014). Such families tend to be less organized, have more conflict, and are less cohesive (Lewandowski et al., 2010). Impaired family functioning has been found across multiple pain populations, including headache (Antilla et al., 2004), abdominal pain (Liakopoulou-Karis et al., 2002), fibromyalgia, and arthritis (Conte et al., 2003). However, it is often difficult to determine the directionality of these findings due to the use of cross-sectional designs; few studies have examined these variables longitudinally. In this limited literature, one

study found that parent but not youth-reported family dysfunction predicted increased pain intensity over a seven-day period in youth with inflammatory bowel disease (Caes et al., 2019).

However, there have been some mixed findings as well. In a systematic review of family functioning in pediatric pain populations, Lewandowski and colleagues (2010) reported that several studies found youth pain symptoms to be associated with positive family functioning. Similarly, Miró and colleagues (2019) found that, for younger children with disabilities and chronic pain, perceived family support was associated with increased pain catastrophizing, a maladaptive coping style often associated with increased pain levels. Further, pediatric research has frequently demonstrated the negative impact of solicitous and protective parenting behaviors; parents may believe these behaviors are a form of family/social support but they are ultimately quite detrimental (Palermo & Chambers, 2005). These parenting behaviors often positively reinforce activity restriction, give attention to youth pain symptoms, and have been associated with increased pain and pain-related disability (Claar et al., 2008). Therefore, it appears that pain does not have an exclusively positive or negative effect on family functioning; instead, it has been hypothesized that pediatric pain can bring families together and bolster cohesion, as well as place increased stress on a family, thereby increasing family conflict (Lewandowski et al., 2010). Further, aspects of family/social support can result in solicitous behaviors thereby increasing youth pain symptoms. These mixed findings underscore the importance of using a longitudinal design to parse apart the bi-directional relations between different family factors and pain to aid in the development of efficacious family interventions.

With regard to the family environment in SB, the current literature has produced similarly complex findings. Research has generally supported a resilience-disruption model (Costigan et

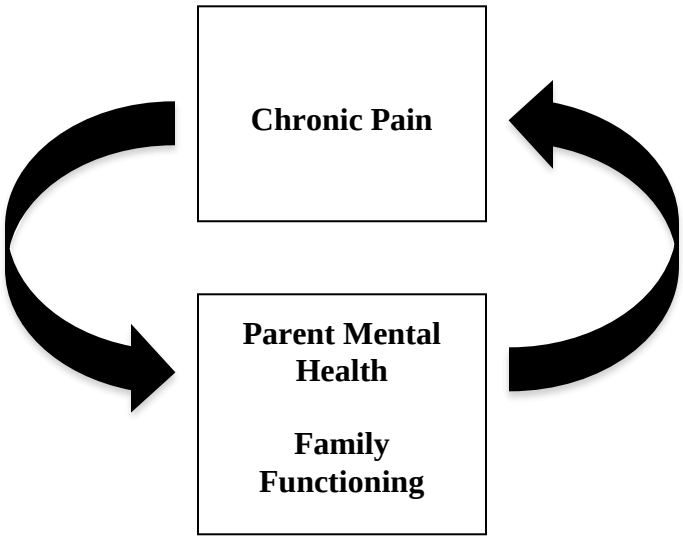
al., 1997; Holmbeck & Devine, 2010), in which families with SB have been found to be more vulnerable in certain areas of family functioning, and also display notable resilience in the face of significant stressors. For example, research suggests that families with SB display less cohesion in pre-adolescence compared to families with TD youth (Lennon et al., 2015). However, research has also highlighted areas of strength later in development; indeed, families with youth with SB display less conflict and more cohesion during adolescence compared to families with TD youth (Jandasek et al., 2009; Lennon et al., 2015).

Current Study

Given the findings from both the chronic pain and spina bifida literature, one might expect chronic pain to potentially represent a further disruption to the family unit or an added stressor to parent psychosocial functioning. Conversely, it may represent a maladaptive youth stress response to an already disrupted family environment or existing parent mental health problems. Further, it may represent an area of potential resilience, in which there is greater cohesion in the face of an added stressor. Finally, it could function differently at different developmental stages (middle childhood, early adolescence, and later adolescence). To our knowledge, there is currently no research examining relations between pain and parent mental health, or pain and family functioning in youth with SB. As demonstrated above, this gap in the literature is problematic given: (1) the prevalence of pain in SB, (2) strong bi-directional associations between pain and reduced parent mental health and family functioning in youth with chronic pain, and (3) the fact that the current literature has shown that families and parents of youth with SB may be vulnerable to family dysfunction and reduced parent mental health. Therefore, this study aimed to address this gap in the literature by examining bi-directional,

longitudinal relations between youth chronic pain and, parent mental health and family functioning (see Figure 4). This study is based on four hypotheses to examine these bi-directional associations: (1) Youth chronic pain status will be associated with reduced parent mental health (increased depressive symptoms, anxiety symptoms, and somatization), (2) Reduced parent mental health will, in turn, be associated with youth chronic pain status, (3) Youth chronic pain status will be associated with reduced family functioning (lower cohesion, increased conflict), and (4) Reduced family functioning, in turn, will be associated with youth chronic pain status. These hypotheses will be examined longitudinally (e.g., over a two-year period), enabling us to understand potential temporal relations between these variables. The longitudinal design will also allow us to determine whether pain in SB functions similarly to other pediatric pain populations wherein there is a bi-directional relationship between pain and parent/family functioning.

Figure 4. Bi-Directional Relations between Chronic Pain and Parent and Family Functioning



Methods

Participants

Participants were recruited from an ongoing longitudinal study examining family and peer relationships, neuropsychological functioning, and psychological adjustment in youth with SB (e.g., see Psihogios & Holmbeck, 2013). The current study examined parent and family functioning and pain symptoms at Time 1 (ages 8-15) and Time 2 (ages 10-17). Families of children with SB were recruited through four hospitals and a statewide SB association. Recruitment was conducted in person at regularly scheduled clinic visits or through recruitment letters. Research assistants assessed (over the phone) whether interested families met the inclusion criteria: (1) a diagnosis of SB (types included myelomeningocele, lipomeningocele, and myelocystocele); (2) age 8-15 years; (3) proficiency in English or Spanish; (4) involvement of at least one primary caregiver; and (5) residence within 300 miles of the laboratory (to allow for data collection at participants' homes). A total of 246 families were approached throughout recruitment and a total of 163 agreed to participate. However, 21 of the 163 families could not be contacted or later declined to participate, and two families did not meet inclusion criteria. The final sample included 140 families of children with SB at Time 1 (53.6% female, M_{age} = 11.40; see Table 1). Youth of families who declined to participate did not differ from participants with respect to type of SB (myelomeningocele or other), χ^2 (1) = .0002, $p > .05$, shunt status, χ^2 (1) = .003, $p > .05$, or occurrence of shunt infections, χ^2 (1) = 1.08, $p > .05$). Data were collected at Time 2 for 111 (79%) of the original 140 participants. Reasons for attrition at Time 2 (N=29) were as follows: 16 participants declined to participate, 12 participants were unable to be contacted, and 1 participant was deceased. Youth of families who did not participate at Time 2

did not differ from participants with respect to gender χ^2 (1)=0.28, p>.05, socioeconomic status (SES), t(128)=-1.86, p>.05, type of SB (myelomeningocele or other), χ^2 (1)=1.19, p>.05, lesion level (thoracic or other), χ^2 (1)=0.72, p>.05, or shunt status, χ^2 (1)=2.73, p>.05.

Table 5. Youth Demographic and Spina Bifida Information at Time 1

	Total M (SD) or N (%)
Participants	140 (100%)
Age	11.43 (2.46)
Gender: female	75 (53.6%)
Race	
Caucasian	74 (52.9%)
African American/Black	19 (13.6%)
Hispanic/Latino	39 (27.9%)
Asian	2 (1.4%)
Bi-racial	6 (4.3%)
Spina bifida type	
Myelomeningocele	122 (87.1%)
Non-myelomeningocele	17 (12.1%)
Unknown/not reported	1 (0.7%)
Lesion level	
Thoracic	23 (16.4%)
Lumbar	69 (49.3%)
Sacral	41 (29.3%)
Unknown/not reported	7 (5.0%)
Shunt: present	109 (77.9%)
Family SES	39.12 (16.09)

Procedure

University and hospital Institutional Review Boards approved the current study. Trained undergraduate and graduate student research assistants collected data during three-hour long home visits; at Time 1, two separate three-hour home visits were conducted and at Time 2 one three-hour home visit was conducted. Prior to data collection, informed consent and assent were

obtained from parents and children, respectively. Release of information forms were also gathered from parents to permit data collections from medical charts, health professionals, and teachers. One or more Spanish-speaking research assistants were present at home visits in which parents primarily spoke Spanish. Questionnaires were offered in both English and Spanish; members of the research team, who were native Spanish speakers, translated and back translated questionnaires that were only available in English. When requested or when youth appeared to have reading difficulties, research assistants read questionnaires aloud to participants. Family members completed questionnaires independently. Questionnaires were counterbalanced prior to mailing and/or being presented to families to avoid order effects. Parents and children participated in four videotaped interaction tasks, the order of which were randomly assigned. Tasks included: (1) an interactive game, (2) discussion of two age-appropriate socially-relevant vignettes, (3) discussion of transferring disease-specific responsibilities from parent to child, and (4) discussion of three conflict issues; the family chose these three issues from five possible conflict areas that they reported to be frequently occurring on a parent-child conflict questionnaire, which is described in further detail below. The current study used youth and parent questionnaire data and family observation data. At each time point, families were compensated for their participation with small gifts (e.g., logo t-shirts, pens, water bottles) and $150.

Measures

Demographics. At Time 1, parents reported on family and youth demographic information (e.g., age, gender, race/ethnicity, income, education, and employment). In accordance with the Hollingshead Four Factor Index of Socioeconomic Status (SES), SES was

calculated based on parent's education and occupation, with higher scores indicating higher SES (Hollingshead, 1975).

Illness severity. Data regarding illness severity was gathered from questionnaire data and medical charts. At Time 1, parents completed the Medical History Questionnaire (MHQ; Holmbeck et al., 2003). This questionnaire assesses youth's disease-specific medical information including bowel and bladder functioning, ambulation method (i.e., ankle-foot orthoses [AFOs], knee-ankle-foot orthoses [KAFOs], hip-knee-ankle-foot orthoses [HKAFOs], wheelchair, or no assistance), medications, frequency of medical care, and surgery history. Data were also collected from medical charts, including information regarding shunt status, type of SB (i.e., lipomeningocele, meningocele, or myelomeningocele), and lesion level (i.e., sacral, lumbar, or thoracic). An illness severity index was then computed based on membership in specific groups: shunt status (no = 1, yes = 2), myelomeningocele (no = 1, yes = 2), lesion level (sacral = 1, lumbar = 2, thoracic = 3), and ambulation status (no assistance/AFOs = 1, KAFOs/HKAFOs = 2, wheelchair = 3). Higher illness severity scores indicate higher levels of severity (range =4-10; Hommeyer et al., 1999).

Parent mental health. Parent mental health was assessed using the Symptom Checklist-Revised (SCL-90-R), a broad-based measure of psychosocial functioning. The SCL-R is a 90-item measure, with a five-point rating scale, ranging from 0=not at all, to 4=extremely. This measure includes nine symptom subscales and an overall severity index (Global Severity Index, GSI). Results from validation studies for the SCL-90-R have indicated that individual subscales demonstrate acceptable levels of internal consistency (e.g., Depression: α=.90; Derogatis, Rickels, & Rock, 1976). An "internalizing subscale" was created for both reporters using items

from the SCL's Depression, Anxiety and Somatization subscales. Scale reliability was quite strong for both the father internalizing (T1: α=.81, T2: α=.87), mother internalizing (T1: α =.88, T2: α =.87) subscales.

Pain symptoms. Pain frequency was measured on a 6-point Likert-type scale, ranging from '0-less than once a month' to '5-daily' (Palermo et al., 2004). A dichotomous pain chronicity variable was created for this study, in which participants were either categorized as having chronic pain or not having chronic pain. Youth were considered to have chronic pain if they reported experiencing pain one or more times per week over the past 3 months (King et al., 2011).

Family functioning. Family functioning was measured using both questionnaire and observational measures.

Questionnaire measures of family functioning. Parents and youth reported on the Parent Adolescent Conflict Scale (PAC), a brief version of the Issues Checklist (Prinz et al., 1979). The PAC lists 15 potential non-medical conflicts that are commonly found in families with a child close to or in the adolescent developmental period. It also includes 10 medical conflict issues that are likely to arise in families with a child with SB. Each item requires three responses; first the respondent indicates whether or not the issue was discussed during the last two weeks (yes or no response), if "yes" the respondent then indicates the number of times it was discussed and the average intensity of the discussions (using a 5-point Likert scale, ranging from "calm" to "angry"). The PAC has demonstrated strong internal consistency in previous studies in this population (Coakley et al., 2002). This study used the conflict intensity variable across both

medical and non-medical conflict items. The conflict scale demonstrated strong internal consistency in this study across all three reporters and time points (α=.79-.96).

Parents also completed the Family Environment Scale (FES; Moos & Moos, 1994), which requires the respondent to answer questions about the social-environmental characteristics of their family using a four-point Likert scale (i.e., 1=Strongly Disagree, 4=Strongly Agree). This study used two of this measure's ten subscales, cohesion and conflict. The FES-R cohesion scale demonstrated adequate internal consistency for both Time 1 and Time 2 and for both mother and father report (α=.60-.72). Similarly, the conflict scale demonstrated satisfactory internal consistency across time points and reporters (α=.69-.79).

Observational measure of family functioning. Observational data were coded using a macro-coding system developed by Holmbeck et al. (2007). This system was adapted from a methodology established by Smetana et al., 1991 (see Holmbeck et al., 2002 for a detailed description). Graduate and undergraduate research assistants were trained for approximately 8-10 hours, and until they reached 90% agreement with ratings on previous videos coded by "expert coders." "Reliable coders" then independently viewed and separately coded four videotaped family interaction tasks; coders used a 5-point Likert scale to provide ratings on dimensions of several family behaviors, including behaviors related to family cohesion and conflict. Composite scores for family cohesion and conflict were created by calculating the mean scores across two coders and four tasks for items included in those scales. Items listed below may reflect observation of one family member, both parents, each individual family member (including the child), or it may reflect family-level functioning (e.g., "family is able to reach agreement/resolution). Family cohesion included the following items: involvement in the task

(child and parent[s]), requests input from other family members (child and parent[s]), parents present a united front (parents), parental promotion of dialogue and collaboration (parent[s]), disengaged (family; *reverse-coded*), openness/warmth (family), and family is able to reach agreement/resolution (family). Family conflict included the following items: levels of conflict observed between mother and child, father and child, and mother and father, and attempted resolution of issues (child and parent[s]*; reverse-coded*). When only one parent was present, this code only included level of conflict between mother/father and child, and attempted resolution of issues (*reverse-coded*). At both Time 1 and Time 2, the FIMS cohesion scale demonstrated acceptable scale reliability scores (T1: $\alpha = .90$; T2: $\alpha = .88$) and interrater reliability coefficients (ICCs =.78-83). The FIMS conflict scale similarly demonstrated acceptable scale reliability (T1: $\alpha = .66$; T2: $\alpha = .78$) and interrater reliability coefficients (ICCs=.62-.68).

Statistical Treatment

Prior to testing the hypotheses, the psychometric properties of all measures were evaluated. Descriptive statistics were computed for all measures to assess for skewness and outliers and to evaluate assumptions and missing data. To conserve power and reduce the potential number of analyses, data transformation techniques were used when appropriate. Associations between measures in which there were two reporters (e.g., mother-report, father-report) or methodologies (e.g., Family Environment Scale Cohesion and Observed Cohesion) were calculated using Pearson correlation coefficients; associations between three or more informants (youth, mother, father) or methodologies were calculated using Cronbach's alpha coefficients (PAC conflict, Observed conflict, FES conflict). Criteria of $r \geq .40$ and $\alpha > .60$ were

used, respectively, to determine which measures could be collapsed across reporters or methodologies (Holmbeck et al., 2002).

With regard to the parent mental health variables, mother and father report were not adequately correlated, and therefore needed to be examined separately. However, for each reporter (i.e., mother and father) the subscales of somatization, depression, and anxiety were strongly associated ($\alpha > .60$), therefore, we created "mother internalizing" and "father internalizing" subscales, which included items across the somatization, depression, and anxiety subscales. With regard to family environment variables, mother and father report of conflict on the PAC and mother and father report of conflict on the FES demonstrated adequate internal consistency ($\alpha > .60$); therefore a "Parent reported conflict" scale was created across both of these measures and reporters. Child report of conflict on the PAC and observed conflict were not significantly associated with the mother or father report on the PAC or FES and were therefore examined in separate regressions. Mother and father report of cohesion on the FES were adequately correlated and therefore were combined to create a "Parent reported cohesion" subscale. Observed cohesion was not adequately correlated with mother or father report of cohesion, and therefore was examined separately.

This study examined relations between chronic pain status (see description above) and parent/family functioning longitudinally (Times 1 and 2) and used a series of hierarchical multiple linear and logistic regression analyses to examine associations between (1) family and parent functioning at Time 1 and chronic pain status at Time 2, and (2) chronic pain status at Time 1 and family and parent functioning at Time 2. Logistic regression was used when pain chronicity was the outcome. Previous findings regarding pain and family functioning guided the

choice of covariates for this study; covariates included child age and gender (King et al., 2011), SES (Dorner et al., 2011; Schreier & Chen, 2013), and illness severity (Hommeyer et al., 1999). Separate regressions were conducted for each combination of predictor and outcome variables. Assuming a power of .80, and an alpha of .05, a sample size of 43 is required to detect large effect sizes, a sample size of 92 is required to detect medium effect sizes, and a sample of 647 is required to detect small effect sizes (Cohen, 1992). Thus, the current study had enough power to detect medium or large effect sizes.

Results

Preliminary Analyses

All variables were examined for outliers and skewness. Variables were considered skewed if skewness values were >1.0 (Tabachnick & Fidell, 2007). No outliers were identified; however, results revealed six study variables to be positively skewed. These included: child report of conflict on PAC at Time 2 (skewness value=7.096), mother report of PAC at Time 2 (skewness value= 1.03), mother report of internalizing symptoms on the SCL-90 at Time 1 (skewness value= 2.27) and Time 2 (skewness value =2.66), and father report of internalizing symptoms on the SCL-90 at Time 1 (skewness value=1.77) and at Time 2 (skewness value=3.46). With one exception, all skewed variables were transformed using a square root transformation. Child report of conflict on PAC was transformed using a natural log transformation.

Objective 1: Longitudinal, bidirectional associations between chronic pain and parent mental health. Results showed no significant relations between youth chronic pain at Time 1 and mother or father internalizing symptoms at Time 2. Further, there were also no

significant relations between mother or father internalizing symptoms at Time 1 and youth chronic pain at Time 2.

Objective 2: Longitudinal, bidirectional associations between pain and family functioning. Results from hierarchical logistic and linear regressions indicated significant bi-directional longitudinal associations between youth chronic pain and family functioning. Specifically, after entering the covariates, chronic pain status at Time 1 was found to significantly predict child reported family conflict at Time 2 (β=-.25, p<.01, ΔR^2=.13; see Table 6), such that having chronic pain was associated with a decrease in family conflict two years later. However, chronic pain did not significantly predict parent reported or observed family conflict (see Table 6).

Table 6. Longitudinal Hierarchical Linear Regression Analyses for Covariates and Pain Chronicity at Time 1 Predicting Child, Parent, and Observed Family Conflict at Time 2

Child-Reported Conflict			
Predictor	b	β	ΔR^2
Child Reported Conflict (T1)	.04	.08	.01
Youth Age	.01	.10	.04
SES	.00	-.02	.04
Gender	.01	.02	.04
Illness Severity	.02	.12	.04
Chronic Pain (T1)	**-.18**	**-.30**	**.08****
Parent-Reported Conflict			
Parent Reported Conflict (T1)	.57	.59	.35**
Youth Age	.01	.04	.01
SES	.00	-.01	.01
Gender	.14	.10	.01
Illness Severity	-.01	-.01	.01
Chronic Pain	-.12	-.07	.00
Observed Conflict			
Observed Conflict (T1)	.11	.27	.07*
Youth Age	-.01	-.14	.07
SES	.00	-.19	.07
Gender	-.01	-.02	.07
Illness Severity	.02	.13	.07
Chronic Pain	.02	.05	.07

Note: All predictor variables were measured at Time 1. The covariates of youth age, gender, SES, and illness severity were entered in a block in enter selection. Youth, parent, or observed conflict at Time 1 was entered at Step 1. *$p < .05$, ** $p < .01$

When examining the alternative direction, results revealed that parent reported family conflict at Time 1 significantly predicted chronic pain group status at Time 2 (Exp(b)=2.98, Wald=4.34, $p<.05$; see Table 7), such that increased family conflict increased the odds of being in the chronic pain group two years later (see Table 7). Child reported conflict and observed family conflict were not significantly associated with chronic pain group status (see Table 7).

Table 7. Longitudinal Hierarchical Logistic Regression Analyses for Covariates and Observed, Parent, and Child Reported Conflict at Time 1 Predicting Chronic Pain Group Membership at Time 2

Conflict Predictors			
Predictor	b	Wald	Exp(b)
Chronic Pain (T1)	.38	.30	1.46
Youth Age	.36	5.91	1.43*
SES	.01	.27	1.01
Gender	.31	.20	1.37
Illness Severity	-.22	1.06	.81
Observed Family Conflict (T1)	-.46	.42	.63
Parent Reported Conflict (T1)	**1.06**	**4.34**	**2.98***
Child Reported Conflict (T1)	-.71	1.14	.49

Note: All predictor variables were measured at Time 1. The covariates of youth age, gender, SES, and illness severity were entered in a block in enter selection. Chronic pain at Time 1 was entered at Step 1. *$p < .05$, ** $p < .01$

With regard to family cohesion, chronic pain status at Time 1 was significantly associated with parent-reported family cohesion at Time 2 ($\beta=.24$, $p<.01$, $\Delta R^2=.05$; see Table 8), such that chronic pain was associated with increased parent reported family cohesion at Time 2. No significant associations between chronic pain and observed cohesion were found (see Table 8). When examining the alternative direction, no significant associations were found between parent reported or observed cohesion and youth chronic pain (see Table 9). Finally, it should be noted that across all logistic regressions, age emerged as a significant predictor, in which older age increased the likelihood of being in the chronic pain group two years later (see Table 8 and Table 9). This finding is in line with previous research that used this study sample (Ohanian et al., 2020).

Table 8. Longitudinal Hierarchical Linear Regression Analyses for Covariates and Pain Chronicity at Time 1 Predicting Parent and Observed Cohesion at Time 2

Predictor	b	β	ΔR^2
Parent-Reported Cohesion			
Parent-Reported Cohesion	.68	.65	.35**
Youth Age	-.02	-.12	.04
SES	.00	.06	.04
Gender	-.07	-.11	.04
Illness Severity	-.02	-.11	.04
Chronic Pain (T1)	**.18**	**.24**	**.05****
Observed Cohesion			
Observed Family Cohesion (T1)	.35	.38	.15**
Youth Age	.00	.01	.07
SES	.01	.20	.07
Gender	.04	.06	.07
Illness Severity	-.05	-.20	.07
Chronic Pain (T1)	.10	.10	.01

Note: All predictor variables were measured at Time 1. The covariates of youth age, gender, SES, and illness severity were entered in a block in enter selection. Parent or observed cohesion at Time 1 was entered at Step 1. *$p < .05$, ** $p < .01$

Table 9. Longitudinal Hierarchical Logistic Regression Analyses for Covariates and Observed and Parent Reported Cohesion at Time 1 Predicting Chronic Pain Group Membership at Time 2

Predictor	b	Wald	Exp(b)
Chronic Pain (T1)	.53	.57	1.70
Youth Age	.41	8.40	1.51**
SES	.01	.44	1.01
Gender	.51	.63	1.66
Illness Severity	-.29	2.39	.75
Observed Family Cohesion (T1)	.55	.42	1.73
Parent Reported Cohesion (T1)	-.50	.28	.61

Note: All predictor variables were measured at Time 1. The covariates of youth age, gender, SES, and illness severity were entered in a block in enter selection. Chronic pain at Time 1 was entered at Step 1. *$p < .05$, ** $p < .01$

Discussion

The purpose of this study was to explore the relationship between chronic pain and family factors in youth with SB. Specifically, this study examined longitudinal associations between youth chronic pain status and parent mental health, as well as between youth chronic pain status and family environment (i.e., cohesion and conflict). To our knowledge no study has

examined relations between chronic pain and these family factors in youth with SB. Therefore, given the demonstrated vulnerabilities in parent mental health and family functioning in both youth with SB and youth with chronic pain, this paper sought to gain an understanding of how pain functions in this population in relation to family factors so as to inform future clinical interventions.

With regard to Objective 1, the study hypothesis was not supported, as results revealed no significant associations between youth chronic pain status and mother or father internalizing symptoms. This may indicate that, while having a child with SB may be associated with reduced parent mental health (Vermaes et al., 2005), the added factor of chronic pain does not significantly affect their psychological functioning. Spina bifida is a complex and demanding condition; therefore, in terms of stressors, youth chronic pain may not be as salient as other condition stressors (e.g., ambulation concerns, catheterization, bowel program, possible shunt revisions). Further, previous research, using the same study sample, found that lower illness severity at Time 1 was associated with higher pain intensity at Time 2 (Ohanian et al., 2020), potentially indicating that youth with greater illness severity are not experiencing or not reporting as much as pain as those with lower illness severity. As such, parents whose children have chronic pain may experience fewer stressors if their child is overall experiencing less illness severity. Further, this study did not find that reduced parent mental health led to an increase in youth chronic pain. Therefore, while parent mental health may affect other domains of functioning in youth with SB, such as youth psychological adjustment (Friedman et al., 2004), it does not appear to be expressed as a propensity toward recurrent pain. Given previous findings that have found child internalizing symptoms to precede pain in this population (Ohanian et al.,

2020), future studies might explore whether youth psychological functioning indirectly mediates the relationship between parent mental health and youth chronic pain.

Regarding relationships between pain and family environment, results revealed several significant associations between both family conflict and youth chronic pain, and cohesion and youth chronic pain. Youth chronic pain at Time 1 was significantly associated with reduced family conflict two years later according to youth report. On the other hand, higher family conflict per parent report at Time 1 was significantly associated with an *increased* likelihood of being in the chronic pain group two years later. While these two findings, may seem contradictory, they actually align well with previous findings regarding family environment and youth pain (Lewandowski et al., 2010). That is, these results may reflect how a tense family environment may increase the likelihood of an adolescent with SB experiencing a maladaptive stress response in the form of chronic pain. These results may also indicate that when a child with SB experiences chronic pain, the family unit may respond by attempting to reduce stressors in the home, such as reducing conflict. Indeed, this study also found that chronic pain at Time 1 was significantly associated with increased parent-reported cohesion two years later; the reverse was not found. Although, one may think that this relationship potentially indicates, that in response to youth pain, parents engaged in solicitous parenting behavior, family cohesion is considered separate construct that is comprised of several behaviors and attitudes including emotional bonding, helping each other, amount of quality time, feelings of togetherness, and group spirit (Olson et al., 1983; Moos & Moos, 1994). It should also be noted that, similar to previous studies, older age was a predictor of chronic pain group status, indicating that older adolescents whose family is experiencing significant conflict may be at an even higher risk of

developing chronic pain. Overall, these findings suggest that, in line with the resilience-disruption model (Costigan et al., 1997), when faced with the added stressor of youth chronic pain, families with youth with SB appear to display notable resilience.

In contrast, no significant associations were found between observed family environment and youth chronic pain. A previous study using this sample similarly found non-significant associations between observed parenting behaviors and SB-related medical responsibility (Driscoll et al., 2020). However, they did find significant associations between self-report of parenting behaviors and SB-related medical responsibility (Driscoll et al., 2020). These findings were in accordance with other studies that have found weak correlations between parent-reported and observed parenting behaviors (Moens et al., 2018). Further, these findings may indicate that parent and child perception of family conflict and cohesion may be more salient to the experience of youth chronic pain than the observed family environment. Given that pain is typically considered an "internal" experience and is often accompanied by internalizing symptomatology (Noel et al., 2016), perception of family environment may be more important than observed family behaviors.

This study had several strengths, including the use of multiple methods of assessing family environment and multiple reporters of both family environment and parent mental health. This study also used a longitudinal design, enabling us to examine directionality between these variables. However, this study also had several limitations. First, while having multiple reporters and methods was a strength of this study, there was often inadequate agreement among these measures and reporters, which meant that we were not able to create composite scores for all of the assessed domains. Therefore, it was necessary for us to run numerous regression analyses,

which increased the likelihood of Type 1 error. Further, chronic pain was only measured using youth report, and current guidelines for measuring pediatric pain recommend obtaining information from multiple reporters (Cohen et al., 2008). This is particularly important for youth with cognitive deficits (Breau & Burkitt, 2009), which are quite common in youth with SB. We also did not examine pain intensity, another important construct in pain research, thereby limiting our ability to obtain a comprehensive understanding of relations between parent/family functioning and pain in this population. In spite of these limitations, this was the first study to examine family and parent functioning in relation to youth chronic pain in SB, thereby addressing a significant gap in the current literature.

Conclusions and Clinical Implications

The results of the current study highlight the importance of understanding the familial context in which youth with SB experience pain. Similar to the larger pediatric pain literature, increased family conflict was associated with increased chronic pain in youth with SB over time. However, youth with chronic pain were also found to have family environments with *less* conflict and more cohesion two years later. These results represent further evidence for utilizing the resilience-disruption model in pediatric populations. Clinically-oriented literature has increasingly emphasized the importance of adopting a strengths-based vs. a deficits-based perspective, wherein identifying strengths that one can build upon for healthy functioning is just as important as identifying risks that require intervention. This study demonstrated that families with a child with SB might experience less conflict and increased cohesion when youth are experiencing chronic pain, a finding which furthers previous work that demonstrates the resilience of these families. Future research should focus on developing clinical interventions to

promote positive family functioning to support both the health of the family unit and potentially mitigate youth with SB's risk of developing chronic pain and associated psychosocial distress.

CHAPTER FOUR

PAIN CATASTROPHIZING IN ASSOCIATION WITH PAIN SEVERITY AND PAIN INTERFERENCE IN YOUNG ADULTS WITH SPINA BIFIDA: EXECUTIVE FUNCTIONING AS A MODERATOR

Introduction

Pain has long been considered a physiological, cognitive, and behavioral experience, and is one of many complications associated with spina bifida (SB; Clancy et al., 2005). SB is a common congenital birth defect and represents a prenatal neural insult. It occurs during the first month of pregnancy when the embryonic neural tube fails to close completely and is frequently accompanied by numerous physical and neurocognitive complications and psychosocial difficulties (CDC, 2011; Copp et al., 2015). In addition to pain, complications include: bowel and bladder incontinence, paralysis of the lower extremities, orthopedic conditions, seizures, hydrocephalus, and deficits in attention and executive functioning (EF; Zukerman et al., 2011; Rose & Holmbeck, 2007). As a result, the presence of both SB *and* pain is likely to impact physiological, cognitive, and behavioral functioning additively and negatively. Current research examining pain in SB has found associations between pain and internalizing symptoms (Oddson et al., 2006; Ohanian et al., 2020), quality of life (Oddson et al., 2006), and social activity and competence (Essner et al., 2014). Still, relations between pain and cognitive processes, such as neuropsychological functioning and cognitive appraisals involved in coping responses remain largely unexplored in SB. Therefore, the current study sought to gain a better understanding of

pain processes unique to SB by examining relations among pain catastrophizing, executive functioning (EF), and pain-related outcomes in young adults with SB.

From an evolutionary perspective, humans are predisposed to interpret pain as a threat (Eccleston & Crombez, 1999). This phenomenon can result in several potential outcomes related to coping, cognition, and the experience of pain itself. When one overestimates, magnifies, and ruminates on the threat of pain, one is likely to increase the potency and impact of the painful experience. In other words, engaging in pain catastrophizing often results in increased pain intensity and may increase the degree to which pain interferes with one's life (Sullivan et al., 2001; Vervoot et al., 2006). Given that catastrophizing is an inherently cognitive experience, one might also hypothesize that the impact of pain-related rumination on pain and pain interference may be amplified for those who have an inability to shift away from or inhibit catastrophic thoughts, due to executive dysfunction.

Pain catastrophizing is defined as a cognitive process that involves magnification, rumination, and feelings of helplessness in response to pain (Sullivan et al., 2001). It is considered a maladaptive coping style and has been increasingly recognized as one of the most reliable predictors of pain intensity, pain disability, and pain interference in both pediatric and adult populations (Vervoot et al., 2006; Cunningham et al., 2014; Sullivan et al., 2001; Thastum et al., 2005). Past research has suggested that pain catastrophizing and anxiety are related but distinct constructs, with pain catastrophizing being a stronger predictor of both pain intensity and pain disability (Tran et al., 2015). Indeed, pain catastrophizing has also been found to predict pain intensity and disability above and beyond certain demographic variables (e.g., sex and age) and negative affectivity (Vervoort et al., 2006). While, to our knowledge, there have been no

studies specifically examining pain catastrophizing in young adults with SB, findings from the greater disability literature suggest that these associations may also be present in SB. Specifically, in a study examining adults with spinal cord injury, greater pain catastrophizing was associated with greater pain intensity, disability, and interference in daily activities (Turner et al., 2002). Further, another study whose sample included youth with SB along with other disability populations (i.e., neuromuscular disorder and cerebral palsy) found that, when controlling for demographics and disability status, pain catastrophizing predicted pain intensity (Engel et al., 2013). Overall, pain catastrophizing has demonstrated a clear and significant impact on the experience of pain in both pediatric and adult populations, including individuals with disabilities.

Executive functions (EF) have also been found to be associated with pain processes in several studies (Moriarty et al., 2000; Weiss et al., 2017). EF is defined as higher order attentional processes, such as inhibition, shifting, and updating (Miyake et al., 2000). Notably, fMRI studies have found that both pain modulation and EF occur in the same regions of the brain (Apkarian et al., 2009; Seminowicz & Davis, 2007). Research has found pain to be negatively associated with inhibitory control (i.e., the ability to control one's attention, behavior, thoughts and/or emotions to override internal predisposition; Diamond, 2013; Legrain et al., 2009) and shifting abilities, or the ability to move back and forth between tasks (Berryman et al., 2014).

Further, stronger EF abilities are associated with increased use of adaptive pain-related coping skills (Hocking et al., 2011). Indeed, one study found that if an individual successfully employs EF while experiencing pain, they are more capable of utilizing positive pain coping strategies and they report less pain-related distress (Verhoeven et al., 2014). Relatedly, Compas

and Boyer (2001) suggested that poor attentional control impedes one's ability to disengage from their condition symptoms. This hypothesis was supported in a study examining relations between pain catastrophizing and EF in a non-clinical sample of older adolescents (ages 16-19; Bell et al., 2018). Specifically, in this study, pain rumination was associated with problems with attentional disengagement, or shifting. Further, pain magnification was associated with poor inhibitory control (Bell et al., 2018). Overall, these findings suggest that when one can shift their attention or inhibit competing attentional demands (i.e., pain symptoms), they can disengage from the pain, and consequently they will cope with pain more adaptively and the experience of the pain will thus be less bothersome.

The population of interest in this study, spina bifida, is characterized by a unique neuropsychological profile, which often includes deficits in attention and EF (Rose & Holmbeck, 2007). Youth with SB have demonstrated difficulties with focusing and shifting attention and orienting toward the most salient stimuli (Burmeister et al., 2005; Dennis et al., 2006; Brewer et al., 2001; Rose & Holmbeck, 2007). Further, metacognitive skills are also often impaired in youth with SB; deficits are seen in working memory, planning and organizing, task initiation, and self-monitoring (Brown et al., 2008). While relations among pain, pain catastrophizing, and EF have yet to be tested in this population, literature examining these constructs in other populations with cognitive deficits sheds light on potential associations among these constructs in SB.

Youth with cognitive impairments may be particularly vulnerable to poor pain outcomes. Low Kapalu and colleagues (2018) suggested that cognitive difficulties, such as executive dysfunction, might represent a significant psychosocial stressor, which can predispose one to the

emergence of chronic pain, similarly to how other stressors have been linked to the emergence of pain. Indeed, youth with cognitive impairments may experience pain more frequently than their typically-developing peers, with one study estimating that 35-52% of children and adolescents (ages 3-18) with a seizure disorder or cerebral palsy and associated severe cognitive impairments experience pain at least one time per week over a four-week period (Breau et al., 2003). Moreover, a recent study found that reduced intellectual functioning in SB was associated with pain chronicity (Ohanian et al., 2020). This finding is noteworthy, as reduced intellectual functioning is not commonly associated with chronic pain (Ho et al., 2009). Therefore, executive dysfunction could underlie the aforementioned findings.

Relations among EF, pain, and pain coping have been found in other populations characterized by executive dysfunction. In a sample of youth with cerebral palsy, EF was found to be significantly associated with pain interference (Tervo et al., 2006). Further, another study found that EF in youth with sickle cell disease, a population that also commonly experiences EF deficits (Schatz et al., 2004), was associated with increased maladaptive pain coping, including a propensity to engage in pain catastrophizing (Ludwig et al., 2018). In the SB literature, an EF intervention study for young adults with SB, found that bolstering EF abilities supported the use of adaptive coping strategies (Stubberud et al., 2015). While this study did not examine pain coping or catastrophizing, it indicates that EF may be important to consider with regard to coping more generally in this population, and indicates that relations among EF, pain catastrophizing, and pain warrant further examination in SB.

Finally, it is important to highlight different developmental contexts in pain research. Emerging adulthood has increasingly been recognized as a distinct developmental period in

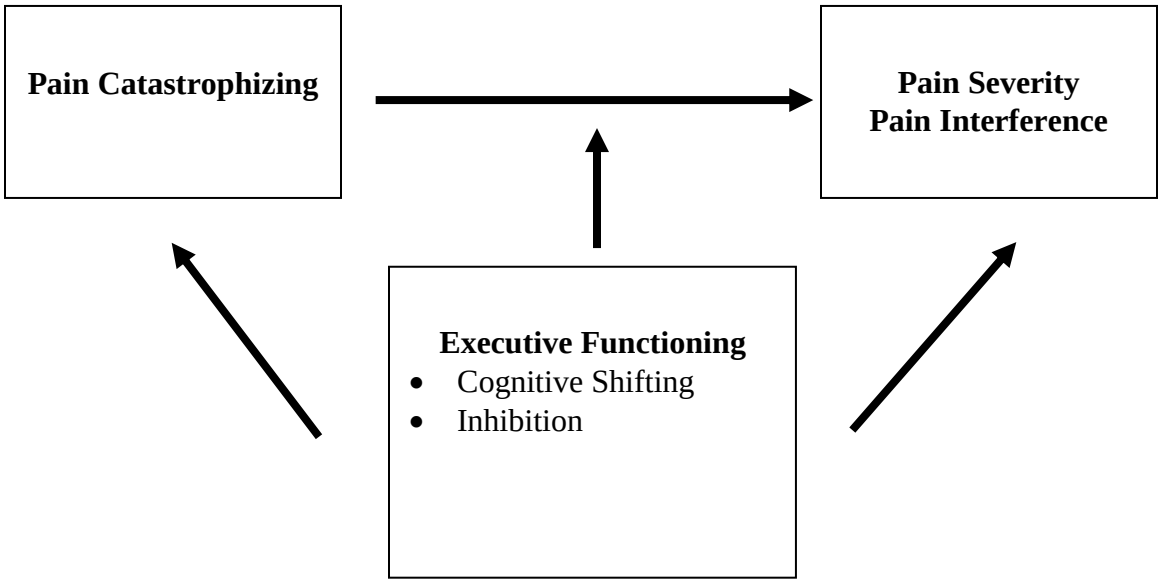
which individuals are required to progress through multiple life transitions and become increasingly independent. Chronic pain may pose a threat to successfully transitioning to adulthood, as it can interfere with educational and vocational attainment, as well as social functioning (Murray et al., 2020). Indeed, a study examining the prevalence of chronic pain in Australia found that, while the prevalence of pain was higher in older adulthood, young adults experienced significantly more pain interference than those who were considered to be in middle or older adulthood (Blyth et al., 2001). There is currently a paucity of research on pain in young adults with SB. Of the few studies that have examined pain in young adults with SB, one study found 25-33% young adults with SB to report experiencing pain at least once per month (Verhoef et al., 2004). Another study found pain severity in this population to be in the moderate range (i.e., around 5 on a 1-10 scale; Bellin et al., 2010). Given the difficulties that young adults with SB face with regard to independent functioning (Zukerman et al., 2011), highlighting pain as another potential area for intervention is critical.

Current Study

The current study had four aims and three hypotheses. The four aims were as follows: (1) to describe the nature of pain catastrophizing, pain severity, and pain interference in young adults with SB, (2) to examine the relationship between pain catastrophizing and pain severity/ interference, (3) to examine the relationship between EF and pain catastrophizing, and pain severity/interference, and finally (4) to examine the potential moderating effect of EF on the relationship between pain catastrophizing and pain severity/interference. The three hypotheses were: (1) pain catastrophizing will be positively associated with pain severity and pain interference, such that greater pain catastrophizing will be associated with greater pain severity

and more pain interference, (2) greater executive dysfunction will be positively related to pain catastrophizing, severity, and interference, such that greater executive dysfunction will be associated with more pain catastrophizing, severity, and interference, and finally, (3) greater executive dysfunction will exacerbate the negative impact of pain catastrophizing on pain severity and pain interference (see Figure 5).

Figure 5. Moderating Role of Executive Functioning on the Relationship between Pain Catastrophizing and Pain Severity/Interference



Methods

Participants

This study was part of an ongoing longitudinal study examining family and peer relationships, neuropsychological functioning, and psychological adjustment in youth and young adults with spina bifida (see Devine, Holmbeck et al., 2012; Psihogios & Holmbeck, 2013). The current study examined pain, pain catastrophizing, and EF during the sixth and final time point of this longitudinal study. Young adults in the current study were originally recruited, along with

their families from four hospitals and a statewide SB association. Such recruitments were either conducted in person at regularly scheduled clinic visits or through recruitment letters. At Time 1, interested families were screened by phone or in person by a trained member of the research team to determine if the child met the following inclusion criteria: (1) a diagnosis of SB (types included myelomeningocele, lipomeningocele, and myelocystocele); (2) age 8-15 years; (3) proficiency in English or Spanish; (4) involvement of at least one primary caregiver; and (5) residence within 300 miles of the laboratory (to allow for data collection at participants' homes).

During recruitment a total of 246 families were approached, of which 163 agreed to participate. However, a subset of these families later declined to participate. Further, we were also not able to contact some of these families; therefore, 21 of the 163 families ultimately were not included in the study sample. Finally, two families did not meet inclusion criteria. The final sample included 140 families of children with SB (at Time 1, 53.6% female, M_{age} = 11.40). For this study, the participants were again recruited during young adulthood to participate in the sixth wave (T6) of the study. T6 data collection was suspended due to COVID-19; 80 participants (ages 18-26) had completed questionnaires and interviews when the analyses for this study were conducted (52.5% female, M_{age} = 22.76; Table 1). Young adults who have not yet participated in this study did not differ from participants in this study with respect to gender χ^2 (1)=0.97, p>.05, socioeconomic status (SES), t(126)=1.58 p>.05, type of SB (myelomeningocele or other), χ^2 (1)=0.49, p>.05, lesion level (thoracic or other), χ^2 (1)=0.57, p>.05, or shunt status, χ^2 (1)=0.00, p>.05.

Procedure

The current study was approved by university and hospital Institutional Review Boards. Data were collected by trained undergraduate and graduate student research assistants during home visits that lasted approximately three hours. For home visits with families who primarily spoke Spanish, at least one bilingual research assistant was always present. Prior to each home visit, informed consent from participants was obtained. Participants also completed releases of information to obtain data from medical charts and health professionals. Participants received $200 in compensation at this wave of the study.

Measures

Demographics. Parent completed a questionnaire reporting on participant and family demographic information at Time 1. This questionnaire includes information on age, gender, race/ethnicity, highest levels of education, and work status and income. The Hollingshead Four Factor Index of Socioeconomic Status (SES) was computed using parent's education and occupation from Time 1, with higher scores indicating higher SES (Hollingshead, 1975).

Illness severity. Illness severity data was extracted from questionnaire data and medical charts. At Time 1, parents completed the Medical History Questionnaire (MHQ; Holmbeck et al., 2003), which asks questions regarding youth's disease-specific medical information including bowel and bladder functioning, ambulation method (i.e., ankle-foot orthoses [AFOs], knee-ankle-foot orthoses [KAFOs], hip-knee-ankle-foot orthoses [HKAFOs], wheelchair, or no assistance), medications, frequency of medical care, and surgery history. Medical chart data was also used to assess type of SB (i.e., lipomeningocele, meningocele, or myelomeningocele), shunt status, and lesion level (i.e., sacral, lumbar, or thoracic). An illness severity index was then

computed based on membership in specific groups: shunt status (no = 1, yes = 2), myelomeningocele (no = 1, yes = 2), lesion level (sacral = 1, lumbar = 2, thoracic = 3), and ambulation status (no assistance/AFOs = 1, KAFOs/HKAFOs = 2, wheelchair = 3). The range of illness severity scores was from 4 to 10 (higher scores indicate higher levels of severity; Hommeyer et al., 1999).

Youth IQ. At Time 1, research assistants administered the Vocabulary and Matrix Reasoning subtests of the Wechsler Abbreviated Scale of Intelligence to participants (WASI; Wechsler, 1999), which yielded an estimate of the Full-Scale Intelligence Quotient (IQ) score. The 42-item vocabulary subtest measures an individual's expressive vocabulary, verbal knowledge, and fund of information. The matrix reasoning subtest assesses nonverbal fluid reasoning and general intellectual ability and is composed of 35 items. These subtests have demonstrated high levels of internal consistency for youth 6-16 years old ($\alpha = .89$ for Vocabulary, $\alpha = .92$ for Matrix Reasoning; Wechsler, 1999).

Pain catastrophizing. Participants completed The Pain Catastrophizing Scale (PCS-EN), a 13-item measure assessing catastrophic thinking in response to pain (Sullivan et al., 1995). Participants responded to 13 items on a 5-point Likert scale (0=not at all to 4=all the time), which assessed cognitive responses to past painful experiences. Sample items include “I worry all the time about whether the pain will end,” “I anxiously want the pain to go away,” and “I keep thinking about how badly I want the pain to stop.” This measure yields a total score and three subscales: rumination, magnification, and helplessness. The pain catastrophizing scale was validated on Nova Scotia workers that had submitted an injury claim (Sullivan et al., 1995). Cutoff scores were developed, wherein if an individual was in the 50^{th} -75^{th} percentile they were

considered at a moderate risk for developing chronic pain, and those whose scores were at the 75th percentile or above were considered at significant risk for developing chronic pain. The 50th percentile starts at 20 for the total score and the 75th percentile starts at 30. The subscale cutoffs at the 50th and 75th percentile are as follows, rumination=8, 11, magnification=3, 5, helplessness=8, 13. To address Aim 1, the three subscales and the total score were employed. To examine Aims 2 and 4, regression and moderation analyses only included the total score. The PCS-EN validation study indicated that the measure demonstrated adequate internal consistency for both the total score (α=.87) and the three individual subscales, with alphas ranging .66-.87 (Sullivan et al., 1995). In this study the total catastrophizing score (α=.96), and the subscales of rumination (α=.95), magnification (α=.88), and helplessness (α=.91), all demonstrated strong internal consistency.

Executive functioning. Participants completed the Behavior Rating Inventory of Executive Functioning-Adult Version (BRIEF-A; Gioia et al., 2000). This measure asks participants to indicate how often (Never, Sometimes, Often) 75 different behaviors have been a problem during the past month. Sample items include "I lose things," "I forget instructions easily," and "I have trouble finishing tasks." This is a valid self-report measure of executive functioning that yields eight sub-domains of executive functions: Inhibit, Shift, Emotional Control, Initiate, Working Memory, Plan/Organize, Organization of Materials, and Monitor, as well as a Global Executive Composite. In accordance with findings from literature on relations among pain, executive functioning, and pain catastrophizing, this study used two of the BRIEF subscales: *Shift* and *Inhibition*. Higher scores indicate greater executive dysfunction. The shift

(α =.77) and inhibition (α =.86) scales both demonstrated adequate internal consistency in this study.

Pain and pain interference. Participants also completed the Brief Pain Inventory (BPI; Cleeland & Ryan, 1994), which measures both pain severity and the degree to which pain interferes with individual functioning. Participants responded to nine items in total. Five items ask participants to rate their sensory pain experiences over the past 24 hours (e.g., location of pain, worst pain, least pain, average pain, current pain). The mean of four of these items are used to compute the *pain severity* scale (e.g., worst pain, least pain, average pain, and current pain). Pain severity was placed into four categories (e.g., 0.0–0.9: none/minimal, 1.0-3.9: mild, 4.0-6.9: moderate, and 7.0-10.0: severe; Breivik et al., 2008) for descriptive analyses; however, for regression analyses this variable (pain severity) was on a continuous scale. Further, participants responded to two questions regarding pain treatment (which treatments and the percentage of relief from the treatments) in the past 24 hours. Finally, the last item asks participants to indicate how much their pain has interfered with seven domains of daily activity in the past 24 hours (general activity, mood, walking ability, normal work, relations with other people, sleep, and enjoyment of life). The mean rating across these domains was used to compute the *pain interference* scale. Due to the nature of SB, we removed "walking ability" from the pain interference scale; this has been done in other studies examining pain in youth with physical disabilities, including youth with SB (see Miró et al., 2019). This study used both the *pain severity* and *pain interference* scales. The Brief Pain Inventory was originally validated in cancer patients and does not have cut off scores. However, they have suggested that "substantial pain" is at the midpoint or higher (i.e., "5"; Cleeland & Ryan, 1994); therefore, some descriptive analyses

in this study reference this category of "substantial pain." The BPI has demonstrated satisfactory internal consistency for pain severity (.81-.89) and pain interference (.88- .95); test-retest reliability was .80 for both pain severity and interference (Pouquet & Lin, 2016). In the current study the BPI demonstrated strong internal consistency for pain severity (α =.87) and pain interference (α =.95).

Statistical Treatment

Prior to hypothesis testing, the psychometric properties of all measures were calculated to examine distributional properties, identify outliers, and evaluate the degree of skewness. Further descriptive statistics were computed to characterize levels of pain, pain interference, and pain catastrophizing. These statistics were also used to address Aim 1 of the current study. Aim 2 of this study was addressed using cross-sectional hierarchical multiple linear regression analyses to examine associations between pain catastrophizing and pain severity as well as associations between pain catastrophizing and pain interference. Aim 3 similarly was addressed using cross-sectional hierarchical multiple linear regression analyses to examine associations between EF and pain catastrophizing, severity, and interference. Several covariates were chosen for this study that have been considered particularly relevant in the pain and SB literature; these included age and gender (King et al., 2011), SES (Dorner et al., 2011; Schreier & Chen, 2013), and illness severity (Hommeyer et al., 1999). Separate regressions were conducted for each combination of predictor and outcome variables; covariates were entered in block 1 followed by the predictor variable (i.e., pain catastrophizing or EF). Finally, to address Aim 4, two-way interactions between pain catastrophizing and EF were computed and examined as predictors of pain severity/interference using the Hayes PROCESS Macro. In this model, executive functions

(inhibition, shifting) served as moderators. Assuming a power of .80, and an alpha of .05, a sample size of 36 is required to detect large effect sizes, a sample size of 77 is required to detect medium effect sizes, and a sample of 550 is required to detect small effect sizes (Cohen, 1992). Thus, the current study had enough power to detect medium or large effect sizes.

Results

Preliminary Analyses

Study variables were examined for skewness and outliers. Using the West et al. (1995) criteria for skewness for smaller samples (values> 2.0), none of the variables were considered significantly skewed. Further, no outliers were identified. Given concerns about the potential cognitive capacity needed to complete the study questionnaires, distributional properties were examined in those participants with an IQ <70 (N=12) and no outliers were identified. Therefore, these participants were included in study analyses.

The total study sample included 80 young adults with SB ages 18-25 (52.5% female; 61.3% Caucasian; see Table 10). This sample predominately had myelomeningocele SB (86.1%) and a shunt (78.5%), and about half of the sample had a Lumbar lesion (48.1%). Mean intellectual functioning fell in the low average range (M=87.78, SD=19.35). Bivariate Pearson correlation coefficients revealed significant associations between gender and pain interference (r=.26, p<.05), and gender and pain catastrophizing (r=.24, p<.05), such that female gender was associated with more pain interference and increased pain catastrophizing. Lesion level, shunt status, IQ, illness severity, age, race/ethnicity were not significantly associated with pain severity, interference or pain catastrophizing.

Table 10. Youth Demographic and Spina Bifida Information at Time 6

	Total M (SD) or N (%)
Participants	80 (100%)
Age	22.76 (2.47)
Gender: female	42 (52.5%)
Race	
Caucasian	49 (61.3%)
African American/Black	11 (13.8%)
Hispanic/Latino	16 (20.0%)
Asian	1 (1.3%)
Bi-racial	3 (3.8%)
Spina bifida type	
Myelomeningocele	68 (86.1%)
Non-myelomeningocele	11 (13.9%)
Unknown/not reported	1 (0.7%)
Lesion level	
Thoracic	14 (17.7%)
Lumbar	38 (48.1%)
Sacral	25 (31.6%)
Unknown/not reported	2 (2.5%)
Shunt: present	62 (78.5%)
IQ	87.78 (19.34)
Family SES	41.02 (15.45)

Objective 1: Descriptive analyses. Using the Brief Pain inventory severity scale, 26.4% (N=18) of young adults reported moderate or severe levels of pain. Participants predominately reported mild to moderate levels of pain interference across most domains, with mood being the most affected (M=2.73, SD=3.05; see Table 11). For young adults who reported substantial pain severity (at the midpoint of scale [5] or higher), 70% (N=6) of them reported a moderate to severe level of pain interference. For this subsample, pain again appeared to interfere with mood the most of any listed domain (M=6.30, SD=1.83).

With regard to catastrophizing, a small but sizable minority of participants (25.7%, N=19; see Table 11) reported moderate to high levels of pain catastrophizing. Means across the subscales of rumination, magnification, and helplessness all fell in the mild range. However, 35.6% (N=26) of respondents reported moderate to high levels of rumination, 41.0% (N=30) reported moderate to high levels of magnification, and 26.0% (N=19) reported moderate to high levels of helplessness (see Table 11).

Table 11. Descriptive Data for Pain Severity, Pain Interference, and Pain Catastrophizing

Variable		
Severity	N	%
None/Minimal	29	42.6
Mild	21	30.9
Moderate	17	25.0
Severe	1	1.5
Interference	M	SD
General Activity	2.20	2.85
Mood	2.73	3.05
Work	2.45	3.09
Relations with other people	1.48	2.61
Sleep	2.59	3.07
Enjoyment of life	2.02	2.98
Average Interference	2.24	2.62
Pain Catastrophizing	M	SD
Rumination	5.59	4.94
Magnification	2.89	3.33
Helplessness	5.59	5.78
Total Score	14.07	13.37
Pain Catastrophizing Classification	N	%
Mild	55	74.3
Moderate	7	9.5
High	12	16.2

Note. M=Mean, SD=Standard Deviation, N=sample size. Percentages reflect frequency of chosen descriptors. Pain severity is presented according to four severity categories (0.0-0.9: none/minimal, 1.0-3.9: mild, 4.0-6.9: moderate, 7.0-10: severe). Pain catastrophizing classification categories correspond to risk for pain chronicity (i.e., 16.2% of respondents catastrophizing total corresponded to a high risk of developing chronic pain).

Objective 2: Associations between pain catastrophizing and pain severity and interference. Hierarchical linear regressions revealed significant cross-sectional associations between pain catastrophizing, and pain severity and interference. With regard to pain severity, after entering the covariates greater pain catastrophizing significantly predicted greater pain severity (β=.31, $p<.05$, $\Delta R^2=.09$; see Table 12). Further, pain catastrophizing also significantly predicted increased pain interference (β=.44, $p<.01$, $\Delta R^2=.19$; see Table 12).

Table 12. Hierarchical Linear Regression Analyses for Covariates and Pain Catastrophizing Predicting Pain Severity and Pain Interference

Pain Severity			
Predictor	b	β	ΔR^2
Age	.07	.09	.05
SES	-.01	-.06	.05
Gender	.50	.12	.05
Illness Severity	-.16	-.12	.05
Pain Catastrophizing	**.33**	**.31**	**.09***
Pain Interference			
Age	.15	.15	.06
SES	-.02	-.12	.06
Gender	.38	.08	.06
Illness Severity	-.22	-.14	.06
Pain Catastrophizing	**.09**	**.44**	**.19****

Note: The covariates of youth age, gender, SES, and illness severity were entered in a block in enter selection.
*$p < .05$, ** $p < .01$

Objective 3: Relations between executive functioning and pain catastrophizing, pain severity, and pain interference. Study hypotheses were partially supported with regard to examining relations between EF and pain catastrophizing, severity, and interference. After entering the covariates greater difficulties with inhibition was associated with increased pain catastrophizing (β=.37, $p<.01$, $\Delta R^2=.13$; see Table 13). Further, greater difficulties with shifting was also associated with increased pain catastrophizing (β=.48, $p<.01$, $\Delta R^2=.23$; see Table 13).

While not statistically significant there were trending relationships between inhibition and pain interference (β=.26, *p*=.07, ΔR^2=.07; see Table 14) and between shifting and pain interference (β=.28, *p*=.06, ΔR^2=.07; see Table 14). Neither inhibition nor shifting was significantly associated with pain severity (see Table 14).

Table 13. Hierarchical Linear Regression Analyses for Covariates and Inhibition and Shifting Predicting Pain Catastrophizing

Predictor	b	β	ΔR^2
Age	.32	.06	.05
SES	-.02	-.03	.05
Gender	5.34	.21	.05
Illness Severity	.50	.06	.05
Inhibition	**12.12**	**.37**	**.13****
Age	.05	.01	.05
SES	-.02	-.02	.05
Gender	3.90	.15	.05
Illness Severity	.32	.04	.05
Shifting	**13.17**	**.48**	**.23****

Note: The covariates of youth age, gender, SES, and illness severity were entered in a block in enter selection.
**p* < .05, ** *p* <.01

Table 14. Hierarchical Linear Regression Analyses for Covariates and Inhibition and Shifting Predicting Pain Severity and Interference

Predictor	b	β	ΔR^2
Pain Severity			
Age	.09	.10	.05
SES	-.01	-.06	.05
Gender	.73	.17	.05
Illness Severity	-.13	-.10	.05
Inhibition	.76	.14	.02
Age	.07	.08	.05
SES	-.01	-.06	.05
Gender	.62	.15	.05
Illness Severity	-.14	-.11	.05
Shifting	.82	.18	.03
Pain Interference			
Age	.16	.14	.06
SES	-.02	-.13	.06
Gender	.77	.15	.06
Illness Severity	-.16	-.11	.06
Inhibition	1.69	.26	.07
Age	.13	.13	.06
SES	-.02	-.12	.06
Gender	.56	.11	.06
Illness Severity	-.17	-.11	.06
Shifting	1.47	.28	.07

Note: The covariates of youth age, gender, SES, and illness severity were entered in a block in enter selection.
*$p < .05$, ** $p < .01$

Objective 4: Examining executive functioning as a moderator. Contrary to hypotheses, inhibition did not significantly moderate the relationship between pain catastrophizing and pain severity, nor did inhibition moderate the relationship between pain catastrophizing and pain interference. Similarly, shifting also did not significantly moderate the relationship between pain catastrophizing and pain severity or pain interference (*p*s>.05).

Discussion

The purpose of this study was to examine associations among pain catastrophizing, executive functions, and pain severity/interference in young adults with SB. Specifically, this

study explored: (1) relations between pain catastrophizing and pain severity/interference, (2) relations between inhibition and shifting and pain catastrophizing, severity, and interference, (3) and whether inhibition and shifting moderated the relationship between pain catastrophizing and pain severity/interference. This study also provided an overarching look at the nature of pain and maladaptive pain responses in this population; we described how pain affects domains of daily life and the prevalence of pain catastrophizing in young adults with SB. To our knowledge no study has explored pain catastrophizing in this population, nor relations among pain severity, pain interference, pain catastrophizing, and EF in young adults with SB. Therefore, this study aimed to inform future interventions for pain management in SB by: (1) highlighting the importance of assessing pain in SB as individuals enter young adulthood, (2) understanding maladaptive pain coping responses in this population, and (3) examining neuropsychological mechanisms, related to the cognitive profile of SB that may underlie pain symptoms in this population.

The first aim of this study, to describe the nature of pain catastrophizing, severity, and interference in young adults with SB, yielded several important results. About a quarter of this sample reported moderate to severe levels of pain. Further, the majority of participants with substantial pain severity indicated that they had significant pain interference. These findings echo previous work that has found that a small but sizeable minority of youth with SB experience significant pain (Oddson et al., 2006; Ohanian et al., 2020) and that these symptoms impact their wellbeing (Bellin et al., 2013; Essner et al., 2014). Still, pain interference was predominately rated as mild in the current study sample. Therefore, most young adults with SB may not consider pain to be a significant impediment to their daily functioning. There are several

potential explanations for this finding. Pain has received relatively little attention in SB, particularly in pediatrics; therefore, healthcare providers may not be asking young adults with SB questions about their pain and, consequently, these participants may not be attuned to the ways that pain may be interfering with their functioning. However, this finding may also indicate that pain is not as impactful as other secondary conditions with which individuals with SB have to contend (e.g., catheterization, bowel program, ambulation difficulties, etc.). Given the paucity of research on pain in young adults with SB, it is difficult to determine how these findings fit within the literature. Within the larger adult literature, one study found that pain significantly impacted daily life in adults with SB (Wagner et al., 2015), whereas another study found that adults with SB report relatively little pain interference (Alriksson-Schmidt et al., 2018). This variability is likely due in part to the methodological characteristics of these studies; specifically, they measured pain interference across only one or two domains (i.e., "sleep and work" or "daily life") and either reduced the range of possible responses or dichotomized the responses, minimizing variability. Therefore, more rigorous research is required to understand pain interference in this population, specifically within the context of young adulthood.

With regard to catastrophizing, a small but significant minority of participants (26%) reported a moderate to high level of pain catastrophizing. From a clinical perspective, these participants are at a significantly increased risk for developing chronic pain and related disability (Sullivan et al., 1995). Participants were also found to endorse pain magnification the most, followed by pain rumination, and then feelings of helplessness related to pain. Results from this study indicated that, similar to previous research in other populations, increased pain catastrophizing is associated with higher levels of pain severity and pain interference in young

adults with SB. Further, females were more likely to indicate elevated levels of pain interference and pain catastrophizing than males. Combined with prior research that has consistently demonstrated the negative impact of pain catastrophizing (e.g., increased disability; Tran et al., 2015; Turner et al., 2002), these results highlight the need to screen for pain and pain catastrophizing in young adults with SB during regular doctors' visits, particularly in young adult females with SB.

The second hypothesis of this study, that executive dysfunction would be positively related to pain catastrophizing, severity and interference, was partially supported. Shifting deficits were associated with increased pain catastrophizing. Further, this relationship was also present between inhibition and pain catastrophizing. Therefore, young adults with SB who have a harder time shifting may not be able to move away from their catastrophic thoughts about pain. Similarly, those who have difficulties controlling their attention may also struggle to override or inhibit catastrophic thinking about their pain symptoms and focus their attention elsewhere. These findings are in line with the greater pediatric literature that has found that executive dysfunction is associated with the utilization of maladaptive coping strategies (Campbell et al., 2009; Hocking et al., 2010). Further, it has been hypothesized that youth who appraise their coping potential, or their ability to effectively cope with stress, as lower are more likely to use maladaptive coping strategies (Lazarus & Folkman, 1984). Indeed, this has been observed in pain populations, wherein youth who are less confident in their ability to positively adapt to stress were found to be less likely to utilize positive coping strategies (Walker et al., 2007). This may indicate that young adults with SB who have EF deficits may appraise their coping potential as lower and are consequently more prone to pain catastrophizing. While neither shifting nor

inhibition were significantly related to pain interference, results indicated a trending relationship such that greater executive dysfunction was associated with more pain interference. Future studies should repeat these analyses to determine whether executive dysfunction may in fact result in increased pain interference in young adults with SB. Finally, neither shifting nor inhibition was significantly associated with pain severity. This may indicate that executive functioning deficits do not necessarily increase the pain itself but instead represent a barrier to proper pain management, thereby leading to more maladaptive pain responses.

The third hypothesis of this study, that executive functions would moderate the relationship between pain catastrophizing and pain severity/interference, was not supported. Therefore, difficulties with shifting and inhibition do not increase the impact of pain catastrophizing on pain severity and interference. Relations among these variables may be better understood in a meditational model. Specifically, given that there was a strong relationship between both shifting and inhibition and pain catastrophizing, strong relations between pain catastrophizing and pain severity/interference, and no significant relations between these executive functions and pain severity/interference, future studies should examine whether pain catastrophizing mediates the relationship between executive functioning and pain severity/ interference. Indeed, it may be helpful to examine these variables through the lens of Lee and colleagues' (2012) cognitive vulnerability model, wherein neuropsychological deficits make one more vulnerable to poor coping and depressive symptoms. A previous study examining neuropsychological functioning, depression, and medical responsibility in youth with SB supported the use of this model (Stern et al., 2018). Moreover, previous findings indicating that improved EF is associated with adaptive coping skills (Stubberd et al., 2015) provide further

support for a possible meditational model. Poor EF may result in youth being more vulnerable to maladaptive coping responses, resulting in more pain and pain interference. We were unable to examine this model as the study data was all cross-sectional and, therefore, we would not have been able to accurately interpret the results (Maxwell & Cole, 2007).

However, the non-significance of the moderation effect in this study may also be due, at least in part, to how we measured executive functioning. This study only used self-report measures; therefore, we were not able to obtain a comprehensive perspective on associations among executive dysfunction, pain catastrophizing, and pain severity/interference. Future studies should include performance-based measures to confirm the current study's findings.

This study had several strengths, including the use of the pain catastrophizing scale in this population and this study's focus on young adulthood, an age group that is often overlooked in both the pain and SB literature. However, this study also had several limitations. First, we used a cross-sectional design, which limits our ability to determine directionality between pain catastrophizing, and pain severity and interference, as well as between executive functions, and pain catastrophizing, severity, and interference. Second, our small sample size reduced the power of our analyses, thereby increasing the likelihood of Type II error. Specifically, we had several trending results, which may not have been statistically significant due to low power. Third, this study was vulnerable to common method variance because the primary study measures were all young adult self-report. Fourth, we were not able to examine pain chronicity, a very important construct in this literature due to the structure of the BPI. Finally, as previously stated, we were not able to conduct a thorough examination of the role of neuropsychological functioning in relation to the pain variables in this study because we did not have objective measures of

neuropsychological functioning. Nevertheless, this study filled a significant gap in the literature, as it was the first study to examine pain catastrophizing and pain interference in young adults with SB, and to examine relations among these variables and executive functioning.

Conclusions and Clinical Implications

The results from the current study further demonstrate that pain is an essential area for assessment and intervention in individuals with SB. Pain is not often considered a fundamental part of the experience of SB. Nevertheless, about 26% of young adults in our study reported having moderate to severe levels of pain. Further, while overall pain interference was reported to be mild, 70% of those with substantial pain severity reported experiencing moderate to severe levels of pain interference. A small but significant minority of participants also reported clinically elevated levels of pain catastrophizing, placing them at an increased risk for developing chronic pain. There is increasing evidence that a sizeable minority of individuals with SB across multiple developmental stages experience elevated pain symptoms. To develop effective interventions for this population, future research should examine whether pain is persistent in this minority across development or whether physical and psychosocial developmental changes result in some individuals experiencing the maintenance of symptoms of pain while others experience a reduction in pain over time. This study also found EF to be significantly related to pain catastrophizing. Thus, it is imperative that interventions for pain in this population account for the unique cognitive profile of this condition, including EF deficits.

CHAPTER FIVE

DISCUSSION

Review of Study Purposes and Results

Increasingly researchers have come to recognize that pain is a common experience among youth and young adults with SB. Given the complexity of SB, it is crucial that we gain an understanding of how pain *uniquely* functions in this condition. SB and pain both impact multiple domains of functioning (e.g., family relations, psychological functioning). Consequently, it is imperative that researchers utilize a framework that considers the impact of SB and pain broadly. To do so requires an examination of how pain affects and interacts with the unique biological, neuropsychological, and psychosocial vulnerabilities and areas of strength that individuals with SB and their families possess. The current collection of research sought to address this need by conducting three studies that were based on the bio-neuropsychosocial model of adjustment (Holmbeck & Devine, 2010).

The first study, Ohanian et al. (2020), focused on relations between pain and biological and psychological factors. This study provided an overarching examination of pain in youth with SB, examined coping responses, and explored longitudinal associations between pain and internalizing symptoms. We found that about a quarter of our participants reported experiencing chronic pain, and a third of this subsample continued experiencing chronic pain two years later. Further, while the full sample reported overall mild pain severity, the chronic pain subsample reported moderate to severe pain. The chronic pain subsample reported pain in their extremities,

back, abdomen and head, and reported using predominantly condition-specific forms of pain coping (e.g., taking medication, removing braces). The subsample and larger sample both reported pain to be primarily "aching" in nature. Females, older children, and individuals with lower intellectual functioning were found to be a higher risk for pain symptoms. Finally, longitudinal associations revealed that internalizing symptoms preceded pain symptoms (rather than the reverse) in this population. Specifically, teacher-reported internalizing symptoms predicted youth chronic pain status, and child-reported internalizing symptoms predicted increased pain intensity two years later.

The second study aimed to understand social factors relevant to the experience of pain. Specifically, we explored relations between youth pain and parent mental health, as well as between youth pain and family functioning (family cohesion and conflict). This study revealed several important associations between family functioning and pain using a longitudinal, multi-method, and multi-informant design. Youth chronic pain status at Time 1 was associated with reduced child-reported family conflict and increased parent-reported cohesion two years later. On the other hand, higher parent-reported family conflict at Time 1 was associated with youth chronic pain status two years later. No significant associations were found between observed family functioning and youth chronic pain. Further, no associations were found between parent mental health and youth chronic pain.

The third study explored relations among neuropsychological factors, cognitive appraisals of pain (i.e., pain catastrophizing), and pain severity/interference in young adults with SB. This study aimed to: (1) describe the nature of pain catastrophizing, pain severity, and pain interference in this population, (2) examine associations between pain catastrophizing and pain

intensity/interference, and (3) understand how the unique neuropsychological profile in SB (executive functioning deficits) interacts with pain and pain catastrophizing. Further, this study examined pain within the context of an important developmental period, namely, young adulthood. We found that about a quarter of our sample reported experiencing significant pain and similarly that about a quarter of our sample reported elevated levels of pain catastrophizing. Pain interference was reported to be relatively mild; however, for those reporting substantial pain, pain interference was often reported to be in the moderate to severe range. As hypothesized, pain catastrophizing was positively associated with pain severity and interference. Further, executive functioning, specifically inhibition and shifting, were associated with pain catastrophizing, such that greater difficulties with inhibition and shifting were associated with more pain catastrophizing. No significant associations between executive functions and pain severity/interference were found. Moreover, our hypothesis that executive functions would moderate the relationship between pain catastrophizing and pain severity/interference was not supported.

These findings provide several important insights into the nature of pain in SB. The first and third studies both documented the prevalence of pain symptoms in this population. Indeed, rates of pain chronicity in children and adolescents with SB are similar to that of typically developing youth (King et al., 2011). Further, pain severity was also significantly elevated in young adults with SB, indicating that youth with SB experience significant pain symptoms across development. Both of these studies also provided information about how pain can be conceptualized in SB *specifically*. Indeed, when examining associations between pain and biological characteristics, such as shunt status and lesion level, few significant associations were

found. This was consistent across the developmental periods assessed in this collection of research (i.e., older children, adolescents, and young adults). In fact, in the first study, illness severity was found to be significantly associated with change in pain intensity, such that *lower* illness severity was associated with higher pain intensity over time. This information is very important for determining who is at risk for developing pain symptoms in this highly variable condition. Although increased illness severity is negatively associated with a number of functional outcomes (Hommeyer et al., 1999), pain-related difficulties may be more impactful for those with *lower* illness severity. With less illness severity, one might expect fewer competing stressors (e.g., catheterization, skin checks, etc.) thereby potentially making pain a more recognizable condition parameter and a more salient contributor to quality of life and functional outcomes. Further, those with lower illness severity may be more likely to have less severe or no paralysis. This may make it easier for them to be mobile and use their muscles, increasingly the likelihood of muscle pain. Nevertheless, it should be noted that illness severity was not significantly associated with pain variables in the third study. Therefore, it may be that as youth grow older some of those with lower illness severity experience less pain and some of those individuals with higher illness severity start to experience more pain, potentially due to the alleviation or increase of other important etiological factors, such as psychological functioning, discussed further below.

On the other hand, gender emerged as an important risk factor for pain. Studies 1 and 3, which utilized the same study sample at different time points, both identified female gender to be significantly associated with pain symptoms. Indeed, females with SB reported more pain severity, interference, and catastrophizing than males with SB. These findings are consistent with

the greater pain literature both in pediatric (King et al., 2011) and adult populations (Husky et al., 2018; Tsang et al., 2008), across pain conditions (e.g., headache, back pain, joint pain; Jiménez-Trujillo et al., 2019; King et al., 2011) and throughout the world (Tsang et al., 2008). Therefore, our findings contribute to this research by further highlighting female gender as a significant risk factor for the development of pain and pain-related disability.

This collection of research also demonstrated that, in addition to being a physical experience, pain in SB is a psychosocial experience. The longitudinal nature of Studies 1 and 2 allowed us to examine directionality with regard to relations between pain and psychosocial functioning, which revealed several important findings. Regarding social functioning, the presence of pain did not lead to reduced family functioning. In fact, chronic pain status was associated with more family cohesion and less conflict over time. Further, pain also did not appear to lead to worse psychological functioning over time. Our findings echo previous research that has found that youth with SB and their families experience disruption *and* demonstrate resilience in the face of significant illness stressors (Holmbeck et al., 2010; Lennon et al., 2015a). Therefore, in addition to areas of risk, we were able to highlight areas of strength that future research can build upon.

In contrast, psychosocial difficulties were associated with more pain over time. Specifically, family conflict was associated with pain chronicity, and youth internalizing symptoms were associated with pain chronicity and intensity. Therefore, when thinking about the etiology of pain in SB, it is important to consider the contribution of psychosocial stressors. These results may indicate that youth with SB struggle to cope with psychosocial difficulties, which results in increased pain symptoms and/or increased attention to pain symptoms. Indeed,

we found some evidence that youth with SB may not use the most effective strategies to manage their pain. In spite of few associations between condition-related variables and pain, youth with SB typically employ condition-related coping methods (e.g., taking ibuprofen, drinking cranberry juice, taking off braces). Further, we found little evidence that youth with SB employ acceptance-based or distraction-oriented strategies. These strategies are very effective for youth with chronic illnesses (Compas et al., 2012), including youth with chronic pain (Gauntlett-Gilbert et al., 2013). However, it may be more difficult for youth with SB to employ these methods, as some have suggested that these techniques require stronger cognitive skills. Specifically, several studies have found that stronger executive functioning predicts more use of these coping strategies in pediatric populations (Campbell et al., 2009; Desjardins et al., 2018).

Indeed, promoting healthy adjustment in this population requires one to consider the unique cognitive profile of youth with SB. Research has shown that multiple areas of functioning in SB are influenced by cognitive functioning, such as medical autonomy (Stern et al., 2018) and adherence (O'Hara & Holmbeck, 2013), social competence (Lennon et al., 2015b), and emotional autonomy (Friedman et al., 2009). Our findings suggest that pain and pain related processes are also associated with cognitive functioning. Unexpectedly, we found associations between lower intellectual functioning and pain chronicity. While lower IQ has not typically been associated with pain symptoms (Ho et al., 2009), this association may indicate that reduced cognitive abilities make it more difficult for one to mitigate pain through adaptive coping skills. Indeed, in a study with youth with sickle cell disease, strong verbal comprehension was associated with increased adaptive coping (Prussien et al., 2018). Further, we hypothesized that executive dysfunction may also underlie this association. We found that executive functioning

was related to pain catastrophizing in young adults with SB. Therefore, executive functioning deficits may indeed make it more difficult to employ positive coping techniques and increase the likelihood that young adults with SB will engage in pain catastrophizing, an extremely maladaptive coping response. As such, these deficits that are often considered intrinsic to the condition, may ultimately leave young adults with SB vulnerable to developing pain-related disability.

Strengths, Limitations, and Future Research

These studies had multiple strengths, including the use of longitudinal data and multiple methods, the inclusion of multiple reporters, and bi-directional designs. Further, this collection of research examined pain in SB, a condition parameter that has received relatively little attention to date. In spite of these strengths, several limitations must be noted. First, there were several weaknesses regarding how pain was measured across these studies. For the first two studies, we used The Spina Bifida Pain Questionnaire (SBPQ), which was created for the larger longitudinal study and was comprised of questions that are commonly found in validated pediatric pain measures. Still, this questionnaire has not been validated in youth with SB. Given that the majority of youth with SB reported mild pain severity and frequency, our methods of measurement may not have been sensitive enough to detect significant variability and give us a true picture of pain in this population. Further, we were not able to assess what youth with SB believe to be the causes of their pain (e.g., shunt revisions, pain due to ambulation method). This would have provided invaluable information about how clinicians should approach intervention and psychoeducation about pain. This collection of studies also relied solely on youth report of pain. The current literature recommends including both parent and youth report when assessing

pediatric pain (Cohen et al., 2008). Moreover, both parent report and observational methods are commonly included in the assessment of pain for youth with cognitive deficits, who may not be reliable reporters (Breau & Burkitt, 2009). Therefore, given the variability in cognitive functioning in this population, multiple methods of pain measurement should be used in future studies. Further, our study methods did not capture day-to-day temporal relations between pain and the psychosocial and neuropsychological variables. Daily diary methods have increasingly been recognized as important methods to use in chronic illness populations (Quittner et al., 2000) and specifically when studying pediatric pain (Stinson, 2009). Finally, there were also some potential issues with the measure used in the third study. While the Brief Pain Inventory is a well-validated measure of pain, it does not include questions about chronicity, limiting our ability to understand if chronicity remained the same across this set of studies, which would have provided insight into the nature of pain chronicity at different developmental stages in this population (i.e., late childhood, adolescence, and young adulthood). Further, this measure was originally created for cancer pain and is therefore oriented toward measuring acute vs. chronic pain. Indeed, it prompts the respondent to think about their pain "over the last 24 hours," thereby only allowing us to obtain a snapshot of pain for these participants. Future research should use pain measures that have been validated with individuals with SB, include multiple reporters of pain, employ observational methods, and include daily diary measures.

It is also important to note several limitations in study design across this collection of studies. First, all three studies had a fairly low sample size which, although common in pediatric research, resulted in reduced power. These studies were only powered to detect medium to large effects, with Study 3 barely making this cut off (Study 3: N=80, Cut off=77; Cohen, 1992).

Therefore, the nonsignificant results of these studies should be interpreted with caution. Moreover, while efforts to reduce analyses were made by collapsing across reporters, all three studies included numerous regression analyses, increasingly the likelihood of Type 1 error. Moreover, while collapsing across reporters for Studies 1 and 2 (Study 1: parent report of child internalizing, Study 2: parent report of family cohesion and conflict) may have reduced the likelihood of common method variance and reduced the number of analyses, we may have also missed subtle differences between father and mother perceptions of their child's internalizing symptoms or the family environment, and these differences may have impacted our findings. Indeed, other studies have found differences between mothers and fathers with regard to associations between parenting stress and parent perception of child vulnerability for youth with SB (Driscoll et al., 2017). Therefore, future studies should consider examining mother and father reports separately. Finally, Study 3 was also cross-sectional, limiting our ability to determine directionality among executive functioning, pain catastrophizing, and pain severity/interference. Overall, future studies should try to replicate these analyses in a larger sample, reduce the number of analyses when possible, and examine pain and pain related processes in young adults with SB longitudinally.

While this research focused on multiple developmental periods, future research should directly examine the trajectory of pain in youth and young adults with SB. Although the current studies have allowed us to identify important risk factors for developing pain, a great deal remains unknown about the timing of pain onset. For example, it would be important to understand if those children who reported chronic pain at Time 1 but not at Time 2 in Study 1 cease experiencing chronic pain or if their pain symptoms re-emerge in relation to psychosocial

or developmental changes later in adolescence or in young adulthood. If we were able to identify sensitive periods wherein the emergence/re-emergence of pain is likely to happen in this population, it would enable researchers to develop effective interventions. Future studies should consider using more complex longitudinal designs, such as growth curve modeling, to obtain a better understanding of the development of pain in this population.

Although these studies examined important aspects of social functioning, several domains remain unexplored. First, while Study 2 revealed important findings regarding family functioning, we were not able to examine parenting behaviors in relation to pain. Specifically, future studies should examine solicitous parenting behaviors in relation to youth pain symptoms in SB. When parents engage in solicitous behaviors, they draw attention to their child's pain, tend to be overly protective, and ultimately reinforce pain behaviors, such as avoiding physical activity. These behaviors have been associated with increased pain-related disability in youth with chronic pain (Palermo et al., 2014). Moreover, we did not explore peer relationships. Indeed, a systematic review of social functioning in pediatric pain found that youth with chronic pain tend to have fewer friends and experience more peer victimization than their TD peers (Forgeron et al., 2010). Moreover, a previous study examining pain in youth with SB found that pain symptoms were associated with reduced social activity involvement (Essner et al., 2014). As such, future studies should also explore relations between pain and peer relationships in youth with SB.

Conclusions and Clinical Implications

Clinically, the results from this collection of research underscore the importance of assessing and treating pain in SB. As such, assessment of pain symptoms should be part of

regularly scheduled clinic visits. Further, these studies found significant associations between pain symptoms, and IQ and executive functioning; therefore, it will be important for clinicians to approach pain assessment in a developmentally appropriate manner. This includes obtaining parent report of youth pain symptoms (Cohen et al., 2008) and asking their patients about their pain using multiple modalities (e.g., scales, pictures, verbal descriptions). For example, younger children often report their pain on a faces pain scale (i.e., line of faces with expressions of increasing distress; Tomlinson et al., 2010). Youth with cognitive deficits may benefit from using this type of measurement. It is also recommended that clinicians alter the language they use to fit their patient's developmental age vs. their chronological age.

Further, psychosocial vulnerabilities emerged as important predictors of pain in this population. Therefore, clinicians should continually assess their patients for symptoms of depression and anxiety as well as difficulties at home (e.g., increased family conflict). Moreover, when psychosocial difficulties arise, clinicians should encourage early intervention to mitigate the potential emergence of pain symptoms. Indeed, psychologists who work with youth with SB should be aware of the relationship between psychosocial stressors and pain in SB. Teaching effective coping strategies for psychosocial stressors might lessen the likelihood that youth will experience elevated or burdensome pain symptoms. Further, for youth with SB who are experiencing pain symptoms, psychologists should teach their patients how to engage in acceptance- and distraction-based coping strategies. They should also assess whether their patients have catastrophic thoughts and use both cognitive restructuring and behavioral techniques to reduce the tendency to catastrophize. Moreover, these strategies should again be tailored to the cognitive level of the patient, with increased structure (i.e., reminders to practice

skills) and adapting the methodology to developmental age (i.e., for cognitive restructuring, distracting oneself from one's thoughts rather than trying to challenge maladaptive thoughts). Finally, like all interventions, psychologists should approach psychotherapy for pain with cultural humility, making efforts to include cultural values in the conceptualization and treatment plan.

This research demonstrated that pain is a prevalent and impactful condition in SB. Using the bio-neuropsychosocial model, we showed that pain in SB affects and is affected by multiple areas of functioning. Relations were found between pain, and psychological, social, and neuropsychological variables. Further, pain was found to be impactful across multiple developmental periods. Several key areas of risk and resilience were identified, allowing future interventions to both address these risks and utilize strengths-based models with individuals and families with SB. Future studies should continue to use this framework to address areas that these studies did not examine to facilitate the development of comprehensive interventions.

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VITA

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