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LOYOLA UNIVERSITY CHICAGO

SLEEP-WAKE DISTURBANCES IN ADOLESCENTS WITH SPINA BIFIDA:
PREVALENCE AND ASSOCIATIONS WITH BIO-NEUROPSYCHOSOCIAL
FUNCTIONING

A DISSERTATION SUBMITTED TO
THE FACULTY OF THE GRADUATE SCHOOL
IN CANDIDACY FOR THE DEGREE OF
DOCTOR OF PHILOSOPHY

PROGRAM IN CLINICAL PSYCHOLOGY

BY

CAITLIN B. MURRAY

CHICAGO, IL

AUGUST 2017

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ABSTRACT

Sleep is a critical component of healthy developing during adolescence, and when disrupted, has been linked to difficulties with physical status, psychological health, family functioning, neuropsychological symptoms, and academic performance. The overarching goal of this project was to examine sleep-wake disturbances in association with bio-neuropsychosocial functioning in a vulnerable pediatric population of adolescents with spina bifida (SB). Specifically, this study aimed to 1) examine sleep-wake patterns in adolescents with SB using a multimodal sleep assessment, 2) identify daily temporal associations between sleep and pain as well as sleep and mood, and 3) identify the relationship between sleep-wake disturbances and bio-neuropsychosocial functioning in adolescents with SB. Sleep-wake patterns in adolescents ages 12 to 18 with SB ($N = 37$) were compared to a matched comparison group of typically developing (TD) peers ($N = 37$). A subjective and objective sleep assessment was conducted; ambulatory actigraphy recordings was completed over 10 days, and adolescents completed several sleep questionnaires (e.g., sleep quality, pre-sleep arousal) and a daily diary. In addition, adolescents and parents completed questionnaires to assess physical (pain, BMI), psychological (internalizing symptoms, health-related quality of life), family (conflict, cohesion), neuropsychological (attention, executive function), and academic functioning (school competence, grades).

Study findings revealed that adolescents with SB experienced higher rates of sleep-wake disturbances compared to their typically developing peers. Results of actigraphy and

questionnaire report data found that adolescents were particularly at-risk for reduced sleep quantity (i.e., lower total sleep time) and poor sleep quality (i.e., difficulties with bedtime settling and staying asleep). Adolescents with SB also experienced higher levels of daytime fatigue compared to their peers. Sleep-wake disturbances were associated with every domain of adolescent functioning within the bio-neuropsychosocial model. In particular, there were consistent data to support the connection between nighttime sleep disturbances and psychological maladjustment (i.e., internalizing, quality of life). To a lesser extent, nighttime sleep disturbances were linked to worse physical health (pain, BMI) and family functioning (family conflict). Furthermore, daytime sleepiness and/or fatigue, but not nighttime sleep disturbances, predicted worse neuropsychological and academic functioning, including inattention/hyperactivity, executive dysfunction, and lower school grades. Ongoing evaluation and treatment of sleep disturbances will be critical to optimize health and functioning in this vulnerable pediatric population.

CHAPTER ONE

INTRODUCTION

Overview

Sleep is a critical aspect of adolescent development, and is associated with multiple domains of adolescent functioning (Mindell & Owens, 2010; Figure 1). Specifically, poor nighttime sleep may adversely impact key bio-neuropsychosocial outcomes, including youths' physical, psychological, family, and neuropsychological functioning (e.g., pain, mood, attention; Alfano, Zakem, Costa, Taylor & Weems, 2009; Chen, Beydoun & Wang, 2008; Kheriandish & Gozal, 2006). Research also indicates a bidirectional relationship between sleep and poor daytime functioning; sleep may contribute to pain and mood disturbance, which, in turn, may exacerbate sleep disruptions (Figure 2; Dahl & Lewin, 2002; Valrie, Bromberg, Palermo & Schanberg, 2013). Sleep-wake disturbances are characterized by nighttime sleep disruption and daytime sleepiness (e.g., frequent nighttime awakenings, excessive daytime fatigue), and are especially prevalent in youth with chronic illnesses and medical conditions (e.g., Hysing, Sivertsen, Stormark, Elgen & Lundervold, 2009). Adolescents with spina bifida (SB), a congenital neural tube defect, are at-risk for sleep-wake disturbances (Quine, 1991) in addition to poor physical, psychological, family, and neuropsychological functioning (Appleton, 1997; Clancy, McGrath, & Oddson, Holmbeck et al., 2003; Holmbeck et al., 2010; Rose & Holmbeck, 2007). However, few studies have comprehensively assessed the relationship between sleep-wake disturbances and key bio-neuropsychosocial outcomes in adolescents with SB. The current

study sought to bridge this critical gap in knowledge through a comprehensive examination of sleep-wake disturbances as predictors of bio-neuropsychosocial functioning in this vulnerable pediatric population.

Figure 1. Theoretical Model of the Associations between Sleep-wake Disturbances and Adolescent Bio-neuropsychosocial Functioning

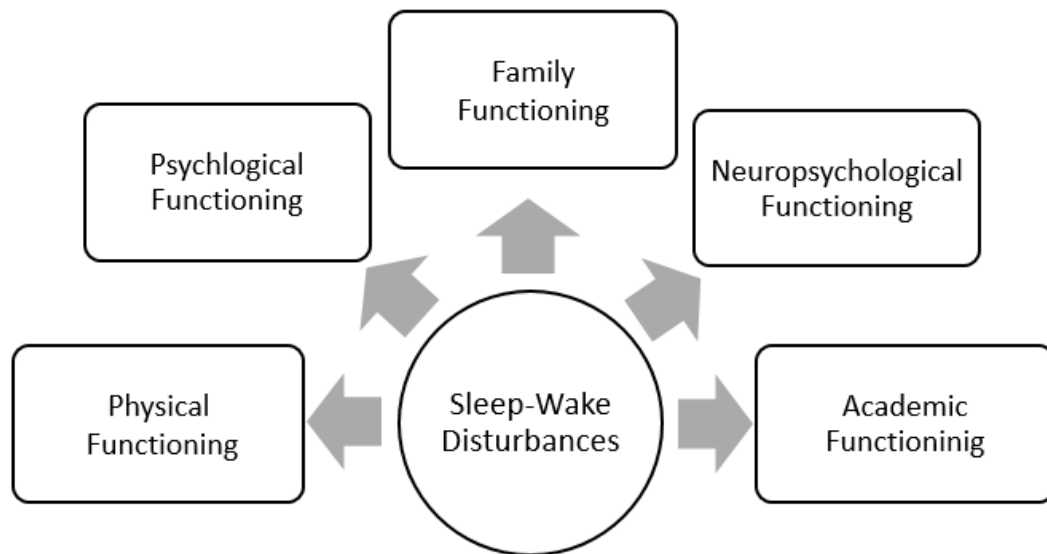
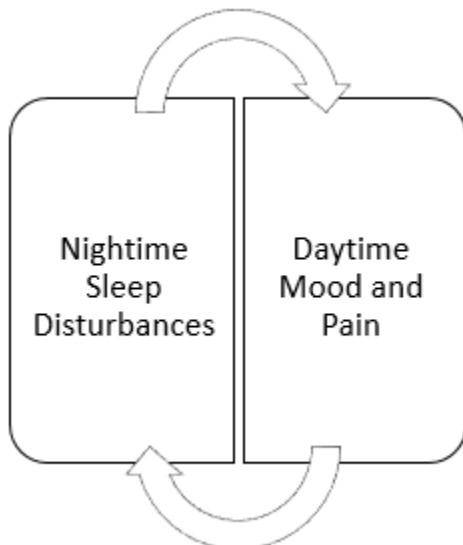


Figure 2. Daily Associations between Adolescent Sleep, Mood, and Pain



The following sections provide an overview of the literature on sleep in adolescence, the definition and assessment of sleep-wake disturbances, and sleep-wake disturbances in youth with

chronic medical illnesses and in youth with SB in particular. Research on associations between sleep and adolescent bio-neuropsychosocial outcomes is reviewed and applied to youth with SB. Finally, the current study is described along with the specific aims and hypotheses of the proposed project.

Sleep in Adolescence

Alterations in sleep patterns and the timing of sleep are normative features of adolescence (Carskadon & Tarokh, 2013). Adolescent sleep patterns are often referred to as the “perfect storm” to describe how several bio-neuropsychosocial factors decrease total sleep time, delay the timing of sleep, and increase daytime fatigue and sleepiness (Carskadon, 2011). Adolescents have been shown to need about 9 hours of sleep according to laboratory studies (Carskadon & Acebo, 2002), yet most sleep only about 7 hours per night (Wolfson & Carskadon, 1998). Therefore, the loss of sleep during this developmental period is not the result of a lower need for sleep. Instead, dramatic changes in adolescent sleep patterns arise from a convergence of biological and psychosocial influences.

Data on biological regulation of sleep during adolescence indicate that adolescents develop a sleep phase delay of about two hours (i.e., later sleep onset and wake times) around the time of puberty onset (Carskadon & Acebo, 2002). Developmental and psychosocial factors may further exacerbate sleep delay and decrease the overall time adolescents spend sleeping. For example, developmentally normative adolescent strivings to achieve independence by self-selecting bedtimes may contribute to delayed sleep onset (Carskadon, 2011). Screen time/technology use in the bedroom, academic responsibilities, extracurricular activities, and social opportunities also tend to increase during adolescence and may further contribute to later

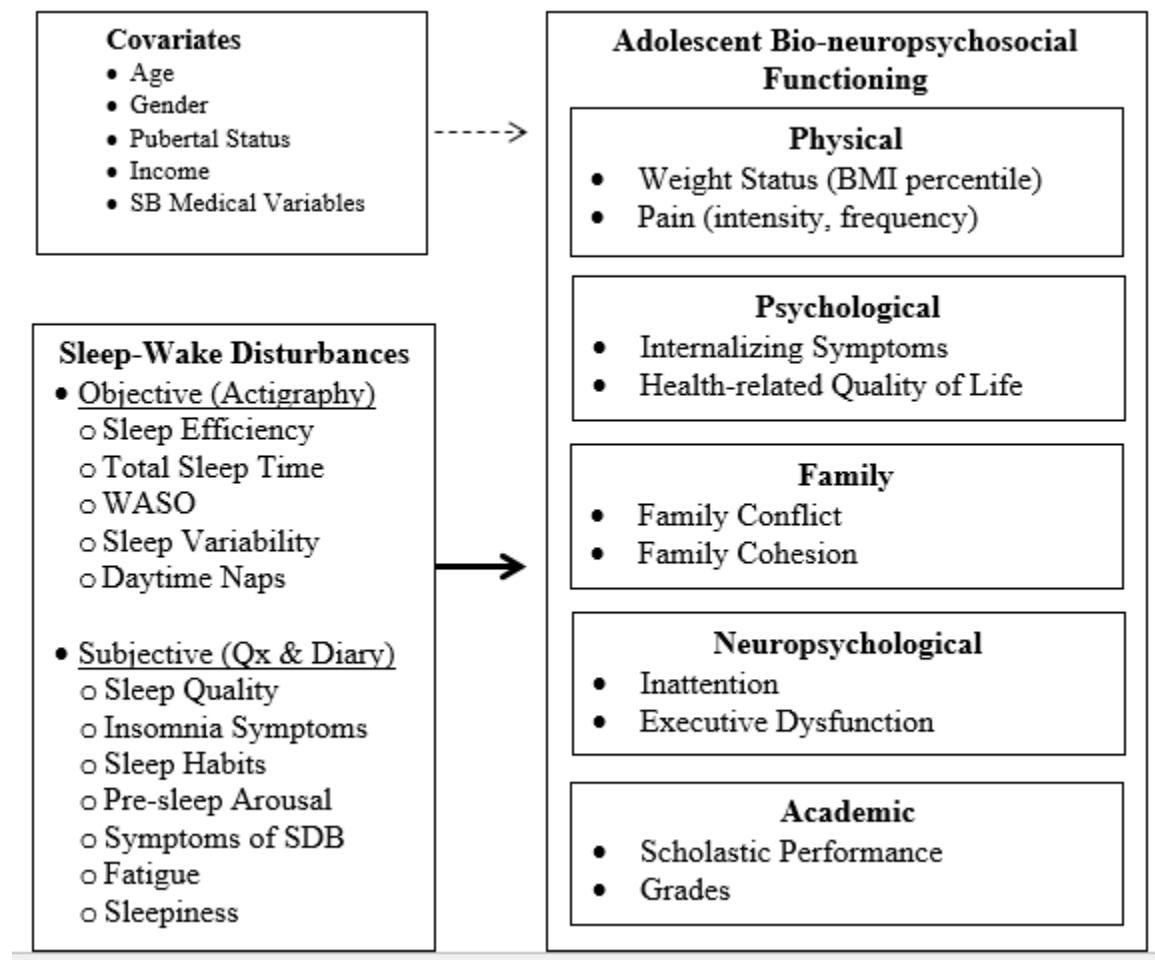
bedtimes and sleep loss (Cain & Gradisar, 2010; Carskadon, 2011). Changes in sleep patterns during this developmental period are also often accompanied by nighttime and daytime sleep disturbances, including daytime sleepiness and fatigue (Carskadon, 1990; ter Wolbeek, van Doornen, Kavelaars, & Heijnen, 2006), increased napping (Shinkoda, Matsumoto, Park, & Nagashima, 2000), and difficulties initiating and maintaining sleep (i.e., insomnia symptoms; Roberts, Lee, Hernandez, & Solari, 2004). Between 6-10% of adolescents are considered to have DSM-defined insomnia (Roberts, Roberts & Chan, 2008; Johnson, Roth, Schultz, & Breslau, 2006), adding to growing evidence that clinically significant sleep disorders among adolescents have prevalence rates comparable to major psychiatric disorders (e.g., mood and behavioral disorders; Roberts et al., 2008). Further, sleep disturbances may also become chronic; several studies have indicated that sleep disturbances persist or recur over time (Morrison, McGee, & Stanton, 1992; Roberts et al., 2008). Thus, early identification and treatment of sleep disturbances may be particularly beneficial.

Definition and Assessment of Sleep Disturbances

Sleep is a multidimensional construct. Nighttime sleep disturbances in adolescents can be categorized as 1) inadequate sleep quantity for age (i.e., insufficient sleep), 2) poor sleep quality (i.e., fragmented sleep), and sleep disorders such as those involving inappropriate timing of sleep (i.e., circadian rhythm and primary sleep disorders; Mindell & Owens, 2010). For the current research project, the majority of the measured sleep constructs fit under the first two categories of nighttime sleep disturbances (see Figure 3). Insufficient sleep is typically the result of difficulty initiating (i.e., delayed sleep onset) and/or maintaining sleep (i.e., prolonged or frequent nocturnal awakenings) with the result being inadequate sleep quantity (Mindell &

Owens, 2010). Sleep fragmentation often involves brief and frequent arousals during sleep (Mindell & Owens, 2010). Further, daytime sleep disturbances may include frequent napping excessive sleepiness, and fatigue (Carskadon, 1990; ter Wolbeek et al., 2006). While closely related, fatigue and sleepiness are distinct constructs. In the current study, sleep-wake disturbances are defined as both nighttime and daytime sleep disturbance, such as nighttime sleep fragmentation and daytime fatigue.

Figure 3. Study Model of the Associations between Sleep-wake Disturbances and Bio-neuropsychosocial Functioning in Adolescents with Spina Bifida



Accurate assessment of sleep-wake disturbances in children and adolescents is important across research and clinical settings. Due to the multidimensional nature of sleep, evaluation of sleep disturbances in children and adolescents often requires a combination of objective and subjective sleep assessment tools (Lewandowski, Toliver-Sokol & Palermo, 2011), which are briefly described in the following section.

Objective Sleep Measurement

The most widely used tools for objective measurement of sleep include polysomnography (PSG) and actigraphy. PSG is a diagnostic test that is typically performed in a sleep laboratory, yielding information on sleep architecture (e.g., distribution of sleep stages) and physiological changes (e.g., eye and body movement, heart rate) that occur during sleep (Mindell & Owens, 2010). Clinicians often use PSG to diagnose the presence and severity of sleep-disordered breathing (SDB), including obstructive sleep apnea (OSA; Mindell & Owens, 2010). OSA is a respiratory disorder characterized by partial or complete obstruction to the upper airway, resulting in sleep fragmentation (Witmans & Young, 2013). Pediatric medical conditions often associated with OSA include craniofacial syndromes, myelomeningocele, Down syndrome, obesity, and sickle cell disease (Mindell & Owens, 2010).

While PSG is considered the gold standard for sleep assessment (Horne & Biggs, 2013), there are several limitations to using this type of sleep data collection. PSG is conducted in a laboratory setting that may alter a participant's typical sleep patterns, is costly, and may be both time-consuming and labor intensive (Horne & Biggs, 2013). A popular alternative to PSG for assessing pediatric sleep is actigraphy. In fact, overall agreement rates of 89% to 97% between actigraphy and PSG have been found in children ages 3-18 (Meltzer, Walsh, Traylor & Westin,

2012). Actigraphy uses an actiwatch with an accelerometer (i.e., a portable wristwatch-like device) to record the presence or absence of movement as an approximation of sleep and wake cycles (Horne & Bigg, 2013). In other words, the presence of movements indicates wakefulness and the absence of movements indicates sleep. Actiwatch data are coded and analyzed using specialized computer software and trained research assistants to approximate sleep-wake patterns (Horne & Bigg, 2013). While actigraphy coding does involve some subjective decisions (e.g., distinguishing daytime periods of quiet wakefulness versus naps), this assessment tool enables researchers and clinicians to gain a clear picture of sleep-wake behavior (e.g., sleep quantity, nocturnal awakenings) in a naturalistic setting (Mindell & Owens, 2010).

Objective sleep measurement tools may be used in conjunction with questionnaire measures to provide a comprehensive examination of sleep in youth. Questionnaire measures and sleep diaries are often dependent on the accuracy and reliability of the person completing them, and thus may have a number of limitations (Horne & Biggs, 2013). As such, it has become standard practice for self-report sleep diaries to be used at the same time as actigraphy recording for greater reliability in reporting sleep/wake patterns as well as to aid in the coding and analyses of sleep-wake patterns (e.g., determination of nap periods; Horne & Biggs, 2013). Moreover, while actigraphy provides information about sleep patterns, this methodology is unable to identify behavioral sleep disturbances (e.g., insomnia) or potential reasons for difficulty falling asleep (e.g., pre-sleep habits and cognitions). Therefore, due to the multidimensional nature of sleep, the use of both objective (i.e., actigraphy) and subjective (i.e., questionnaires, sleep diaries) sleep measurement tools is essential.

Subjective Sleep Measurement

Developmentally appropriate questionnaire measures of sleep disturbances are an essential component of sleep assessment. A recent review article by Lewandowski and colleagues (2011a) identified several subjective pediatric sleep measures that fell into the following categories: 1) daytime sleepiness, 2) sleep habits or hygiene, 3) attitudes and cognitions associated with sleep, 4) sleep initiation and maintenance, 5) and multidimensional constructs. Measures of daytime sleepiness (e.g., Pediatric Daytime Sleepiness Scale; Drake et al. 2013) typically assess behaviors such as falling asleep and alertness during the day, while measures of fatigue (e.g., the Pediatric Quality of Life Inventory- Fatigue; Varni, Burwinkle, & Szer, 2004) may include items related to general feelings of tiredness, need for rest during the day, and cognitive fatigue (Varni et al., 2004). Measures of sleep hygiene and sleep habits (e.g., the Adolescent Sleep Hygiene Scale; LeBourgeois, Giannotti, Cortesi, Wolfson & Harsh, 2005) examine bedtime routines, activities surrounding bedtime, and the sleep environment (Lewandowski et al., 2011a). Measures of sleep-related beliefs and cognitions (e.g., the Pre-sleep Arousal Survey for Children; Gregory, Willis, Wiggs, & Harvey, 2008) examine children's dysfunctional thoughts about sleep (e.g., worry about falling asleep) as well as somatic arousal (e.g., rapid pulse, sweating palms) prior to sleep onset (Lewandowski et al., 2011a). Measures of sleep initiation and sleep maintenance (e.g., the Adolescent Sleep Wake Scale; LeBourgeois et al., 2005) examine sleep quantity and difficulties with falling and staying asleep. Self-reported sleep diaries are also often used to assess sleep-wake patterns in children and adolescents, in which bedtime, the time the adolescent went to sleep, the number and duration of nocturnal awakenings, the time the child awoke in the morning, and any daytime napping are recorded

over a 1-2 week period of time (Horne & Biggs, 2013). Multidimensional pediatric sleep measures (e.g., the Pediatric Sleep Questionnaire, Chervin, Hedger, Dillon, & Pituch, 2000) may assess a broad range of sleep disturbances including sleep habits, fatigue, sleepiness, symptoms of sleep-disordered breathing (e.g., snoring), and nocturnal awakenings (Lewandowski et al., 2011a).

Sleep Disturbances in Youth with Chronic Medical Illnesses

Assessment of sleep is particularly important for pediatric populations, due to the increased risk of sleep disruption among youth with acute and chronic medical conditions (Lewandowski, Ward & Palermo, 2011). Sleep disturbances occur more frequently in children with acute and chronic medical illnesses compared to otherwise healthy or TD peers (Hysing et al., 2009). Sleep disturbances are also more likely to become chronic and persistent in youth with chronic conditions compared to those without chronic conditions (Sivertsen, Hysing, Elgen, Stormark, & Lundervold, 2009). Comprehensive sleep assessments in pediatric populations have expanded in the past decade, identifying disturbed sleep as a prevalent concern across several pediatric medical illnesses: youth with asthma (Sadeh, Horowitz, Wolach-Benodis, & Wolach, 1998), cancer (Hinds et al., 2007), chronic pain conditions (Palermo, Toliver-Sokol, Fonareva, & Koh, 2007), craniofacial abnormalities (e.g., cleft palate, MacLean, Fitzsimons, Hayward, Waters & Fitzgerald, 2008), sickle cell disease (Jacob, Miaskowski, Savedra, Beyer, Treadwell, & Styles, 2006), cystic fibrosis (Amin, Bean, Burklow, & Jeffries, 2005), and neurological conditions (e.g., epilepsy; Maganti, Hausman, Koehn, Sandok, Glurich & Mukesh, 2006). In particular, youth with chronic illnesses may achieve less nighttime sleep and report more difficulties initiating and maintaining sleep compared to TD peers (Hysing et al., 2009).

Various aspects of a chronic illness may affect sleep; specifically, physical and disease-related mechanisms of the illness (e.g., BMI, pain, airway restriction), hospitalization, and medication may all significantly impact sleep (Lewandowski et al., 2011b). Certain brain structures are also connected to sleep, including the hypothalamus, brain stem, and basal forebrain, all of which can be impacted by brain tumors, neurological conditions, and illness treatments (e.g., chemotherapy; Rosen, Shor & Geller, 2008). Medical illnesses such as asthma and epilepsy may have worse symptoms at night, and others (e.g., diabetes) require nighttime medication (Hysing et al., 2009). Further, some medical conditions are associated with particular sleep disorders (e.g., sleep disordered breathing and spina bifida, Waters et al., 1998). Conversely, diminished sleep quantity and quality may exacerbate a chronic illness and disease-related symptomatology (Lewandowski et al., 2011b). Supporting this, Hanson and Chen (2008) found that worse self-reported sleep quality predicted poorer lung function in youth with asthma the following day. Thus, the impact of sleep disturbances in youth with chronic medical conditions may be more concerning given potential bidirectional relations between sleep and health. Poor sleep may worsen the medical condition, and, in turn, disease-related symptoms may exacerbate sleep disturbances (e.g., asthma and cystic fibrosis; Hanson & Chen, 2008; Amin et al., 2005).

Sleep Disturbances in Youth with Spina Bifida

Despite their importance, relatively little research attention has been directed toward assessing sleep disturbances in youth with spina bifida (SB). SB is a relatively common congenital neural tube defect, occurring in 18 out of 100,000 births in the United States (Centers for Disease and Control Prevention, 2010). This condition arises due to the failed closure of one

or more vertebrae surrounding the embryo's developing spinal cord, and is associated with varying degrees of motor impairment, neuropsychological deficits, and difficulties with bladder and bowel management (Liptak 1997). In children and adolescents with SB, studies have revealed the presence of behavioral sleep problems including insomnia symptoms (e.g., difficulty initiating and maintaining sleep; Quine 1991; Edelstein, Cirino, Hasher & Fletcher, 2012) as well as physiological sleep disorders such as central and obstructive sleep disordered breathing (SDB; Waters et al., 1998; Kirk, Morielli & Brouillette 1999). High rates of SDB and other sleep disturbances in this population may be due to central nervous system malformations and pulmonary function abnormalities (Waters et al., 1998). Despite research indicating a high incidence of sleep problems across several illness groups, studies that assess sleep in adolescents with SB remain scarce. Multi-method studies that comprehensively assess sleep-wake disturbances using objective (i.e., actigraphic) and subjective sleep instruments are essential for characterizing the nature of these problems.

Associations between Sleep and Adolescent Bio-neuropsychosocial Functioning (Figure 3)

Sleep is a critical aspect of adolescent development and may be associated with several domains of adolescent functioning (Figures 1 & 3). While adolescents with SB are at-risk for physical, psychological, family, and neuropsychological difficulties, few studies have examined the link between sleep and relevant bio-neuropsychosocial domains in this population. Further, due to increased numbers of individuals with SB surviving into adulthood, and the impediment that bio-neuropsychosocial difficulties may have on self-management in chronic illness populations (Gadalla, 2008), research investigating sleep as a modifiable behavioral mechanism that may improve functioning in adolescents with SB is essential.

Sleep and Physical Functioning

Research on sleep and physical health has tended to focus on two important physical health parameters: pain and weight status or obesity.

Sleep and pain. Persistent or recurring pain is common among adolescents, occurring in 25-40% of community samples (Stanford, Chambers, Biesanz, 2008). Pain may be part of a medical condition's presentation (e.g., sickle cell disease, juvenile idiopathic arthritis), or the condition itself (e.g., migraine headaches; Valrie et al., 2013). Youth with persistent or recurrent pain often complain of disturbed sleep, characterized by difficulties initiating or maintaining sleep and shorter sleep quantity (i.e., insufficient sleep), poor subjective sleep quality, and poor sleep hygiene. Beyond the difficulty of initiating and maintaining sleep when experiencing painful somatic symptoms, increased commonality of sleep disturbances in pediatric pain populations may be due to medications (e.g., analgesics) and hospitalizations that affect sleep (Lewandowski et al., 2011b).

The sleep-pain association has been well documented in adult and pediatric pain populations, with the majority of studies finding evidence of bidirectionality (see review; Valrie et al., 2013). Review articles of adult and pediatric literature have found evidence that pain disrupts sleep, and poor sleep may exacerbate pain (Figure 3; Onen, Onen, Courpron et al, 2005; Valrie et al., 2013), possibly by increasing pain sensitivity (Lautenbacher, Kundermann & Krieg, 2006; Smither, Edwards & McCann, 2007). In juvenile idiopathic arthritis (JIA), sickle cell disease (SCD), headache, and mixed pediatric pain samples, cross-sectional objective (i.e., PSG and actigraphy) and questionnaire-based sleep assessments have generally found that greater pain intensity and frequency is associated with greater sleep disruption (Valrie et al., 2013). In a

daily diary study of youth with SCD ages 8 to 12 years, investigators found that higher pain intensity predicted poor sleep quality that night, and, conversely, poorer sleep quality predicted higher pain intensity the following day (Valrie, Gil, Redding-Lallinger, & Daeschner, 2007). Yet, other diary studies of children with JIA and adolescents with idiopathic chronic pain have discovered that pain intensity did not predict sleep quality, but poor sleep quality did predict higher pain intensity the following day (Bromberg, Gil, & Schanberg, 2012; Lewandowski, Palermo, De la Motte et al., 2010). These findings may highlight the complex nature of sleep and pain. Moreover, each chronic medical illness is characterized by a unique clinical profile that may contribute to variation in findings.

Sleep and weight status/BMI. Pediatric obesity is considered a worldwide epidemic and has risen dramatically in the past several decades; recent estimates indicate that about 30% of children and adolescents in the United States are overweight or obese (i.e., BMI greater or equal to the 85th percentile; Center for Disease and Control and Prevention, 2011). Given the rise of obesity and its association with medical (e.g., type 2 diabetes, cardiovascular disease; Freedman, Dietz, Srinivasan, & Berenson, 1999; Ludwig & Ebbeling, 2001) and psychosocial risk factors (e.g., poorer health-related quality of life; Wardle & Cooke, 2005), increased attention has been paid to prevention and intervention efforts. Effective treatment approaches for pediatric obesity typically combine dietary, physical activity, and behavioral components to achieve reductions in body mass index (BMI; Oude et al., 2009). These treatment approaches are promising, yet, despite their effectiveness, they often do not yield enough weight losses to help children and adolescents achieve healthy weight status (Hart, Hawley, Kuhl, & Jelalian, 2013). Thus, identification of other modifiable behaviors associated with weight loss in children may be

imperative to enhance current treatment approaches (Hart et al., 2013).

Sleep has been identified as one such target. With the rise of pediatric obesity, there has been a parallel decrease in child and adolescent sleep quantity (Matricciani, Olds & Perkov, 2011). In support of this trend, meta-analyses of cross-sectional and longitudinal studies demonstrate that shorter sleep times in childhood is associated with a 58-89% increased risk of obesity (Cappuccio, Taggart, Kandala, & Currie, 2008; Chen, Beydoun & Wang, 2008) after controlling for race, ethnicity, age, sex, parent BMI, and birth weight (Cappuccio et al., 2008; Chen, Beydoun & Wang, 2008). Mechanisms underlying the sleep-obesity link are unclear, yet experimental research in adults suggests that strong associations between reduced sleep and weight gain may be due to neuroendocrine changes. For example, sleep restriction may lead to hormonal fluctuations, including decreased leptin (Spiegel et al., 2004; Teheri, Lin, Austin, Young & Mignot, 2004) and increases ghrelin (Teheri et al., 2004) that are associated with the regulation of hunger and appetite (Wren et al., 2001; Mars, De Graaf, De Groot, Van Rossum, & Kok, 2005).

Physical Functioning in Youth with SB

Adolescents with SB are at-risk for poor physical health, including increased rates of chronic pain and obesity. Pain is a common experience for adolescents (Stanford et al., 2008), and there is evidence to suggest that children, adolescents, and young adults with SB experience recurrent pain that may impact their health-related quality of life and psychological health (i.e., depression and anxiety; Oddson, Clancy, & McGrath, 2006; Bellin et al., 2010; Bellin et al., 2013). Clancy and colleagues (2005) found that about half of their sample (56%) of children and adolescents with SB (ages 8 to 19 years) experienced pain once per week or more often, which is

higher than prevalence rates of weekly pain reported in a population-based studies of TD youth (Stanford et al., 2008). Further, over half of the sample reported moderate to severe pain at one or more locations (Clancy, McGrath, & Oddson, 2005). The type and location of pain in youth with SB may be due to medical sequelae related to this condition. Specifically, youth may experience joint and musculoskeletal pain due to the long-term effects of spasticity and overusing certain muscle groups necessary for ambulation (e.g., shoulder pain due to wheelchair overuse, Marge, 1994), back and lower body pain due to tethered cord (Bowman, McLone, Grant, Tomita, & Ito, 2001), and headache and migraine due to intracranial pressure imbalances and shunt infection or malfunction (Stellman-Ward, Bannister, Lewis & Shaw, 1997).

Children and adolescents with SB are also at risk for obesity and overweight. Obesity rates (BMI-percentile greater than or equal to the 95th percentile) among individuals with SB have been assessed with a variety of methods and range widely from 37-58% in children and adolescents, with many more being overweight (BMI-percentile between the 85th and 95th percentile; Essner, Murray, & Holmbeck, 2014; Mita, Akataki, Itoh, Ono, Ishida, & Oki, 1993; van den Berg-Emons, Bussmann, Meyerink, Roebroek, & Stam, 2003). Causes of obesity in this population are not well-researched, yet non-ambulatory status, sedentary lifestyle, neuroendocrine disturbances related to hydrocephalus and Chiari malformation, and short stature have been identified as potential implicating factors (Dosa et al., 2009). Further, caring for a child with obesity in addition to having a physical disability may impose greater demands on the caregiver in performing activities of daily living or medical management tasks as well as increase health care costs (Young, Steele, Fehlings, Jutai, Olmsted & Williams, 2005).

Sleep and Psychological Functioning

Sleep disturbances are intricately connected to adolescent psychological functioning. Much of research to date has focused on associations between sleep disturbances and internalizing symptoms, yet emerging research has also supported links between sleep disturbances and broader areas of adolescent psychological health, including health-related quality of life.

Sleep and internalizing symptoms. Researchers have often noted the bidirectional nature of sleep and internalizing symptoms (i.e., symptoms of anxiety and depression); sleep disturbances may have a negative impact on mood, and subsequent emotional difficulties may interfere with sleep, thus creating a negative feedback cycle (Dahl & Lewin, 2002; Figure 2). Research has consistently demonstrated associations between disturbed sleep and internalizing symptoms in typically adolescents, such that sleep disturbances are associated with increased levels of anxiety and depression (Alfano, Zakem, Costa, Taylor & Weems, 2009; Moore et al., 2009; Roberts, Roberts, & Chen, 2001; Roberts, Roberts & Chen, 2002). Further, internalizing symptoms frequently co-occur with sleep disorders (e.g., insomnia; Johnson, Roth, & Breslau, 2006; Ohayon, Caulet, & Lemoine, 2012), and likewise, sleep disturbances are common in youth with mood disorders (e.g., depression and anxiety; Alfano, Ginsburg, & Kingery, 2007; Murray, Murphy, Palermo, & Clarke, 2012; see Chorney, Detweiler, Morris, & Kuhn, 2008 for a review). For example, adolescents with Major Depressive Disorder (MDD) often report symptoms of insomnia, poor sleep quality, and fatigue (Murray et al., 2012).

Although research is less substantial, the relation between sleep and internalizing symptoms has also been assessed in mixed samples of pediatric chronic conditions as well as

illness-specific samples. In a population-based study, Hysing and colleagues (2009) found that difficulties initiating and maintaining sleep predicted emotional difficulties in a mixed sample of youth with chronic illnesses (e.g., asthma, epilepsy, and SB). Sleep disturbances have also been shown to be predictive of internalizing symptoms in children with asthma (Fagnano et al., 2009), epilepsy (Becker, Fennell, & Carney, 2004), and chronic pain conditions (Palermo & Krista, 2005; Meltzer, Logan, & Mindell, 2005). Despite emerging evidence of the impact of sleep disturbances on internalizing symptoms in TD and pediatric medical populations, these associations have not yet been investigated in youth with SB.

Sleep and health-related quality of life. Although current investigations have found that sleep disturbances are associated with specific impairments daytime functioning (e.g., behavior problems, depression), there are relatively few studies that have investigated the association between sleep disturbances and more global measures of functioning such as health-related quality life (HRQOL). Existent literature has tended to evaluate this association within the context of pediatric sleep disorders, finding that youth with SDB and insomnia are at-risk for significantly greater decrements in physical and psychosocial HRQOL (Hart, Palermo & Rosen, 2005; Rosen, Palermo, Larkin & Redline, 2002). Even youth with mild to moderate symptoms of SDB may be at-risk for reduced physical HRQOL (Rosen et al., 2002), and investigators have theorized that physiological sleep disorders such as SDB may impact sleep quality that is important for promoting overall health and functioning.

Other research has started to investigate the relationship between sleep and HRQOL in youth with chronic illnesses. In a sample of adolescents with three chronic pain conditions (juvenile idiopathic arthritis, headache, and sickle cell disease), a range of sleep disturbances

were associated with decrements in HRQOL (Palermo & Kiska, 2005). Sleep disturbances showing robust associations with HRQOL included daytime sleepiness or fatigue and sleep/wake problems (e.g., irregular sleep habits, prolonged sleep latency, and difficulties getting up in the morning; Palermo & Kiska, 2005). This emerging body of research suggests the importance of including HRQOL as an outcome domain in pediatric sleep research and investigating the link between sleep and HRQOL in other chronic illness samples.

Psychological Functioning in Youth with SB

Prevalence rates of internalizing symptoms (i.e., depression, anxiety) rise dramatically in adolescence (Garber, 2002), and likewise, youth with SB may have an elevated risk of internalizing symptoms during the adolescent period (Appleton, 1997; Essner & Holmbeck, 2010) that extends into young adulthood (Bellin et al., 2010). Indeed, adolescent-onset depression has been associated with an increased risk for recurrent episodes of depression in adulthood (Graber, 2004), necessitating research on factors that contribute to internalizing symptoms in adolescents with SB. Further, children and adolescents with SB may have impaired physical and psychosocial HRQOL compared to TD and chronically ill pediatric populations (Cate, Kennedy & Stevenson, 2002; Murray, Holmbeck, Ros, Flores, Mir, & Varni, in press).

Sleep and Family Functioning

As is true of many of the links between sleep and bio-neuropsychosocial functioning discussed thus far, associations between sleep and family functioning are likely complex and reciprocal; a close, warm family environment may promote appraisal of safety and healthy sleep, and conversely, stress and conflict may lead to increased worry, rumination and disrupted sleep (Dahl & El-Sheikh, 2007). Just as conflict may disrupt sleep, child and adolescent sleep may also

impact the family environment (Meltzer & Westin, 2011). For example, adolescent sleep disturbances may increase the likelihood of emotional problems, thereby contributing to a more stressful and high-conflict home environment (Kelly & El-Sheikh, 2011). Cross-sectional work has supported relations between child and adolescent sleep disturbance and family functioning. El-Sheikh and colleagues (2006) found that decreased sleep quantity and quality as measured by actigraphy was associated with increased child-report of marital conflict. Congruent with this research, a study on Mexican-American adolescents found that self-reported sleep disturbances were associated with marital and parent-child conflict (McHale, Kim, Kan, & Updegraff, 2011). Longitudinal research has also indicated that disturbed sleep as measured by actigraphy predicted increases in marital conflict (Kelly & El-Sheikh, 2011). While research examining the potential impact of sleep on family functioning is in its infancy, available evidence suggests that family environment may be a highly relevant outcome of interest.

Family Functioning in Youth with SB

Research on family functioning in children and adolescents with SB has revealed variable results. Family relationships are particularly salient and influential social relationships for youth with SB, given that social isolation from peers is more common in this population than TD youth (Holmbeck et al, 2003). Generally, families of youth with SB may be at-risk for decreased levels of family cohesion, particularly those from lower socio-economic backgrounds (Holmbeck, Coakley, Hommeyer, Shapera, & Westhoven, 2002). Yet, research has also revealed similar levels of conflict in families of youth with SB compared to TD youth (Holmbeck et al., 2002) and, further, families of youth with SB do not evidence normative increases in family conflict as a function of pubertal development (Coakley, Holmbeck, Friedman, Greenley, & Thil, 2002).

These findings support a “resilience-disruption” view of family functioning (Costigan et al., 1997); the experience of having a child with SB may disrupt some aspects of family functioning, yet these families still demonstrate considerable resilience on other adjustment domains.

Families of youth with SB may be less cohesive while still experiencing lower levels of conflict.

Sleep and Neuropsychological Functioning

There is growing evidence that sleep disturbances and daytime sleepiness affects neuropsychological functions involving the prefrontal cortex (i.e., attention and executive function) in TD children and adolescents. The majority of literature on the neuropsychological consequences of sleep disturbances in children and adolescents has focused on the cognitive impact of experimental sleep restriction and deprivation (to induce subjective and physiological sleepiness) and sleep disorders (e.g., obstructive sleep apnea; Kheriandish & Gozel, 2006). This research has documented the association between sleep disturbances and a range of neuropsychological deficits, including attention, concentration, memory, and executive function (Kheriandish & Gozal, 2006). However, current studies have tended to focus on the neuropsychological impact of sleep disturbances on younger children, with much less attention paid to adolescent populations (Beebe et al., 2008). Attention and executive function are particularly important neuropsychological outcomes to study in adolescent populations; while disrupted sleep often manifests in young children as hyperactivity, inattention, and poor concentration, adolescents are more likely to exhibit symptoms of inattention and higher-order cognitive processes (i.e., executive dysfunction) than hyperactivity (O’Brien, 2013). It should be noted that executive function is a system of related constructs (i.e., planning, working memory, problem solving, inhibition, and mental flexibility) believed to be primarily located in the

prefrontal cortex (O'Brien, 2013). Further, prefrontal brain regions are believed to be particularly vulnerable to sleep deprivation (Dahl, 2002) and the prefrontal cortex may undergo a period of protracted development during adolescence (Blakemore & Choudhury, 2006). Such cognitive vulnerability, coupled with the knowledge that adolescents often obtain inadequate sleep, necessitate further studies that examine the impact of sleep disturbances on neuropsychological outcomes in during the adolescent developmental period (Beebe et al., 2008).

Overall, reduced sleep quantity as measured by experimental sleep restriction studies is associated with decrements in parent and teacher report of questionnaire-based inattention and executive dysfunction (Beebe et al., 2008; Fallone, Seifer, Acebo, & Carskadon, 2005; Moreau, Rouleau, & Morin, 2013), as well as behavioral inattention as measured by research assistants (Fallone, Acebo, Arnedt, Seifer, & Carskadon, 2001). Findings are variable for performance-based measures of inattention, with some research suggesting a link between sleep quantity and quality and performance-based inattention (Gruber, Wiebe, Montecalvo, Brunetti, Amsel & Carrier, 2011; Sadeh, Gruber & Raviv, 2002) while others have found no such association (Fallone et al., 2001; Meijer, Habekoth, Van Den Wittenboer, 2000).

Further, increased inattention and executive dysfunction has been documented in pediatric sleep disorders characterized by sleep fragmentation (i.e., increased night awakenings) and increased daytime sleepiness, including SDB, delayed sleep phase syndrome, and restless legs syndrome (Beebe et al., 2004; Giannotti, Cortesi, Sebastiani, & Ottaviano, 2002; Blunden, Lushington, Kennedy, Martin, & Dawson, 2000; Cortese et al., 2005). For example, children with mild SDB demonstrated diminished performance-based selective and sustained attention compared to controls (Blunden et al., 2000). Researchers have even suggested the presence of a

dose-dependent response in attention scales, such that children with moderate to severe OSA show greater attention deficits (Owens-Stively et al., 1997). Further, viewing the association between sleep and inattention from a developmental psychopathology perspective, sleep disturbances and daytime sleepiness are also common in children and adolescents with ADHD (Chervin, Dillon, Bassetti, Ganoczy, & Pituch, 1997; Cortese, Konofal, Yateman, Mouren, & Lecendreux, 2006; Gau & Chiang, 2009); an estimated 25 to 50% of children and adolescents with ADHD have difficulties initiating and maintaining sleep (Corkum, Tannock & Moldofsky, 1998). Researchers have speculated that youth with ADHD may have intrinsic factors associated with sleep problems, including a hyper-arousal mechanism preventing or delaying sleep onset (Dahl, 1999).

Neuropsychological Functioning in Youth with SB

Due in part to brain malformations characteristic of SB and the consequences of shunt placement at birth (i.e., shunt infections and replacement), the majority of youth with SB have mild to moderate neuropsychological impairments, such as executive dysfunction, inattention, and impaired intellectual functioning (Rose & Holmbeck, 2007). Youth with SB tend to exhibit attention problems that are similar to children with attention-deficit/hyperactivity disorder ADHD- inattentive type (e.g., high distractibility, poor organization of materials, and difficulty staying on task), and have higher rates of ADHD compared to their TD peers (Burmeister et al., 2005). Burmeister and colleagues (2005) reported a 31% prevalence rate of ADHD among youth with SB (the majority of whom were the inattentive type), compared to 17% of TD peers. However, there are distinct differences between the manifestation of attention problems in youth with SB compared to those with ADHD; children with SB show greater difficulty with posterior

brain attention processes, rather than the anterior attention deficits that are commonly associated with ADHD (Brewer, Fletcher, Hiscock, & Davidson, 2001). The anterior brain attentional system is related to sustaining and maintenance functions of attention, whereas the posterior system mediates attention functions related to focusing and shifting. In particular, youth with SB typically have difficulties with orienting to important information, focusing, and shifting attention (Dennis, Landry, Barnes, & Fletcher, 2006; Brewer et al., 2001; Rose & Holmbeck, 2007) but not sustained attention. Therefore, a child with ADHD may showcase more classic clinical symptoms of this disorder, such as difficulty attending to verbal and visual stimuli. In contrast, a child with SB may be able to attend to exogenous information, yet will require more time to disengage, shift, and orient to another stimulus.

Moreover, there are strong and consistent data to support the commonality of executive function deficits in youth with SB, which may include difficulties in the areas of cognitive flexibility, abstract reasoning, visual planning, sequencing, and switching (Burmeister et al., 2005; Fletcher et al., 1996; Hampton et al., 2011; Rose & Holmbeck, 2007; Snow, 1999; Tuminello et al., 2012) as well metacognitive tasks (i.e., task initiation, working memory, planning, organization, and self-monitoring; Brown et al., 2008; Zabel et al., 2011). Although executive function skills are believed to be controlled by frontal brain regions (Anderson, 1998), executive dysfunction in individuals with SB may be explained by anomalies in posterior and other brain areas (e.g., the cerebellum). In fact, the measurement of executive function is often complicated because other neuropsychological constructs that are known to be weaker in this population are implicated, including posterior attention system-mediated attentional shifting skills, as well as fine motor skills controlled by the cerebellum (Burmeister et al., 2005).

Further, it has been hypothesized that the under-arousal and disengagement in problem solving tasks typically seen in this population may be due to posterior controlled arousal-activation system. Yet, to date, the neurological origins of executive function skills in this population are not fully understood.

Sleep and Academic Functioning

Intricately related to neuropsychological functioning, research demonstrates that learning capacity and school performance (e.g., lower grades; Beebe, Rose, & Amin, 2010; Curcio, Ferrara, De Gannaro, 2006; Fallone, Owens & Deane, 2002; Wolfson & Carskadon, 2003) may be significantly impacted by poor sleep, increased sleep fragmentation, and late bedtimes and early awakenings (e.g., shortened sleep time). A recent meta-analysis found that three sleep domains (sleep quantity, sleep quality, and daytime sleepiness) had small but significant effects on children and adolescents' school performance (Dewald, Meijer, Oort, Kerkhof, & Bogels, 2010). Effect sizes were larger for daytime sleepiness and smaller for sleep quantity; smaller effects of sleep quantity may be due to each individual's differential sleep need and individual vulnerability to sleep loss (Dewald et al., 2010). While the neural mechanisms of the impact of sleep on neuropsychological performance are unclear, researchers have posited that short or disrupted sleep may reduce overnight brain activation that is necessary for optimal neuropsychological functioning; insufficient or poor quality sleep in adolescence may increase daytime sleepiness (thereby decreasing alertness) and impair executive function of the PFC, and consequently, learning capacity and school performance (Dewald et al., 2010).

Academic Functioning in Youth with SB

Finally, given that neuropsychological deficits are common in this population, it unsurprising that children with SB also have difficulties in certain academic areas, including arithmetic, reading comprehension, and spelling (see Wills, 1993 for a review). Holmbeck and colleagues (2003) found that preadolescents with SB, and particularly those from low SES homes, scored lower on measures of scholastic competence, several teacher-reported indicators of academic performance (e.g., school grades), and parent report of future educational aspirations compared to their TD peers. Such cognitive and academic struggles may also impact achievement of academic milestones, as emerging adults with SB report lower college attendance compared to their peers (41% vs 70%; Zukerman Devine, & Holmbeck, 2011)

The Current Study and Hypotheses

Despite emerging evidence of the prevalence and impact of sleep disturbances in pediatric populations, there are very few studies that have assessed sleep in youth with SB. Multi-method studies that comprehensively assess sleep-wake disturbances using objective and subjective sleep instruments are essential for characterizing the nature of these problems. The proposed research bridged a critical gap in knowledge by assessing sleep-wake disturbances in adolescents with SB, and examining the impact of sleep on adolescents' physical, psychological, family, and neuropsychological functioning (Figures 2 & 3). Sleep represents an important modifiable behavioral domain for diagnosis and intervention, and several researchers have advocated for behavioral sleep interventions to improve adolescent functioning (e.g., mood and attention symptoms; Dahl & Harvey, 2007; Mindell, Kuhn, Lewin, Meltzer & Sadeh, 2006; Gordon, King, Gullone, Muris, & Ollendick, 2007). Further, targeted interventions in childhood

and adolescence are particularly important due to the potential impact of sleep disturbances on emotional and cognitive development across the lifespan (Gregory, Caspi, Eley, Moffitt & O'Connor, 2005; Gregory & O'Connor, 2002; Johnson, Chilcoat, & Breslau, 2000). Knowledge gained from this study informs the development of treatments designed to improve overall health and well-being in adolescents with SB.

Part one (addressing Hypotheses 1-3) of this project is a two-group study design utilizing a comprehensive sleep assessment including objective (actigraphy) and subjective (questionnaire and daily diary) measures. The two matched groups include: (1) adolescents with SB, and (2) typically developing (TD) peers. Specifically, we plan to use a matched comparison group recruited from a separate NIH-funded study to assess group differences in sleep-wake disturbances. *Part two (addressing Hypothesis 4)* of this project assessed the impact of sleep-wake disturbances on bio-neuropsychosocial functioning *in the SB group only*. Primary outcome variables include Body Mass Index (BMI), pain, internalizing symptoms, health-related quality of life (HRQOL), family functioning, inattention/hyperactivity, executive function, and academic functioning.

Hypothesis 1

Subjective survey measures will indicate that adolescents with SB have worse sleep quality and sleep habits, greater pre-sleep arousal, increased problems with fatigue, and more symptoms of insomnia as compared to TD adolescents. Further, utilizing actigraphic recordings, adolescents with SB will demonstrate reduced sleep efficiency, shorter total sleep time, more time awake after sleep onset (i.e., nighttime awakenings), and longer daytime naps compared to TD adolescents.

Hypothesis 2 (Figure 2)

Daily diary and actigraphic measures will indicate that poorer sleep quality and greater sleep fragmentation (reduced sleep efficiency, shorter total sleep time, and more nighttime awakenings) will predict worse pain and mood the following day. The chronic physical, medical, cognitive, and social demands of a complex medical illness such as SB may impede youths' ability to cope with poor sleep quality, painful symptoms, and negative mood. Thus, it is likely that such symptoms will have a significant impact on daytime functioning for adolescents with SB in particular; in other words, it is expected that temporal associations between sleep quality, pain, and mood will be stronger for adolescents with SB as compared to TD adolescents.

Hypothesis 3 (Figure 2)

Conversely, daily diary and actigraphic measures will indicate that higher pain intensity and poorer daytime mood will predict worse nighttime sleep quality and greater sleep fragmentation. Similar to hypothesis 2, it is expected that associations will be stronger for adolescents with SB compared to TD adolescents.

Hypothesis 4 (Figure 3)

Greater sleep-wake disturbances will be associated with higher Body Mass Index (BMI) and greater pain (intensity and frequency), internalizing symptoms, family conflict, inattention/hyperactivity, and executive dysfunction in adolescents with SB. It is also expected that greater sleep-wake disturbances will be associated with lower health-related quality of life (HRQOL), family cohesion, and academic functioning (lower scholastic competence and school grades) in this population.

CHAPTER TWO

METHODS

Participants and Recruitment

This project included 74 adolescents and their parents, ages 12-18: (1) adolescents with SB (SB group; N = 37) and (2) typically developing peers (TD group; N = 37). Specific details regarding each group are outlined below.

SB Group

The sample of adolescents with SB were recruited from a pool of adolescents participating in a longitudinal NIH-funded research study led by Grayson Holmbeck, PhD: Chicago Healthy Adolescents Transition Study (CHATS). The overarching aim of the CHATS study is to assess family, peer relations, neuropsychological correlates, and psychological adjustment outcomes in youth as they progress from childhood to young adulthood. The subsample used for the current study were drawn from participants entering their third, fourth, or fifth data collection waves of the CHATS study. Inclusion criteria for the subsample were: 1) diagnosis of SB, including myelomeningocele, lipomeningocele, or myelocystocele; 2) 12 to 18 years of age; 3) proficiency in English or Spanish; 4) involvement of at least one parent; 5) cognitive ability to complete questionnaires; and 6) residence within 300 miles of the laboratory to allow for home visits. Participants in the SB group were matched across several demographic variables to the TD Group (see below for further details). The proposed project was integrated

into the CHATS study by adding a 10-day objective and subjective sleep assessment for 40 youth with SB meeting the above criteria. This study used measures of height/weight (to calculate BMI), pain, internalizing symptoms, HRQOL, family conflict and cohesion, inattention/hyperactivity, executive function, and academic performance that were already part of CHATS study procedures. During a regularly scheduled home visit, qualified youth were informed that a sleep study was added to the larger study and were invited to participate. Youth assent and parent consent was added to the study assent and consent forms under an “Additional Studies” section in which youth and parents mark “yes” or “no” to study participation. IRB-approval from Lurie Children’s Hospital and Loyola University Chicago was received for modifications of study procedures and consent forms. Participants received \$50 for participating in the optional 10-day sleep assessment. Further, consent from families was provided for health professionals and research assistants to conduct medical chart reviews for adolescents in the SB group.

A total of 51 eligible participants were invited to participate between October 2012 and July 2015. Of the 51 families, 9 participants declined to participate in the sleep assessment portion of the study due to lack of interest or time constraints and 5 declined from the larger, longitudinal study. Thus, the total analyzed cohort included 37 adolescents with SB. Adolescents who declined ($N = 14$) participation did not differ from those who accepted participation ($N = 37$) with respect to age ($t[49] = 1.69, p = .10$), sex ($\chi^2 [1] = .01, p = .98$), race (Caucasian vs. other; $\chi^2 [1] = .35, p = .55$) ethnicity (Hispanic vs. non-Hispanic; $\chi^2 [1] = .27, p = .60$), type of SB (e.g. myelomeningocele or other; $\chi^2 [1] = .01, p = .91$), or shunt status ($\chi^2 [1] = .61, p = .44$)

TD Group

The TD comparison group were recruited from an existing NIH-funded study, conducted by Dr. Tonya Palermo. The comparison sample was recruited as part of Dr. Palermo's larger, longitudinal study (one year in length with three data collection waves) comparing sleep-wake disturbances in adolescents with and without chronic pain (Palermo, Law, Churchill, & Walker, 2012); sleep assessment data collection procedures for this sample are complete. The cohort of typically developing adolescents (without chronic pain) were recruited in the Pacific Northwest via community advertisements. Inclusion criteria were: 1) 12 to 18 years of age; 2) proficiency in English; 3) absence of a serious co-morbid health condition (e.g., cancer, diabetes) or documented development delay; and 4) no current parent-reported psychiatric diagnoses. Participants in Dr. Palermo's study received \$50 for completing study procedures. Children of families from the TD group who declined participation did not differ from those who accepted participation with respect to age and gender (Palermo et al., 2011). An initial pool of 78 adolescents between the ages of 12-18 were available to facilitate matching procedures.

Matching Procedures

Adolescents in the SB group who agreed to participate in the sleep study were matched to the initial TD sample. Participants were matched utilizing a case-by-case sorting process, prioritizing the following sociodemographic variables: age, gender, ethnicity, and income level. A balanced selection approach was utilized; for example, when the best match for a 14-year-old Hispanic female with SB was a typically-developing 14-year-old Hispanic female with a *higher* family income, the next match was selected to correct the imbalance- i.e., a youth with SB that

has *higher* family income was paired with a TD youth that has a *lower* family income. See below (pg. 53 and Table 2) for results of matching procedures.

Procedures

The SB group completed actigraphic and subjective sleep procedures identical to those utilized in Dr. Palermo's study to ensure compatible comparison data. Parents and youth completed informed consent forms and subjective sleep measures (outlined below) which were added to existing study questionnaire packets. Participants received instruction in using the Actiwatch 64 (AW64) and completion of paper diaries. As previous research indicates that at least five nights of actigraphy recording are necessary to obtain reliable sleep measures (Acebo et al., 1999), participants in this study were asked to wear the Actiwatch for ten consecutive days to ensure participants wore the watch for at least five nights total. Actiwatch and paper diaries were mailed back to study staff using pre-paid envelopes, and periodic follow-up phone calls helped to ensure compliance. Dr. Tonya Palermo (project consultant) provided actigraphy equipment (including watches, hardware, and software) necessary for actigraphic assessment and coding procedures.

Measures

Demographic Information

Parents completed a demographics form to assess child's age, gender, ethnicity, race, as well as parent education, occupation, family annual income, and family structure.

Spina Bifida Medical Information

Medical records were reviewed to gather the following SB medical information: type of SB (i.e., myelomeningocele, meningocele, or lipomeningocele), shunt status, and lesion level.

For this study, we analyzed lesion level as a continuous variable. Specifically, participants were assigned a score that ranged from 1 to 14, with lower numbers representing lower-level lesion. A score of '1' represented a lesion in the S3 region (the lowest lesion level among the group) whereas a score of '13' represented a T7 lesion (the highest lesion level among the group). In the absence of chart data, parent-report was used to assess type of SB (91.9% of data derived from chart review, 8.1% parent report) and shunt status (83.8% chart review, 16.2% parent report).

Parents of youth with SB completed a medical form to collect additional medical information on their child's current prescription medications, number of shunt revisions (since birth), number of surgeries and hospitalizations (in the previous two years), and current gross motor functioning. Prescription medication information was re-coded to create a summary variable indicating the total number of different prescription medication types taken by each participant. For example, a participant taking an antidepressant and antispasmodic would be coded as having two prescription medication types total. To assess gross motor functioning, we utilized a modified version of *The Gross Motor Function Classification System Expanded and Revised* (GMFCS-E&R) developed by Palisano, Rosenbaum, Bartlett, & Livingston (2007). Specifically, we gathered information from mother-report on a Medical History Questionnaire (or father-report in the absence of mother data) to assign participants a gross motor classification scale level based on the GMFCS-E&R: Level I: No braces, crutches, walker, or wheelchair (i.e., 100% unassisted walking), Level II: Uses braces, crutches, or walker, Level II: Some wheelchair use, but able to walk with braces (> 50% walking), and Level IV: Uses wheelchair at school and/or long outings, may walk for short distances with a walker (< 50% walking).

Finally, The Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999) from the first data collection wave was used as a proxy for general intellectual functioning. The WASI includes tasks within the performance and verbal domains, and is frequently utilized to provide an intelligence quotient (IQ). Specifically, the Vocabulary and Matrix Reasoning subtests were administered to participants in the present study to obtain an estimate of IQ. The Vocabulary subtest is a 42-item measure that assesses for expressive vocabulary, verbal knowledge, and fund of information. In addition, it is a reliable measure of crystallized intelligence and general intelligence (e.g., Wechsler, 1999). On items one through four, the examinee is required to name pictures (e.g., bucket). On items five through 42, words are orally and visually presented, and the examinee is required to provide a definition (e.g., what is a car?). The Matrix Reasoning subtest assesses nonverbal abstract problem solving, inductive reasoning, and spatial reasoning skills. In addition, it has been found to be a reliable and valid measure of fluid intelligence, correlating .81 with another common measure of fluid intelligence (Wechsler, 1999). Matrix Reasoning consists of 35-items, each consisting of an incomplete pattern. The examinee is asked to complete the pattern by selecting the best choice from five options. In general, higher scores on these measures represent higher levels of intellectual abilities. Standardized norms for both of these subtests have been obtained across 2,245 individuals aged six through 89, and average test-retest reliability coefficients of .89 (Vocabulary) and .92 (Matrix Reasoning) were obtained for children 6 to 16 years old (Wechsler, 1999).

Pubertal Status

Parent report of pubertal status was measured with the Pubertal Development Scale (PDS; Petersen, Crockett, Richards, & Boxer, 1988). Responses on the PDS range from 1 to 4

and were scored and categorized such that higher scores indicated more advanced pubertal status. For the SB and TD samples, internal consistency was adequate for boys (SB: $\alpha = .90$; TD: $\alpha = .74$) and slightly lower for girls (SB: $\alpha = .66$; TD: $\alpha = .71$).

Objective and Subjective Sleep-Wake Variables

An objective and subjective sleep assessment was conducted for this study; ambulatory actigraphic recordings was completed over 10 days, and adolescents completed a daily diary and several sleep questionnaires. Table 1 displays all sleep variables by measurement category.

Objective Sleep Variables (Actigraphy Monitoring)

The Actiwatch 64 (AW64; Phillips Respironics) was used to assess sleep patterns. The small, lightweight actiwatch is a wristwatch-like device that records sleep-wake patterns by monitoring movement (or lack of movement) during the daytime and nighttime (See Figure 4). Movement is “sensed” by an omni-directional mercury switch that opens when movement is detected. The AW64 is not recommended for diagnosis of physiological sleep disorders (e.g., sleep disordered breathing); however, it is considered a highly effective, reliable, and unobtrusive tool for examining day-to-day variation in sleep quality and quantity, with 85-95% agreement rates with polysomnographic sleep studies (Gruber & Sadeh, 2004). Actigraphy data was stored and scored using computer software (Actiware 5.0; Webster, Kripke, Messin, Mullaney, & Wyborney, 1982) and a manualized coding procedure. Dairies (see below) were also used as a tool to facilitate coding of actigraphic sleep patterns, such as to confirm whether a period of inactivity/low activity is due to a participant removing the actiwatch or taking a nap during the daytime.

Table 1. Sleep-Wake Variable Definitions

Sleep-Wake Variables	Definition
<u>Actigraphy</u>	
Total Sleep Time	Sleep quantity, or time spent sleeping between sleep onset and offset (reported in minutes or hours)
Wake After Sleep Onset (WASO)	Number of minutes scored as awake between sleep onset and sleep offset
Sleep Efficiency	[Total sleep time ÷ time in bed] x 100 (expressed as a percent)
Sleep Variability	Variability in total sleep time (SD of total sleep time/ total number of nights) for each individual
Nap Duration	Number of minutes spent sleeping during the daytime (predetermined minimum nap duration ≥ 15 min)
<u>Diary Report</u>	
Sleep Quality	Daily self-report of sleep quality (0 = extremely poor to 10 = extremely good) derived from paper diary
<u>Questionnaire Report</u>	
Sleep Quality	Self-reported sleep quality, derived from total score and subscales of the ASWS ^a
Insomnia Symptoms	Self-reported difficulty falling asleep and/or maintaining sleep 60% or more of the time (2 items of ASWS)
Pre-sleep Arousal	Self-reported arousal before sleep onset, derived from total score and subscales ^b of the PSAS
Sleep Habits	Self-reported sleep-facilitating or –inhibiting practices, derived from total score and subscales of the ASHS ^c
Sleep-Disordered Breathing	Parent-reported symptoms of sleep-disordered breathing. Total score ≥ 0.33 on PSQ ^d suggestive of SDB
Fatigue	Self-reported daytime fatigue, derived from total score and subscales of the PedsQL-MFS ^e

^aThe ASWS (Adolescent Sleep Wake Scale) includes a total sleep quality score as well as 5 subscales: going to bed, falling asleep, maintaining sleep, reinitiating sleep, and returning to wakefulness.

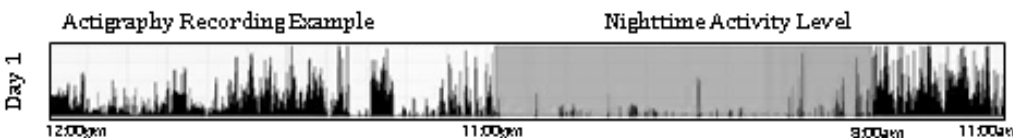
^bThe PSAS (Pre-sleep Arousal Scale) includes a total pre-sleep arousal score as well as 2 subscales: cognitive and somatic arousal.

^c The ASHS (Adolescent Sleep Hygiene Scale) includes a total sleep habits score and 6 subscales (physiological, cognitive, emotional, sleep environment, substances, and sleep stability. The physiological and subscales subscales were not included in this study due to low reliability.

^d The PSQ (Pediatric Sleep Questionnaire) *was administered to the SB group only* and includes a total score and three subscales: daytime sleepiness, snoring, and problem behaviors. The current study used the total score to provide descriptive data on symptoms of sleep-disordered breathing (SDB) and the *snoring* and *daytime sleepiness* subscales in multivariate analyses (i.e., Hypothesis 4).

^eThe PedsQL-MFS (PedsQL Multidimensional Fatigue scale) includes a total score and three subscales: general, sleep/wake, and cognitive fatigue. Higher scores indicating less difficulties with fatigue.

Figure 4. Actigraphy Recording Example



Utilizing these scoring methods, the following sleep parameters were obtained: (1) *total sleep time*, or duration of time spent sleeping while in bed; (2) *wake after sleep onset (WASO)*, or the total number of minutes scored as awake during total sleep time period; (3) *sleep efficiency*, or the ratio of estimated total sleep time and time in bed expressed as a percent ($[\text{total sleep time} \div \text{time in bed}] \times 100$) with values closer to 100 representing good sleep efficiency; and (4) *nap duration* or number of minutes spent sleeping during the daytime. Further, to assess night-to-night variation in sleep patterns we also calculated (5) *sleep variability* or the variability in total sleep time, by computing the standard deviation in total sleep time over the total number of nights for each individual.

The study investigator coded the length of daytime naps (average of all naps in minutes, minimum nap duration of 15 minutes) by assessing participant actigraphy data and using daily diaries for verification and validation. Nap data was coded if present in noted in the daily diary and confirmed using daytime actigraphy records. Actigraphy has been shown to be a valid sleep assessment tool in youth and adults with severe neurological disorders and mild mobility impairments (Scheer, Zeitzer, Ayas, Brown, Czeisler, & Shea, 2005; Zollman, Cyborski, & Duraski, 2010). As recommended by researchers utilizing actigraphy in individuals with lower limb mobility impairments (Zollman, Cyborski, & Duraski, 2010), the actigraph was worn on the wrist or upper arm to ensure valid actigraphic data collection. The graduate student investigator scored all actigraphy data using manualized coding procedures.

Subjective Sleep-Wake Variables

Sleep Quality

Diary-reported sleep quality and diary assessment procedures. The 10-day diary was used to assist with actigraphy coding procedures and assess the adolescent's daily subjective perception of *sleep quality*, as well as *mood* and *pain level*. Each morning, participants answered questions pertaining to: quality of sleep the previous night (Likert scale; 0 = extremely poor to 10 = extremely good), mood (Likert scale; 0 = extremely poor to 10 = extremely good), as well as bedtime/wake time, sleep latency, and number and duration night awakenings. Each evening, participants reported on pain intensity (Likert scale; 0 = No pain at all to 10 = Worse pain), daytime napping, and if/when the actiwatch was removed. *Daily perception of sleep quality, mood, and pain were only included in HLM analyses testing temporal associations between sleep, mood, and pain (i.e., Hypotheses 2 and 3).*

Questionnaire-reported sleep quality. Adolescents completed the Adolescent Sleep Wake Scale (ASWS; LeBourgeois et al., 2005). The ASWS is a 32-item self-report scale that assesses self-perceived quality of sleep across five behavioral dimensions: going to bed, falling asleep, maintaining sleep, reinitiating sleep, and returning to wakefulness. Items assess the occurrence and frequencies of these sleep behaviors in the past month along a 6-point scale (1 = never to 6 = always). The ASWS has demonstrated adequate internal validity and consistency in adolescents (LeBourgeois et al., 2005). Higher scores on the ASWS represent better sleep quality. In the current sample, internal consistency was adequate for the total scale (SB: $\alpha = .84$; TD: $\alpha = .83$) and majority of the subscales (SB: α s = .62- .72; TD: $\alpha = .64$ -.78). Internal consistency for the Falling Asleep subscale was inadequate for the SB group ($\alpha = .56$, TD group:

$\alpha = .74$), thus one item (“When it’s time to go to sleep, I feel sleepy”) was dropped to improve reliability (SB: $\alpha = .66$; TD: $\alpha = .79$).

Insomnia Symptoms

Insomnia symptoms were evaluated using two items from the ASWS (described above) assessing difficulty falling asleep (i.e., “I have trouble going to sleep” item from the falling asleep subscale) and difficulty maintaining sleep (i.e., “After waking up during the night, I have trouble going back to sleep” item from the reinitiating sleep subscale). Adolescents who responded “quite often,” “frequently, if not always,” or “always” (i.e., 60% of the time or more often) to either of these items were judged to have symptoms of insomnia. Previous research has used similar methods to assess for insomnia symptoms in adolescents (Palermo et al., 2011).

Pre-Sleep Arousal

Adolescents reported on pre-sleep arousal using the 16-item Pre-Sleep Arousal Scale (PSAS; Nicassio, Mendlowitz, Fussell, & Petras, 1985), which measures somatic (e.g., tense muscles, pounding heart) and cognitive arousal (e.g., heart palpitations, racing thoughts) prior to sleep onset during a typical week. Respondents rate the presence and intensity of these symptoms using a 5-point scale (1 = not at all to 5 = extremely). Higher scores on the PSAS indicate greater arousal before falling asleep. In the current sample, internal consistency was adequate for the somatic subscale (SB: $\alpha = .93$; TD: $\alpha = .85$), cognitive subscale (SB: $\alpha = .91$; TD: $\alpha = .83$), and total scale (SB: $\alpha = .91$; TD: $\alpha = .83$).

Sleep Habits

Sleep habits were measured with the Adolescent Sleep Hygiene Scale (ASHS; LeBourgeois et al., 2005). The ASHS is a 24-item self-report scale that assesses sleep-facilitating

and sleep-inhibiting habits or practices across six conceptual dimensions: physiological, cognitive, emotional, sleep environment, substances (e.g., caffeine), and sleep stability. Items are scored on a 6-point scale (1 = always to 6 = never), with higher scores indicating better sleep habits. Internal consistency was also adequate for the total scale (SB: $\alpha=.87$; TD: $\alpha = .82$) and the emotional (SB: $\alpha=.85$; TD: $\alpha = .78$) and sleep environment subscales (SB: $\alpha=.69$; TD: $\alpha = .78$) of this measure. Reliability was inadequate for the TD group on the cognitive ($\alpha = .50$) and sleep stability subscales ($\alpha = .58$). Dropping one item on the cognitive subscale (“I use my bed for things *other than sleep*, for example, talking on the phone...”) and one item on the sleep stability subscale (“During the school week, I ‘sleep in’ more than one hour past my usual wake time”) improved reliability (α s = $.62$ & $.67$, respectively). Internal consistency was inadequate for both groups on the physiological subscale (SB: $\alpha=.42$; TD: $\alpha = .55$), yet dropping items from the physiological subscale did not improve internal consistency. Further, both items on the substances scale had very little variance because the majority of adolescents (97.2%) endorsed never using alcohol or tobacco in the evening. Thus, the physiological and substance subscales were not included in study analyses.

Fatigue

Fatigue was assessed via adolescent self-report with the PedsQL Multidimensional Fatigue scale (PedsQL-MFS; Varni et al., 2004). This is an 18-item scale designed to assess the presence and severity of fatigue over the past month along three domains: general fatigue, sleep/rest fatigue, and cognitive fatigue. Much like the PedsQL Generic Scale, responses on the PedsQL-FS are on a five-point scale (0 = never to 4 = almost always). Items are reverse-scored and linearly transformed to a 0-100 scale, with higher scores indicating less difficulties with

fatigue. In the current study, internal consistency was adequate for the subscales (SB: $\alpha=.77-.87$; TD: $\alpha = .69-.83$) and total scale (SB: $\alpha=.91$; TD: $\alpha = .81$) of this measure.

Sleep Disordered Breathing and Daytime Sleepiness

For the SB group only, one caregiver completed the 22-item sleep disordered breathing scale of the Pediatric Sleep Questionnaire (PSQ; Chervin et al., 2000). The PSQ is frequently used in sleep clinics to assess symptoms of sleep-disordered breathing and related symptoms when polysomnography is not feasible, and has demonstrated adequate reliability and validity (Chervin et al., 2000). The 22-item total score is a composite scale that assesses symptoms of sleep-disordered breathing and also contains subscale items specifically related to snoring (4 items; e.g., “*Does your child snore more than half the time?*”), daytime sleepiness (4 items; e.g., “*Does your child wake up feeling unrefreshed in the morning?*”), and problem behaviors (6 items; e.g., “*Interrupts or intrudes on others*”) that can be a manifestation of a sleep disorder.

We utilized the total score of this measure to provide descriptive data on symptoms of sleep-disordered breathing (SDB) due to the high incidence of this disorder. SDB scores are calculated as the proportion of symptoms present; a score of 0.33 or greater has been determined to be suggestive of SDB (i.e., 33% of 22 questions or more answered positivity; Chervin et al., 2000). We also chose to utilize the snoring and sleepiness subscales of the PSQ as predictor variables in multivariate analyses (to test ***Hypothesis 4***) instead of the total score of the PSQ, which contains several items that overlap with the study’s outcome measures including BMI (i.e., “*Is your child overweight?*”) and inattention (“*This child is often easily distracted by extraneous stimuli*”). Internal consistency was adequate for the total Sleep Disordered Breathing score ($\alpha = .71$) and Sleepiness subscale ($\alpha = .65$). Internal consistency of the 4-item Snoring

subscale was inadequate ($\alpha = .45$), thus one item (“*While sleeping, does your child have ‘heavy’ or loud breathing?*”) was dropped to improve reliability ($\alpha = .71$).

Bio-neuropsychosocial Outcome Variables (SB Group Only)

Diary-reported mood and pain level (see above) were the only two bio-neuropsychosocial variables collected for the TD and SB samples to test *Hypotheses 2 & 3* utilizing HLM analyses. The following bio-neuropsychosocial variables were collected *in the SB group only* in multivariate analyses testing sleep-wake variables as predictors of bio-neuropsychosocial outcomes (*Hypothesis 4*).

Forming Composite Measures

Study investigators created composites that included multiple reporters of bio-neuropsychosocial outcomes, which has been recommended as a useful multisource data management technique to decrease the number of analyses and reduce the possibility of shared method variance (Holmbeck et al., 2002). Pearson correlations were run for measures with two or more reporters (e.g., mother-, father-, teacher-, child-report) to assess associations between reporters of the same construct. Reports were averaged to create composite scores if at least moderately correlated ($r_s > .40$).

Body Mass Index (BMI)

Weight status was measured using Body Mass Index (BMI) z-scores. BMI is obtained by dividing the child’s weight from his or her height squared (i.e., kg/m^2). A BMI z-score was then computed based on tables published in 2000 by the U.S. Centers for Disease and Control Prevention. BMI z-scores take into account developmental changes in body composition as well as variations by age and gender (Kuczmarski et al., 2002). Height and weight was obtained

through chart review, or parents' written estimates of their child's height and weight when chart data was unavailable.

Pain

The current study assessed youths' report of pain intensity and frequency over the last three months. Pain intensity was rated using a Visual Analogue Scale (VAS), which has demonstrated adequate reliability and validity in children and adolescents (Cohen et al., 2008). Participants marked the point along a 10-cm line that most accurately represented their pain severity, with anchors ranging from "no pain" to "worst pain ever." Scores were calculated by measuring the distance from the lower end point of the scale to the mark made by the child. Pain frequency was rated on a 6-point Likert scale rating (0 = less than once per month to 5 = daily). Similar assessment of pain frequency has been utilized in previous research (e.g., Palermo, Valenzuela, & Stork, 2004).

Internalizing Symptoms

Parent and teacher responses to items on the Internalizing scale of the CBCL (Achenbach & Rescorla, 2001) were used to assess adults' perceptions of child internalizing symptoms. Additionally, adolescents reported internalizing symptoms using the Child Depression Inventory (CDI), a 27-item self-rated measure of depressive symptoms. Two items from the CDI that pertain to sleep ("I have trouble sleeping" and "I am tired") were dropped to reduce overlap between the sleep-wake variables and CDI scores. The CDI has been well validated in general populations, and has demonstrated adequate reliability in the current sample of adolescents with SB ($\alpha = .85$). Mother- and father-reports of internalizing were significantly correlated ($r = .45$) and averaged together to form a composite score. Child and teacher report internalizing scores

were not correlated with any other reporter ($r_s = .02 - .31$). Thus, we separately examined parent, teacher, and child report of internalizing.

Health-related Quality of Life

Youths' HRQOL was assessed using youth, mother, and father report on the Pediatric Quality of Life Scale (PedsQL™ 4.0 Generic Core Scales; Varni, Seid, & Kurtin, 2001). The PedsQL has well-established reliability and validity in children with both acute and chronic health conditions, and yields an 8-item physical scale and 15-item psychosocial scale. The psychosocial scale is comprised of three subscales: emotional (5 items), social (5 items), and school functioning (5 items). Youth and parents answered how much of a problem a given task had been over the past month using a 5-point Likert scale rating (0 = never a problem to 4 = almost always a problem). Raw scores were reverse-scored and transformed into standard scores that ranged from 0 to 100, with higher scores indicating better HRQOL. The proposed study used adolescent and parent-report of the psychosocial HRQOL total scale, which demonstrated adequate reliability in the current sample across reporters ($\alpha_s = .85 - .89$). One item ("I have trouble sleeping") from the PedsQL emotional subscale was dropped to reduce overlap between sleep variables and this subscale. The physical HRQOL scale of the PedsQL was not utilized in the current study as it contains items that may not be appropriate for youth with physical disabilities (e.g., "It is hard for me to run"; Oddson et al., 2006). Mother, father, and child reports of HRQOL were averaged given significant correlations between reporters ($r_s = .43 - .53$).

Family Functioning

The Family Environment Scale (FES; Moos & Moos, 1994) measures social and

environmental characteristics of the family and is completed by parents. The current study used Form R, which measures people's perceptions of their family environment. The FES includes three main dimensions (Relationship, Personal Growth, System Maintenance) comprising a total of ten subscales. For the purpose of this study, the cohesion and conflict subscales from the Relationship dimension were utilized. Examples of items on each subscale include: "there is a feeling of togetherness in our family" and "we fight a lot in our family," respectively. Because internal consistency has been low in some studies using the original true-false response format (Alderfer et al., 2008), this study used a four-point Likert-type scale to increase internal consistency and gather richer data about the family environment (1 = strongly disagree to 4 = strongly agree). Reliabilities for mother and father report on the conflict and cohesion subscales were adequate (α s = .71 - .83, respectively). Mean scores for mother and father reports of conflict and cohesion were averaged to create composite scores due to adequate correlations between reporters (conflict: $r = .64$; cohesion: $r = .51$).

Inattention/hyperactivity

The Swanson, Nolan, and Pelham Teacher and Parent Rating Scale version IV (SNAP-IV; Swanson et al., 2001) provides a dimensional scaling of the DSM-IV items for inattention, impulsivity, and hyperactivity. In the current study, the 18-item version of the SNAP-IV was used. The items are from the DSM-IV (American Psychiatric Association, 1994) criteria for Attention-Deficit/Hyperactivity Disorder (ADHD), and include two subscales of symptoms: inattention (items 1-9) and hyperactivity/impulsivity (items 11-19). The SNAP-IV is based on a four-point Likert rating scale (0 = not at all = 0 to 3 = very much). Subscale scores are calculated by averaging the item scores within the domains of Inattention and Hyperactivity/Impulsivity.

The SNAP-IV total score was used as a measure of teacher and parent-reported inattention/hyperactivity in the current study. Higher scores on the SNAP-IV total score represent greater inattention/hyperactivity. Internal consistency for the SNAP-IV total score was adequate across reporters (α s = .92 - .93). Mother-, father-, and teacher-report of the SNAP-IV total score were averaged together to form a single composite score due to significant correlations across reporters (r = .44 -.67).

Executive Dysfunction

The Behavior Rating Inventory of Executive Function (BRIEF; Gioia, Isquith, Gray, & Kenworthy, 2000) is a questionnaire measure of executive function that identifies eight sub-domains classified within two broader indexes: Behavioral Regulation Index (i.e., inhibit, shift, emotional control) and Metacognition Index (i.e., initiate, working memory, plan/organize, organization of materials, monitor). The BRI and MI are combined to obtain an overall Global Executive Composite (GEC) score. Higher scores on the BRIEF represent higher levels of executive dysfunction. The current study used the GEC to capture mother, father, and teacher report of youths' executive dysfunction, which demonstrated satisfactory internal consistency across reporters (α s = .97 - .98). Mother- and father-reports on the BRIEF-GEC were averaged given adequate correlations between reporters (r = .79). Parent- and teacher-report on the BRIEF-GEC were separately examined as teacher-report was not correlated with either mother or father report (r s = .31 & .16, respectively).

Academic Functioning

The scholastic competence subscale from the child, parent and teacher version of the Harter (1985) Self-Perception Profile for Children (SPPC) was used to assess academic

functioning. Internal consistency was adequate across reporters on the Harter scholastic competence subscale ($\alpha = .77 - .88$). Mother- and father reports on this subscale were significantly correlated ($r = .81$) and averaged together to form a composite score. Teacher report was not correlated with either mother or father report ($r_s = .19$ & $.21$, respectively). Therefore, we separately examined parent and teacher report on the scholastic performance subscale.

Academic functioning was also assessed using mother, father, and teacher reports of the grades on the adolescent's most recent report card. Grades were coded on a scale of 1 to 8, with higher scores indicating better grades (e.g., a score of 8 indicated *an excellent performance* or the letter-grade *A*, a score of 5 represented *a satisfactory performance* or the letter-grade *B/C*, a score of 1 indicated *an unsatisfactory performance* or the letter-grade *F*). Grade point averages were computed for four classes: science, social studies, English, and math. Grades across the four subjects were first averaged together for each of the three reporters owing to adequate reliabilities ($\alpha = .63-.85$). A composite score was then computed by taking the average of all three reporters due to adequate correlations ($r_s = .65-.90$).

Statistical Treatment

Preliminary Analyses

Analyses determined whether variables were significantly skewed. Descriptive statistics of sleep and diary data were summarized using mean values, frequency distributions, and cross-tabulations. Descriptive analyses also determined the degree of association among 1) objective and subjective sleep summary variables and 2) demographic covariates and sleep summary variables, and 3) SB medical variables and sleep summary variables.

Demographic and Medical Covariates

The following demographic covariates were chosen due to their potential relation to adolescent sleep and bio-neuropsychosocial outcomes: child's gender, age, pubertal status, and family income. During the adolescent period, females are more likely than males to develop depression (Nolen-Hoeksema, 2001) and chronic pain (Perquin et al., 2000). Developmental (e.g., increased social and academic pressures) and biological (e.g., puberty) may also influence sleep patterns in adolescents (Becker, Langberg, & Byars, 2015; Carskadon, Vierira, & Acebo, 1993), thus, chronological age and pubertal stage may be linked to increased sleep disruption. In addition, family income has emerged as a significant correlate of sleep disturbances in youth with chronic illnesses (Boergers & Koinis-Mitchell, 2010), as well as obesity and depression in the general adolescent population (Goodman, Slap, & Huang, 2003). Low SES may also be viewed as a stressor that disrupts parenting and family functioning; lower SES families of preadolescents with SB have been found to exhibit higher levels of parent-child conflict and less family cohesion (Holmbeck et al., 2002).

Medical covariates related to condition severity were used in regression analyses to test associations between sleep and bio-neuropsychosocial outcomes in the SB group only. These medical variables included: lesion level, total shunt revisions (since birth), surgeries and hospitalizations (total in previous two years), current gross motor functioning, total prescription medication types, and cognitive functioning (IQ). Condition severity has been associated with worse neuropsychological functioning in youth with SB, including lower scholastic competence and greater symptoms of inattention (Hommeyer et al., 1999). Further, youth with SB with greater condition severity often have more complex treatment regimens (e.g., catheterization, use

of medications) and a greater number of surgeries and hospitalizations that may disrupt sleep-wake cycles. A number of prescription medications have the potential to disrupt sleep-wake cycles in adolescents with SB, such as antidepressants, anticonvulsants, and antispasmodics (e.g., Oxybutynin; Diefenbach et al., 2003).

Analytic Plan Part 1 (Two-group Analyses; Hypotheses 1 - 3)

Actigraphic sleep variables were aggregated across nights 1 to 10 for each of the sleep parameters. First, MANCOVA/ANCOVA and chi-squared analyses tested for group differences in objective and subjective sleep (*Hypothesis 1*). MANCOVA analyses determined group differences in questionnaire report of sleep disturbances (total and subscales scores of the ASWS, ASHS, PSAS, and PesdQL-Fatigue). Separate ANCOVAs were run for each of the objective sleep scores due to mathematical dependency among several the actigraphy variables (Tabachnick & Fidell, 2007), for example, sleep efficiency scores are calculated from average total sleep times. The presence of insomnia symptoms (yes/no) was dummy coded (0 = no; 1 = yes) and chi-square analyses was utilized to examine group differences in rate of insomnia symptoms.

Daily non-aggregated diary (i.e., daily mood and sleep quality) as well as actigraphic data (i.e., total sleep time, efficiency, and minutes awake after sleep onset) were utilized to clarify the relationships among daily reports of sleep, mood, and pain (*Hypotheses 2 & 3*). These data were analyzed using hierarchical linear modeling (HLM; Bryk & Raudenbush, 1992) as this is an excellent technique specifically designed to test multiple time series data points nested within participants. Multilevel modeling offers a number of advantages. First, this technique is appropriate for data that have a hierarchical structure. Multilevel modeling also allows for the

simultaneous estimation of day-to-day within-person (or Level 1) effects and between-person (or Level 2) effects. Other advantages include ability to 1) handle missing data and serial dependency without imputing values for within-person (Level 1) missing data, 2) handle unequal numbers of observations for each participant, and 3) utilize observations rather than individuals as the unit of analysis, expanding the number of degrees of freedom and power. Participants with less than 4 consecutive daily diary data were dropped from these analyses.

To test whether poorer sleep quality and greater sleep fragmentation would predict worse pain and mood the following day (*Hypothesis 2*), we used a 1-day lagged multilevel modeling procedure (Model 1 and 2 below). In these first two models, sleep on a given day is hypothesized to predict changes in mood and pain from one day to the next. To rule out the possibility that any lagged effect of sleep may be due to initial level of the dependent variable, the previous day's mood and pain was included in the model as a control variable. In these first two models, the dependent variable is the change in mood or pain from day t to day $t + 1$. The analysis model for changes in mood and pain for each individual can be expressed by using the Level 1 equations as shown in Models 1 and 2 below. For example, in Model 1 (*Sleep predicts mood the following day*), $Mood_{jt+1}$ represents the change in person j 's mood score between day t and day $t + 1$; π_{0j} is a regression intercept representing the mean change in daily mood; π_{1j} is a partial regression slope representing an individual's mood on day t ; and e_{ti+1} is a residual component of change in mood.

Further, to test *Hypothesis 3* that poor daytime mood and more intense pain will predict worse nighttime sleep quality and fragmentation (Models 3 and 4 below), daily levels of pain and mood were included in the Level 1 equation as predictors of nighttime sleep. Because pain and

mood on a given day is hypothesized to predict changes in sleep that night, a 1-day lag model was not utilized for these models. The four models were as follows:

Model 1: Sleep predicts mood the following day

$$\text{Level 1: } \text{Mood}_{jt+1} = \pi_{0j} + \pi_{1j}\text{Mood}_{jt} + \pi_{2j} (\text{Sleep Parameter}_{jt}) + e_{ti+1}$$

$$\text{Level 2: } \pi_{0j} = \beta_{00} + \beta_{01}\text{Covariate1}_j + \beta_{02}\text{Covariate2} + \beta_{03}\text{Covariate3}_j + \beta_{04}\text{Covariate4}_j + \beta_{05}\text{GroupStatus}_j + r_{0j}$$

$$\pi_{1j} = \beta_{10}$$

$$\pi_{2j} = \beta_{20} + \beta_{21}\text{GroupStatus}_j$$

Model 2: Sleep predicts pain the following day

$$\text{Level 1: } \text{Pain}_{jt+1} = \pi_{0j} + \pi_{1j}\text{Pain}_{jt} + \pi_{2j} (\text{Sleep Parameter}_{jt}) + e_{ti+1}$$

$$\text{Level 2: } \pi_{0j} = \beta_{00} + \beta_{01}\text{Covariate1}_j + \beta_{02}\text{Covariate2} + \beta_{03}\text{Covariate3}_j + \beta_{04}\text{Covariate4}_j + \beta_{05}\text{GroupStatus}_j + r_{0j}$$

$$\pi_{1j} = \beta_{10}$$

$$\pi_{2j} = \beta_{20} + \beta_{21}\text{GroupStatus}_j$$

Model 3: Daytime mood predicts nighttime sleep

$$\text{Level 1: } \text{Sleep}_{jt} = \pi_{0j} + \pi_{1j} (\text{Mood}_{jt}) + e_{ti}$$

$$\text{Level 2: } \pi_{0j} = \beta_{00} + \beta_{01}\text{Covariate1}_j + \beta_{02}\text{Covariate2} + \beta_{03}\text{Covariate3}_j + \beta_{04}\text{Covariate4}_j + \beta_{05}\text{GroupStatus}_j + r_{0j}$$

$$\pi_{1j} = \beta_{10} + \beta_{11}\text{GroupStatus}_j$$

Model 4: Daytime pain predicts nighttime sleep

$$\text{Level 1: } \text{Sleep}_{jt} = \pi_{0j} + \pi_{1j} (\text{Pain}_{jt}) + e_{ti}$$

Level 2: $\pi_{0j} = \beta_{00} + \beta_{01}\text{Covariate1}_j + \beta_{02}\text{Covariate2} + \beta_{03}\text{Covariate3}_j + \beta_{04}\text{Covariate4}_j +$

$$\beta_{05}\text{GroupStatus}_j + r_{0j}$$

$$\pi_{1j} = \beta_{10} + \beta_{11}\text{GroupStatus}_j$$

In all of the study models, the Level 2 equation included all relevant covariates that represent characteristics of the participants. In particular, participant characteristics (age, gender, income, pubertal status) and were first entered into baseline multilevel models to determine the influence of between-person covariates on daytime mood and pain and nighttime sleep. Next, separate multilevel models were run to evaluate 1) sleep parameters (nighttime sleep efficiency, WASO, total sleep time, and diary rating of sleep quality) as predictors of mood and pain the next day (total of 8 models), and 2) daytime mood and pain as predictors of nighttime sleep parameters (total of 8 models), above and beyond the effects of covariates. Therefore, a total of sixteen models (four models with each of the four sleep parameters) were run for Hypothesis 2 and 3 of Analytic Plan Part 1

Finally, in order to investigate whether day-level relationships between sleep, mood, and pain varied as a function of person-level differences in group status (i.e., whether the individual had SB or was typically developing), coefficients from the day-level as described at Level 1 of model equations were analyzed at Level 2. Thus, study group and the interaction term (Study Group X Sleep Parameter, i.e., Study Group X Sleep Efficiency) were added to the models to evaluate group as a moderator of sleep, pain, and mood relationships, or person j 's Level 1 slope (π_{2j}) is predicted as a function of the intercept and group status. Interaction effects were trimmed from study models if non-significant.

Analytic Plan Part 2 (SB Group Analyses; Hypothesis 4)

The final aim of the current study was to identify the relationship between sleep-wake disturbances and bio-neuropsychosocial outcomes in adolescents with SB. These analyses were conducted for *the SB group only*. Separate linear regression analyses determined whether study independent variables pertaining to sleep-wake disturbances were associated with bio-neuropsychosocial dependent variables. Specifically, hierarchical multiple regression models examined objective and subjective sleep variables as predictors of the following dependent variables: BMI, pain intensity, pain frequency, internalizing symptoms, HRQOL, family conflict, family cohesion, inattention/hyperactivity, executive function, and academic performance. Independent variables were entered in the following order: (Step 1) demographic and medical covariates; (Step 2) actigraphic sleep variables; (Step 3) subjective sleep variables. Sleep variables (Step 2 and 3) were entered in the forward selection fashion to determine which sleep variables were the best predictors of bio-neuropsychosocial functioning.

Given that the relatively large number of potential objective and subjective sleep variables and the smaller sample size used in these single-group analyses presented significant power limitations, Pearson correlations were conducted to determine which demographic and medical covariates and sleep predictors would be included in multivariate regression models. The demographic, medical, and sleep variables that were significantly correlated ($p < .05$) with a bio-neuropsychosocial outcome were included in the corresponding regression model. Covariates, subjective sleep, and objective sleep variables were entered into separate steps. If more than one objective variable was correlated with a particular bio-psychosocial outcome variable, they were run in separate models (one objective variable at a time) due to mathematical

dependency among these variables. Models were trimmed to only include significant effects ($p < .05$). Using these methods and because several adult- and child-reported bio-neuropsychosocial variables were combined (as described in the Measures section above), a total of 16 regressions were run for part two of data analyses.

Power Analysis

Power analyses were conducted using G*Power 3 (Faul, Erdfelder, Lang, & Buchner, 2007). A power of .80 and an alpha of .05 were assumed for all power calculations. For analyses including both the SB and TD groups, a sample size of 74 participants were available for MANOVA analyses (*Hypothesis 1*) and 70 participants (4 were excluded, see page 54 below) for HLM analyses (*Hypotheses 2 & 3*). Regression analyses that utilized the SB group only (*Hypothesis 4*) included a sample size of 37 participants.

For a MANCOVA model with two groups and a maximum of 8 predictors/covariates (e.g., five sleep variables and three demographic covariates included), a sample of 70 is required to detect medium effect sizes ($f^2 = .25$). Thus, our sample size of 74 was deemed to be sufficient to detect medium effects for MANCOVA analyses (*Hypothesis 1*).

Power calculations are often complex for intensive longitudinal data analyses, and therefore power analysis for a two-tailed fixed multiple regression model was used as a conservative approximation. This approach for estimating power is likely an underestimation of power for fixed effects, as it does not take into account multiple data points that are nested within people. For a fixed linear multiple regression model with a maximum of 8 predictors (for the most complex hierarchal linear model), a sample size of 70 is required to detect medium effects

($f^2 = .25$; Hypotheses 2 & 3). Therefore, a sample size of 70 for combined group analysis was determined to be sufficient for Analytic Plan Part 1 (*Hypotheses 1-3*)

Finally, for a fixed linear multiple regression model with a maximum of 3 predictors, a sample of 37 is required to detect large effects ($f^2 = .35$; *Hypothesis 4*). Therefore, a sample size of 37 for single group analysis was determined to be sufficient to detect large effects for Analytic Plan Part 2 (*Hypotheses 4*).

CHAPTER THREE

RESULTS

Preliminary Analyses

Skewness and Outliers

Skewness and outlier analyses were run on covariates and non-aggregated and aggregated sleep and diary variables for each group and on biopsychosocial variables in the SB group only. Conservative alpha levels (.001) were employed to evaluate the significance of skewness and outliers, in which z-score values greater than 3.29 were considered significant. A conservative approach was taken in the attempt to retain data such that transformations were performed first to reduce the impact of outlier values. Outliers were retained if variable transformations were sufficient. However, if transformed variables continued to be significantly skewed, significant outliers were removed and data were re-transformed. Square root transformations were run first, followed by logarithm and reciprocal transformations, respectively.

Analyses revealed that the following covariates were significantly skewed: lesion level ($z = 3.74$), number of shunt revisions ($z = 4.07$), and hospitalizations ($z = 3.36$). The following non-aggregated/raw data were skewed for both groups: pain level (SB: $z = 12.54$; TD: $z = 14.71$), sleep efficiency (SB: $z = -15.85$; TD: $z = -14.59$), and WASO (SB: $z = 6.99$; TD: $z = 18.61$). Aggregated (averaged) sleep variables that were significantly skewed for the SB group only included the pre-sleep arousal total score ($z = 4.74$) and somatic subscale ($z = 5.90$). Aggregated sleep variables that were skewed for the TD group only included the pre-sleep arousal cognitive

subscale ($z = 3.51$), the adolescent sleep wake scale- reinitiating sleep subscale ($z = -3.80$), WASO ($z = 3.83$), and nap duration ($z = 3.41$).

The majority of variables were no longer significant skewed after transformation procedures (z -values < 3.29). Non-aggregated WASO and sleep efficiency variables remained significantly skewed for the TD group only ($z = 8.55$ & $z = 5.83$, respectively). Removal of significant outliers corrected the skew for non-aggregated sleep efficiency ($z = 0.56$) and WASO (TD: $z = 1.23$). Non-aggregated pain level for both groups continued to be significantly skewed after reciprocal transformation (SB: $z = -5.26$; TD: $z = -7.77$). However, outliers were retained for this variable as re-transforming this variable after their removal did not improve skew. Several variables required reversed scale scoring prior to transformation and are displayed (in tables) and interpreted (within text) in the opposite direction.

Descriptive Statistics

Demographic characteristics and matching information (Table 2). Complete demographic information is provided in Table 2. Participants included 37 adolescents with SB (56.8% female; M age = 16.1) and a matched group of 37 TD peers (59.5% female; M age = 16.0). The majority of participants in the total sample were Caucasian (70.3%) and the remaining identified as African American (18.9%); Asian American (2.7%), American Indian/Alaskan Native (1.4%) or “other” (6.8%). Further, 20.3% of the total sample identified as Hispanic/Latino. While a wide range of family incomes were represented in both samples, the median (50-59,000k) annual household income suggests a generally middle income sample. The majority of the sample (86.1%) was at the late- to post-pubertal stage of development. The groups were successfully matched on eight sociodemographic variables: the five primary

variables utilized in matching procedures (child age, gender, race, ethnicity, income level) and three additional variables (family structure, number of people in the household, pubertal status).

Table 2. Characteristics of the Study Samples

Characteristic	SB Group (N = 37)	TD Group (N = 37)	Statistical test	Notes.
Child age in years: <i>M (SD)</i>	16.1 (1.4)	16.0 (1.5)	$t(72) = 0.45^{\text{NS}}$	Pubertal status was measured using the combined gender score on the Pubertal Development Scale (possible range = 1 to 4). Marital status was collapsed to intact vs. not intact (i.e.,
Gender: <i>N (%)</i>				
Male	16 (43.2%)	15 (40.5%)	$\chi^2(1) = 0.06^{\text{NS}}$	
Female	21 (56.8%)	22 (59.5%)		
Child race: <i>N (%)</i>				
Caucasian	26 (70.3%)	26 (70.3%)	$\chi^2(1) = 0.00^{\text{NS}}$	
Other	11 (29.7%)	11 (29.7%)		
Child ethnicity: <i>N (%)</i>				
Hispanic	8 (21.6%)	7 (18.9%)	$\chi^2(1) = 0.03^{\text{NS}}$	
Non-Hispanic	29 (78.4%)	28 (75.7%)		
Not Reported/Missing	0 (0.0%)	2 (5.4%)		
Family income level				
Median level	6.0 (50-59,000k)	6.0 (50-59,000k)	$F(1) = 1.39^{\text{NS}}$	
Categories: <i>N (%)</i>			$\chi^2(3) = 5.77^{\text{NS}}$	
<\$29,000	10 (29.4%)	4 (10.8%)		
\$30,000 – 49,000	6 (17.6%)	8 (21.6%)		
\$50,000 – 69,000	3 (8.8%)	9 (24.3%)		
>\$70,000	15 (44.1%)	16 (43.2%)		
Family structure: <i>N (%)</i>				
Two-parent intact	25 (71.4%)	27 (73.0%)	$\chi^2(1) = 0.02^{\text{NS}}$	
Not intact	10 (28.6%)	10 (27.0%)		
People in household: <i>M (SD)</i>	4.5 (1.4)	4.6 (1.6)	$t(70) = -0.31^{\text{NS}}$	
Pubertal Development				
Total Score: <i>M (SD)</i>	3.3 (0.5)	3.2 (0.7)	$t(70) = 0.55^{\text{NS}}$	
Categories: <i>N (%)</i> ^a			$\chi^2(2) = 1.72^{\text{NS}}$	
Early pubertal	1 (2.9%)	4 (10.8%)		
Mid pubertal	2 (5.7%)	3 (8.1%)		
Late pubertal	19 (54.3%)	19 (51.4%)		
Post pubertal	13 (37.1%)	11 (29.7%)		

mother/stepfather, single-mother, separated, other). NS = non-significant. SB = Spina Bifida; TD = Typically Developing.

^aEarly and mid-pubertal status categories were combined to perform Pearson chi-square test

Medical characteristics. Information on a number of physical status/illness severity variables for the SB group was obtained from medical chart review and/or parent report when chart data were unavailable: (a) *SB type* (chart and parent): 91.9% myelomeningocele, 2.7% lipomeningocele, and 5.7% lipoma, (c) *shunt status* (chart and parent): 78.4% shunt, 21.6% no shunt (b) *spinal lesion level* (chart): average lesion level was approximately in the L4 region, (d) *gross motor functioning class* (GMFC; parent): 13.5% Level I, 24.3% Level II, 27.0% Level III, 32.4% Level IV (2.7% or $N = 1$ unknown), (f) *total number of shunt revisions* among those with shunts (parent; $M = 2.2$, $SD = 2.2$; range = 0-8), (g) *number of surgeries in previous two years* (parent; $M = 0.7$, $SD = 0.0$; range = 0-3), and (h) *current prescription medication* (parent): 8.1% antidepressants, 8.1% anticonvulsants, 48.6% antispasmodics, 8.1% stimulants, and 0% opioids. None of the adolescents in the TD group were prescribed any of the aforementioned medications.

Sleep disordered breathing. Symptoms of sleep-disordered breathing were also assessed in the SB group only. Habitual snoring, which was defined as snoring more than half the time while asleep, was reported in 5.4% ($N = 2$) of adolescents with SB. A score suggestive of sleep disordered breathing (SDB; $> .33$) was found in one fifth (21.6%) of the sample of adolescents with SB.

Sleep assessment completion information. All participants completed actigraphic monitoring ($N = 74$). However, 4 youth from the SB sample had incomplete or unusable diary data: 3 participants were diary non-completers and 1 had non-consecutive diary data. SB participants with complete ($N = 33$) and unavailable/unusable ($N = 4$) diary data were similar according to age ($t[35] = -.48$, $p = .64$), sex ($\chi^2 [1] = .08$, $p = .77$), race (Caucasian or other; $\chi^2 [1] = 1.90$, $p = .17$), ethnicity (Hispanic or Non-Hispanic; $\chi^2 [1] = 0.03$, $p = .86$) income ($t[35] =$

-.96, $p = .35$), type of SB (myelomeningocele or other; $\chi^2 [1] = 1.72, p = .19$), and shunt status ($\chi^2 [1] = .001, p = .97$). Pearson product correlations determined that demographic and medical characteristics were unrelated to sleep assessment completion rate (i.e., number of actigraphy days and number of diary days, $ps = -.23 - .33$).

SB participants completed an average of 8.9 days out of the possible 10 days of actigraphic monitoring (89% completion rate; *range* = 5–12) and TD youth completed an average of 9.2 days (92% completion rate; *range* = 6–11). Youth with SB with valid diary data completed an average of 18.2 diary entries (91% completion rate; *range* = 6–20) out of the possible 20 diary entries (2 entries per day for 10 days) during the data collection period, and TD youth completed an average of 18.4 diary entries (92% completion rate; *range* = 10–29). Data from 614 diary days and 607 actigraphy nights were available for analyses.

Associations among objective and subjective sleep summary scores (Tables 3 & 4).

There were significant correlations among actigraphic sleep and self-reported sleep variables by group. Both diary-report and questionnaire-report of sleep quality were included in these analyses. In the SB sample, lower average total sleep time was associated with lower diary-reported sleep quality ($r = .37, p = .03$). Greater variability in total sleep time (i.e., sleep variability) was associated with poorer self-reported sleep habits ($r = -.34, p = .04$) in this sample. For TD youth, greater sleep variability was associated with greater pre-sleep arousal ($r = .38, p = .03$) and longer average nap duration was associated with greater symptoms of insomnia ($r = .39, p = .02$).

There were also several significant associations found among the actigraphy sleep variables (see Table 3). Strong correlations were found among total sleep time, WASO, and

sleep efficiency across both groups, with shorter total sleep time associated with greater minutes awake after sleep onset (SB: $r = -.75$; TD: $r = -.55$; p -values $< .01$) and poorer sleep efficiency (SB: $r = .83$; TD: $r = .70$; p -values $< .01$). Greater sleep variability was also associated with longer daytime naps in the SB group ($r = .38$, $p = .03$).

Table 3. Bivariate Correlations among Demographic and Actigraphy Sleep Summary Variables by Group

Variables	1	2	3	4	5	6	7	8	9
1. Gender ^a	--	-.03	-.25	.28	-.22	.15	-.16	.23	.35*
2. Age	.07	--	-.01	.52*	.13	-.09	.10	.29	.27
3. Income ^b	.13	.03	--	.21	-.23	.10	-.13	-.08	.16
4. Pubertal Status	.71*	.20	.18	--	-.09	.07	-.11	.13	-.03
5. Total Sleep Time	.30	-.40*	-.11	.27	--	-.75*	.83*	-.18	-.21
6. WASO	-.55*	.31*	-.32	-.50*	-.55*	--	-.96*	.07	.09
7. Sleep Efficiency	.62*	-.30	.19	.51*	.70*	-.93*	--	-.14	-.09
8. Sleep Variability	.11	.31	-.26	.28	-.05	-.07	-.01	--	.38*
9. Nap Duration	-.18	.05	.09	.00	-.21	-.10	.02	-.05	--

Notes. The numbers above the diagonal comprise correlations in sample of adolescents with spina bifida (Ns range from 34 to 37) and the lower triangle comprises correlations in sample of typically developing adolescents (Ns range from 35 to 37); WASO = minutes awake after sleep onset; Sleep Variability = average variation in Total Sleep Time.

^aGender coded as 0 = male, 1 = female

^bSpearman's rho

* $p < .05$

Of note, there was a strong correlation between WASO and sleep efficiency (SB: $r = -.96$; TD: $r = -.93$; p -values $< .01$), likely due to mathematical redundancy between the two variables. While these two actigraphic variables have strong quantitative overlap, previous sleep research has included both variables due to their conceptual differences. Sleep efficiency is frequently reported in sleep research as an interpretable and meaningful metric of sleep quality or the amount of time adolescents spend sleeping while taking into account sleep latency and nighttime awakenings; however, WASO may be useful to specifically determine the average amount of time youth are awake during the nighttime. Thus, both measures were included in our

analyses. However, due to mathematical redundancy, these two variables were never included in the same analytic models (e.g. regression and MANOVA models).

Finally, there were significant correlations among subjective sleep disturbances in the expected directions (see Table 4; $r_s = -.75 - .72$, $p_s < .05$). However, for youth with SB, symptoms of insomnia were unrelated to sleep habits, pre-sleep arousal, and fatigue.

Table 4. Bivariate Correlations among Demographic and Subjective Sleep Summary Scores by Group

Variables	1	2	3	4	5	6	7	8	9	10
1. Gender ^a	--	-.03	-.27	.28	-.55*	-.16	-.17	.32	.13	-.42*
2. Age	.07	--	-.01	.52*	.14	.10	-.36*	-.07	-.14	.07
3. Income Level ^b	.13	.03	--	.21	.08	-.27	.05	-.12	.04	.27
4. Pubertal Status	.71*	.20	.18	--	-.05	-.22	-.34*	-.01	.11	-.03
5. Sleep Quality (Q)	-.01	-.10	.17	.03	--	.29	.39*	-.64*	-.44*	.72*
6. Sleep Quality (D)	-.14	-.04	-.08	-.02	.55*	--	.17	-.35*	-.07	.35*
7. Sleep Habits	.11	-.09	.09	.11	.72*	.46*	--	-.49*	-.25	.59*
8. Pre-sleep Arousal	-.05	.14	-.18	-.07	-.75*	-.46*	-.76*	--	.25	-.72*
9. Insomnia ^c	.03	.14	-.10	.05	-.61*	-.48*	-.42*	.39*	--	-.25
10. Fatigue ^d	.14	-.08	.01	-.02	.55*	.38*	.49*	-.59*	-.33*	--

Notes. The numbers above the diagonal comprise correlations in sample of adolescents with spina bifida (N_s range from 34 to 37) and the lower triangle comprises correlations in sample of typically developing adolescents (N_s range from 35 to 37); Q = Questionnaire report of sleep quality (i.e., the ASWS, Adolescent Sleep Wake Scale); D= Diary report of sleep quality averaged across diary days (Global Score 1-10).

^aGender coded as 0 = male, 1 = female

^bSpearman's rho

^cInsomnia coded as 0 = no, 1 = yes, derived from two items on the ASWS (sleep initiation and maintenance).

^dLower scores on the PedsQL Multidimensional Fatigue Scale indicate greater problems with fatigue

* $p < .05$

Across both groups, lower diary-reported sleep quality was associated with greater pre-sleep arousal (SB: $r = -.35$; TD: $r = -.46$; p -values $< .05$) and fatigue (SB: $r = .35$; TD: $r = .38$; p -values $< .05$). Lower diary-reported sleep quality was also associated with questionnaire-report of sleep quality (i.e., ASWS total score, $r = .55$, $p < .01$), insomnia symptoms ($r = -.48$, $p = .003$), and sleep habits $r = .46$, $p = .005$) in the TD group. For the SB group, snoring was

unrelated to any other subjective sleep measures ($ps > .05$), however, more symptoms of sleepiness were related to lower questionnaire-report of sleep quality ($r = -.34, p = .04$) and greater fatigue ($r = -.41, p = .01$).

Associations among demographic and sleep summary scores by group (Tables 3 & 4). Correlations among the demographic and sleep-wake variables were calculated to determine which demographic covariates would be included in Part 1 of study analyses. Results indicated that, for the SB group, female gender was associated with lower questionnaire-report of sleep quality ($r = -.55$), greater fatigue ($r = -.42$), and longer daytime naps ($r = .35$). Further, older age and more advanced pubertal status were associated with worse sleep habits ($r = -.36$ & $r = -.34$, respectively $ps < .05$) in this sample. For the TD group, female gender and more advanced pubertal status was associated with better sleep efficiency ($r = .62$ & $r = .51$, respectively) and fewer minutes awake after sleep onset ($r = -.55$ & $r = -.50$, respectively). Finally, older age was associated with shorter total sleep time ($r = -.40$) and more time awake after sleep onset ($r = .31$) in the TD sample. In sum, while females and those with more advanced pubertal status reported *worse subjective sleep* disturbances in the SB group, the opposite was true for the TD group (i.e., females and those with more advanced pubertal status showcased *better objective sleep*). However, older adolescent age was associated with greater sleep disturbances in both groups. Income was not significantly associated with sleep disturbances in either group ($ps > .05$). *Results of bivariate correlations indicated that the following demographic variables were included in Part 1 of study analyses (two-group study design): age, gender, and pubertal status.*

The sleepiness and snoring subscales of the PSQ were calculated for the SB group only (not included in Tables 3 & 4); bivariate associations among demographic characteristics and the

two PSQ subscales indicated that male gender and less advanced pubertal status was associated with more snoring symptoms ($r = -.39$ & $r = -.52$ $p = .002$).

Associations between medical and sleep summary scores in the SB group (Tables 5 & 6). Descriptive analyses indicated that the presence of a shunt was associated with longer average sleep time ($r = .37$; see Table 5). Further, more limited gross motor functioning was associated with worse fatigue ($r = -.46$) and greater sleepiness ($r = .44$; see Table 6).

Table 5. Bivariate Correlations among Medical Covariates and Actigraphy Sleep Summary Scores in the SB Group

Variables	1	2	3	4	5	6	7	8	9	10	11	12	13
1. Shunt Status ^a	--												
2. Lesion Level ^b	-.12	--											
3. Shunt Revisions ^c	.52*	-.03	--										
4. Surgeries ^c	.24	-.30	.27	--									
5. Hospitalizations ^c	.06	.04	.17	.58*	--								
6. GMF	.08	.47*	.04	.06	.30	--							
7. Rx Medication ^d	-.03	-.01	.23	.24	.34*	.10	--						
8. WASI	-.03	-.40*	.07	.22	.04	-.04	.04	--					
9. Total Sleep Time	.37*	.29	.04	-.12	-.06	.17	-.20	-.23	--				
10. WASO	-.13	-.16	.17	.19	.13	-.12	.25	-.08	.75*	--			
11. Sleep Efficiency	.17	.19	-.14	-.18	-.11	.13	-.21	-.01	.83*	-.96*	--		
12. Sleep Variability	-.06	.15	.08	.25	.20	.30	.23	-.01	-.18	.07	-.14	--	
13. Nap Duration	-.11	-.11	-.21	.04	-.32	.07	-.22	.07	-.21	.09	-.09	.38*	--

Notes. GMF = Gross motor functioning (higher scores indicate higher level of impairment); WASI = Wechsler Abbreviated Scale of Intelligence; WASO = minutes awake after sleep onset; Sleep Variability = average variation in Total Sleep Time.

^aShunt status coded as 1 = present; 0 = absent/no shunt

^bLesion level coded such that sacral lesions levels had lowest scores and thoracic had highest scores

^cTotal number; Shunt revisions are total in lifetime, surgeries and hospitalizations are total in previous two years

^dRx medications represent the total number of *types* of prescription medications taken (e.g., antidepressants, anticonvulsants, etc.)

* $p < .05$.

Hypotheses Testing

Overview of Part I (Addressing Hypotheses 1-3)

Part I of this project is a two-group study design utilizing a comprehensive sleep assessment including objective (actigraphy) and subjective (questionnaire and daily diary) measures. First, MANCOVAs and ANCOVAs first tested for group differences in objective and subjective sleep using aggregated actigraphic and subjective measures, respectively (*Hypothesis 1*). Next, multilevel regression models tested for relationships among daily reports of sleep, mood, and pain among the two groups using non-aggregated diary data (*Hypotheses 2 & 3*). All two-group analyses controlled for the effects of child age, gender, and pubertal status.

Group comparisons on objective and subjective sleep disturbances (Hypothesis 1)

Objective sleep-wake disturbances (Table 7). As hypothesized, there were significant differences on actigraphic assessment of sleep-wake patterns between adolescents with SB and typically developing (TD) adolescents. Adolescents with SB experienced significantly shorter total sleep time at 6.15 hours (368.9 min) per night on average ($SD = 1.11$ hrs) compared with 6.62 hours (397.0 min) per night in TD adolescents ($SD = 0.84$ hrs, $p = 0.04$). Exploratory chi-squared analyses found that almost half (43.2%) of adolescents with SB experienced extremely short sleep (defined as less than 6 hours), compared with 21.6% of TD adolescents ($\chi^2 [1] = 3.95$, $p < .05$). Youth with SB also evidenced greater sleep variability, such that variability in nightly total sleep time was greater compared to their peers.

Table 6. Bivariate Correlations among Medical Covariates and Subjective Sleep Summary Scores in the SB Group

Variables	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16
1. Shunt Status ^a	--															
2. Lesion Level ^b	-.12	--														
3. Shunt Revisions ^c	.52*	-.03	--													
4. Surgeries ^c	.24	-.30	.27	--												
5. Hospitalizations ^c	.06	.04	.17	.58*	--											
6. GMF	.08	.47*	.04	.06	.30	--										
7. Rx Medication ^d	-.03	-.01	.23	.24	.34*	.10	--									
8. WASI	-.03	-.40*	.07	.22	.04	-.04	.04	--								
9. Sleep Quality (Q)	.02	.05	.04	.04	-.03	-.25	-.22	-.12	--							
10. Sleep Quality (D)	.32	.02	.28	-.11	-.26	-.17	-.22	-.09	.29	--						
11. Sleep Habits	-.01	.05	.15	.01	.01	-.30	-.07	-.13	.39*	.17	--					
12. Pre-sleep Arousal	-.09	.06	-.01	.20	.26	.21	.12	.08	-.64*	-.35*	-.49*	--				
13. Insomnia ^e	.22	-.08	.11	-.07	-.04	.13	-.12	.08	-.44*	-.07	-.25	.25	--			
14. Fatigue ^f	.09	-.19	.18	.06	-.15	-.46*	-.09	.02	.72*	.35	.59*	-.72*	-.25	--		
15. Sleepiness ^g	-.19	.21	-.11	.03	.10	.44*	.08	.13	-.34*	-.29	-.21	.13	.08	-.41*	--	
16. Snoring ^g	-.25	.21	-.02	-.07	.13	-.05	-.33	-.13	.17	-.09	.04	.06	-.01	.04	-.10	--

Notes. GMF = Gross motor functioning (higher scores indicate higher level of impairment); WASI = Wechsler Abbreviated Scale of Intelligence; Q = Questionnaire report of sleep quality (i.e., the ASWS, Adolescent Sleep Wake Scale); D= Diary report of sleep quality averaged across diary days (Global Score 1-10).

^aShunt status coded as 1 = present; 0 = absent/no shunt

^bLesion level coded such that sacral lesions levels had lowest scores and thoracic had highest scores

^cTotal number; Shunt revisions are total in lifetime, surgeries and hospitalizations are total in previous two years

^dRx medications represent the total number of *types* of prescription medications taken (e.g., antidepressants, anticonvulsants, etc.)

^eInsomnia coded as 0 = no, 1 = yes, derived from two items on the ASWS (sleep initiation and maintenance).

^fLower scores on the PedsQL Multidimensional Fatigue Scale indicate greater problems with fatigue.

^gSubscales of the Pediatric Sleep Questionnaire; *calculated for the SB group only.*

Table 7. Between-Group Comparisons on Actigraphy Sleep Summary Scores

Sleep Variable	SB Group Mean (SD)	TD Group Mean (SD)	<i>F</i> -value	<i>p</i> -value	η^2
Total Sleep Time (min) [<i>M</i> hrs, min]	368.9 (65.6) [6 hrs, 9 min]	397.0 (50.3) [6 hrs, 37 min]	$F_{1,67} = 4.28^*$.042	.06
Sleep Efficiency (%)	75.0 (11.6)	83.3 (5.4)	$F_{1,67} = 15.35^{**}$	<.001	.19
WASO (minutes)	117.2 (62.3)	64.9 (26.3)	$F_{1,67} = 21.84^{**}$	<.001	.25
Sleep Variability	19.3 (8.4)	15.3 (6.6)	$F_{1,67} = 5.10^*$.027	.07
Nap duration (minutes)	44.8 (62.2)	44.2 (61.6)	$F_{1,63} = 0.001$.972	<.01

Notes. ANCOVAs were run for each objective score (vs. one MANCOVA) because several of the variables were mathematically dependent. Displayed means and *SD*s are non-transformed variables adjusted for covariates (age, gender, pubertal status). WASO = minutes awake after sleep onset; Sleep Variability = average variation in total sleep time; SB = spina bifida; TD = typically developing; η^2 = partial eta squared.

+*p* = .05-.09; **p* < .05; ***p* < .01

Further, the SB group also spent more time awake after sleep onset, averaging 1.95 hrs (117.2 minutes) per night on average (*SD* = 62.3) compared with a little over an hour (64.5 minutes) in TD adolescents (*SD* = 38.3; *p* < .001). Consequently, adolescents with SB had significantly poorer sleep efficiency (*M* = 75.0; *SD* = 11.6) compared with TD adolescents (*M* = 83.3, *SD* = 5.4; *p* < 0.001). Chi-squared analyses indicated that over half (56.8%) of adolescents with SB had poor sleep efficiency (defined as less than 80%) compared to 18.9% of TD adolescents ($\chi^2[1] = 11.26, p < 0.01$). There were no group differences in average daytime nap duration. Group differences in wake after sleep onset and sleep efficiency represented large effect sizes, while differences in total sleep time and sleep variability were medium effects.

Subjective sleep-wake disturbances (Tables 8 & 9). Table 8 presents group differences and adjusted means/standard deviations of questionnaire sleep-wake summary variables and Table 9 displays between-group comparisons on insomnia symptoms.

Table 8. Between-Group Comparisons on Questionnaire Sleep Summary Scores

Variable	SB Group Mean (SD)	TD Group Mean (SD)	<i>F</i> -value	<i>p</i> -value	η^2
Summary Scores					
			$F_{4,64} = 4.10^{**}$.005	.20
Sleep Quality (ASWS)	4.0 (0.7)	4.5 (0.6)	$F_{1,67} = 10.67^{**}$.002	.14
Sleep Habits (ASHS)	4.7 (0.7)	5.0 (0.5)	$F_{1,67} = 4.21^*$.044	.06
Pre-sleep Arousal (PSAS)	29.1 (12.6)	25.4 (6.5)	$F_{1,67} = 2.20$.143	.03
Fatigue (PedsQL- MFS)	64.5 (18.2)	76.7 (11.9)	$F_{1,67} = 12.14^{**}$.001	.15
Subscale Scores					
Sleep Quality (ASWS)			$F_{5,61} = 6.56^{**}$	<.001	.35
Going to bed	3.5 (1.0)	4.2 (0.9)	$F_{1,65} = 10.57^{**}$.002	.14
Falling asleep	4.2 (1.0)	4.7 (0.9)	$F_{1,65} = 6.14^*$.016	.09
Maintaining sleep	4.5 (0.9)	4.7 (0.7)	$F_{1,65} = 1.70$.196	.03
Reinitiating sleep	4.4 (0.9)	5.3 (0.7)	$F_{1,65} = 24.23^{**}$	<.001	.27
Returning to wakefulness	3.6 (1.4)	3.5 (0.9)	$F_{1,65} = <0.03$.884	<.01
Sleep Habits (ASHS)			$F_{4,61} = 1.35$.237	.08
Cognitive	4.0 (1.0)	4.2 (0.8)	$F_{1,64} = 0.25$.619	<.01
Emotional	4.9 (1.3)	5.1 (0.7)	$F_{1,64} = 0.66$.419	.01
Sleep environment	4.7 (1.1)	5.2 (0.8)	$F_{1,64} = 4.54^*$.037	.07
Sleep stability	3.5 (1.5)	4.0 (1.0)	$F_{1,64} = 2.08$.154	.03
Pre-Sleep Arousal (PSAS)					
Somatic	12.1 (5.6)	10.4 (2.6)	$F_{1,67} = 2.60$.112	.04
Cognitive	17.0 (8.1)	14.9 (4.8)	$F_{1,67} = 1.50$.226	.02
Fatigue (PedsQL- MFS) ^a			$F_{3,65} = 7.32^{**}$	<.001	.25
General	70.8 (19.9)	78.6 (13.0)	$F_{1,67} = 3.97^+$.050	.06
Sleep/Rest	60.7 (21.4)	70.6 (14.7)	$F_{1,67} = 5.42^*$.023	.08
Cognitive	62.0 (22.1)	80.9 (14.2)	$F_{1,67} = 20.83^{**}$	<.001	.24

Notes. Bolded lines represent omnibus MANCOVA results, with univariate follow-up analyses presented underneath. ANCOVAs were run for each subscale of the ASHS (vs. one MANCOVA) due to lower correlations among the subscales. Displayed means and SDs are non-transformed variables adjusted for covariates (child age, gender, pubertal status). SB = spina bifida; TD = typically developing; η^2 = partial eta squared. ASWS = Adolescent Sleep-Wake Scale; ASHS = Adolescent Sleep Hygiene Scale; PSAS = Pre-Sleep Arousal Scale; PedsQL- MF = Pediatric Quality of Life Inventory, Multidimensional Fatigue Scale.

^a Lower scores on the PedsQL Multidimensional Fatigue Scale indicate greater problems with fatigue

+*p* = .05 - .09; **p* < .05; ***p* < .01

Table 9. Between-Group Comparisons on Insomnia Symptoms

Insomnia Symptoms	SB Group N (%)	TD Group N (%)	χ^2	<i>p</i> -value
1) Difficulty falling asleep (>60% of the time)	7 (19.4)	7 (18.9)	.003	.955
2) Difficulty staying asleep (>60% of the time)	9 (25.7)	3 (8.1)	4.01*	.045
Either 1) or 2)	11 (31.4)	8 (21.6)	0.89	.345
Both 1) and 2)	5 (14.3)	2 (5.4)	1.62	.204

Notes. Insomnia symptoms were evaluated using two items from the ASWS assessing difficulty falling asleep and difficulty maintaining sleep. SB = spina bifida; TD = typically developing
* $p < .05$

The overall MANCOVA including questionnaire sleep-wake summary scores was significant ($F_{4, 64} = 4.10, p = .01$). As hypothesized, adolescents with SB reported significantly worse overall sleep quality (on the Adolescent Sleep Wake Scale; ASWS) and greater fatigue (PedsQL-Fatigue Scale; PedsQL-FS) compared with healthy adolescents, which were large effects ($ps < .001$; Table 8). Adolescents with SB also endorsed worse sleep habits on the Adolescent Sleep Habits Scale (ASHS) compared to their peers ($p = .04$), which was a medium effect. There were no differences in the pre-sleep arousal (PSAS) summary score between the two groups

The overall MANCOVA for the subscales of the ASWS was also significant ($F_{5, 61} = 6.56, p < .001$; Table 8). Compared with typically developing peers, adolescents with SB reported more difficulties with going to bed and reinitiating sleep, which were large effects. They also reported greater difficulties falling asleep ($p = 0.02$), which was a medium effect. Gender emerged as a significant covariate in the MANCOVA model ($F_{1, 61} = 2.99, p = .02$); compared to adolescent males, females had greater difficulties going to bed ($F_{1, 65} = 5.50, p = .02$), falling asleep ($F_{1, 65} = 10.48, p = .002$), reinitiating sleep ($F_{1, 65} = 5.59, p = .02$), and returning to wakefulness ($F_{1, 65} = 5.96, p = .02$).

Further, the overall MANCOVA for the subscales of the ASHS was not significant, indicating similarity between groups on sleep habits ($F_{4, 61} = 1.34, p = .24$). However, because adolescents with SB had significantly worse ASHS total scores in the overall MANCOVA including questionnaire summary scores, we proceeded to examine mean differences among individual subscales of the ASHS. As shown in Table 8, there was a significant group difference in mean levels of the sleep environment subscale of the ASHS, such that adolescents with SB reported worse sleep environments compared to TD peers ($p = 0.04$).

Further, the overall MANCOVA for the subscales of the PedsQL-FS was significant, indicating that adolescents with SB reported greater daytime fatigue compared with healthy adolescents ($F_{3, 65} = 7.32, p < .001$). Examining the individual subscales of the PedsQL-FS indicated that adolescents with SB reported more difficulties with sleep/rest ($p = 0.02$) and cognitive fatigue ($p < .001$) compared to their TD peers, which were medium and large effects, respectively. There was also a trend toward adolescents with SB reporting more difficulties with general fatigue ($p = .05$).

There were no differences in the pre-sleep arousal subscale scores (cognitive and somatic arousal before bedtime) between the two groups. However, pubertal development emerged as a significant covariate for somatic pre-sleep arousal ($F_{1, 61} = 2.96, p = .019$), such that more advanced pubertal development was associated with less somatic pre-sleep arousal.

As shown in Table 9, the prevalence of insomnia symptoms was evaluated using two items from the ASWS assessing difficulty falling asleep and difficulty maintaining sleep. Adolescents reporting either symptom were judged to have insomnia symptoms. Chi-squared analyses indicated that a similar proportion of adolescents with SB reported insomnia (31.4%)

compared to typically developing adolescents (21.6%; $\chi^2 = 0.89, p = .345$). However, when examining individual insomnia symptoms, difficulties staying asleep were reported at significantly higher rates in adolescents with SB (25.7%) compared to their TD peers (8.1%; $\chi^2 = 4.01, p = .04$). There were no group differences in self-reported difficulties falling asleep.

Daily Associations among Sleep, Mood, and Pain (Hypotheses 2 & 3)

Descriptive daily actigraphy and diary information. Descriptive analyses were performed on non-aggregated (multilevel or raw) actigraphy and diary data. Across all diaries, the average sleep quality ratings were moderate ($M = 7.4$ [out of 10], $SD = 2.0$, range = 1.0 - 10) average mood ratings were positive ($M = 6.8$ [out of 10], $SD = 2.2$, range = 0.0- 10), and average pain intensity was mild ($M = 1.1$ [out of 10], $SD = 2.0$, range = 0 - 10). Youth with SB reported sleep quality in the lower range (1SD below the mean of sleep quality, i.e., < 5.4 , reflecting poorer sleep quality) on a similar number of observations as their TD peers (15.0% vs. 18.7% of days; respectively; $\chi^2 [1] = 1.59, p = 0.21$). Adolescents with SB also reported mood in the lower range (1SD below the mean mood, i.e., < 4.6 , reflecting more negative or lower mood) and on a similar percentage of diary days as the TD participants (16.7% vs. 12.8% of days, respectively; $\chi^2 [1] = 1.94, p = 0.16$). Finally, youth with SB reported pain on a similar number of diary days (36.0%) compared to their TD peers (29.4%) of days during the study period ($\chi^2 [1] = 3.13, p = 0.08$), and average pain ratings on days in which pain occurred was largely equivalent and in the mild range across the the two groups ($M = 3.36$ vs. $M = 3.74$ in SB and TD youth, respectively; $t(204) = -1.34, p = .18$).

Descriptive analyses of non-aggregated actigraphy data indicated that adolescents with SB and TD adolescents experienced a higher percentage of nights of extremely short total sleep

time (<6.0 hrs sleep; SB: 44.9%; TD: 30.7%, $\chi^2 [1] = 13.51, p < 0.001$) and low sleep efficiency (i.e., less than 80%; SB: 49.0% vs. TD: 24.3%, $\chi^2 [1] = 40.62, p < 0.001$). Youth with SB also experienced time awake after sleep onset (WASO) in the upper range (>1SD above the mean WASO, i.e., > 158.7 minutes) on a higher percentage of (27.1%) compared to 1.5% of TD youth, $\chi^2 [1] = 85.42, p < 0.001$.

Baseline multilevel model analyses. Participant characteristics (group, age, gender, income, pubertal status) and the previous' day mood or pain variable (when applicable) were entered into baseline multilevel models. Results indicated that previous day's mood was positively associated with next-day mood (est = 0.20, $SE = .04, p < .001$). Previous day's pain intensity was also positively associated with pain intensity the following day (est = .20, $SE = .04, p < .001$). Study group emerged as a significant predictor of total sleep time, sleep efficiency, and diary-reported sleep quality. Specifically, youth with SB experienced shorter sleep time (est = 30.0, $SE = 14.3, p = .04$), lower sleep efficiency (est = .91, $SE = .26, p = .001$), and higher WASO (est = 2.33, $SE = .54, p < .001$) across the sleep assessment period. Counterintuitively, youth with SB reported *better* sleep quality (est = -.84, $SE = .38, p = .03$). None of the demographic covariates predicted sleep, daytime mood or pain intensity in these models. Our original data analytic plan was to estimate the slope parameter and interaction term (e.g., Study Group X Sleep Efficiency) in each model. However, preliminary analyses found that none of the interaction terms were significant and were therefore dropped from all models to increase power. Dropping the interaction term also allowed for the elimination of slope parameters, and, thus, estimation of random-intercept-only models. This decision was made because estimating slope parameters decreases power to detect within-person effects (Bolger & Laurenceau, 2013).

Nighttime sleep disturbances as predictors of daytime mood and pain (Hypothesis 2;

Table 10). Separate models evaluated actigraphic sleep efficiency, WASO, total sleep time, and diary-reported sleep quality as predictors of mood (four models) and pain (four models) the next day, above and beyond the effects of demographic covariates. As hypothesized, nighttime sleep efficiency, total sleep time, and diary-reported sleep quality were significantly related to mood the following day ($ps < .05$). Specifically, lower sleep efficiency, shorter sleep time, and poorer diary-reported sleep quality predicted worse mood the next day. Contrary to hypotheses, there was no significant effect of WASO, or number of minutes awake following sleep onset, on daytime mood. In addition, as hypothesized, WASO predicted pain intensity the next day, with greater number of minutes awake after sleep onset associated with higher pain the following day ($p = .04$). Contrary to hypotheses, nighttime sleep efficiency, sleep time, and diary-reported sleep quality did not predict pain intensity the next day.

Table 10. Estimates for Multilevel Models of Nighttime Sleep Disturbances Predicting Daytime Mood and Pain

Fixed Effects	Daytime Mood			Daytime Pain		
	Est	SE	<i>t</i>	Est	SE	<i>t</i>
Sleep Efficiency	0.19	0.08	2.41*	-0.02	0.02	-1.05
Study Group	-0.67	0.53	-1.27	-0.04	0.04	-1.03
WASO	0.03	0.03	1.07	0.02	0.01	2.04*
Study Group	-0.66	0.52	-1.28	-0.04	0.04	-1.01
Total Sleep Time	0.01	<0.01	5.79**	<-0.01	<0.01	-0.23
Study Group	-0.50	0.39	-1.28	-0.04	0.04	-0.98
Sleep Quality	0.49	.05	10.74**	0.01	0.01	0.73
Study Group	-0.50	.41	-1.22	-0.03	0.05	-0.60

Notes. The number of subjects was 68 and the total number of observations was 475-539 in models of nighttime sleep predicting daytime mood or pain. Models controlled for previous day's mood/pain, age, gender, and pubertal status. Sleep variables were person-centered by subtracting each person's mean rating from all of their daily ratings to represent within-person effects. Est = Estimate; WASO = Time Awake after Sleep Onset; Study Group is coded as 0 = SB and 1 = TD.

* $p < .05$; ** $p < .01$

Daytime mood and pain as predictors of nighttime sleep disturbances (Hypothesis 3;

Table 11). Next, separate models evaluated daytime mood and pain as predictors of all the four sleep parameters, above and beyond the effects of demographic covariates. There was no significant main effect of daytime pain on nighttime sleep parameters. However, contrary to hypotheses, higher daytime mood predicted *reduced* total sleep time ($p = .03$) and *lower* sleep quality. Consistent with baseline models, study group emerged as a significant main effect for the majority multilevel models predicting sleep parameters; youth with SB experienced lower sleep efficiency, shorter sleep time, higher WASO, and *better* nighttime sleep quality ($ps < .05$). Group status became non-significant in the model with pain intensity predicting total sleep time ($p > .05$).

Table 11. Estimates for Multilevel Models of Daytime Mood and Pain Predicting Nighttime Sleep Disturbances

Fixed Effects	Sleep Efficiency			WASO			Total Sleep Time			Sleep Quality		
	Est	SE	<i>t</i>	Est	SE	<i>t</i>	Est	SE	<i>t</i>	Est	SE	<i>t</i>
Daytime Mood	-0.02	.03	-0.66	-0.05	.07	-0.47	-4.72	2.39	-1.97*	-0.10	0.04	-2.39*
Study Group	0.85	.26	3.24**	-2.18	.53	-4.08**	29.65	14.27	2.08*	-1.12	0.38	-3.00**
Pain Intensity	0.08	.16	0.51	-0.50	0.38	-1.30	-5.23	11.97	-0.44	-0.31	0.22	1.39
Study Group	0.87	.27	3.24**	-2.29	0.55	-4.17**	27.92	14.55	1.92	-0.93	0.39	-2.40*

Notes. The number of subjects was 68 and the total number of observations was 521-551 in models of daytime mood/pain nighttime sleep. Models controlled age, gender, and pubertal status. Pain and mood variables were person-centered by subtracting each person's mean rating from all of their daily ratings to represent within-person effects. Est = Estimate; WASO = Time Awake after Sleep Onset; Study Group is coded as 0 = SB and 1 = TD.

* $p < .05$; ** $p < .01$.

Overview of Part II (Addressing Hypothesis 4)

Part II of this project examined the relationship between sleep-wake disturbances and bio-neuropsychosocial functioning in adolescents with SB. Multivariate linear regression

analyses examined aggregated objective and subjective sleep variables as predictors of biopsychosocial functioning in the SB group only. Regression models included covariates and sleep variables that were significantly correlated with each measure of adolescent bio-neuropsychosocial functioning as determined by bivariate correlation analyses. Variables included in each regression model are shown in Table 14.

Correlations among covariates, sleep, and bio-neuropsychosocial variables

Covariates and bio-neuropsychosocial variable (Tables 12 & 13). Among the demographic covariates (Table 12), older adolescent age and more advanced pubertal status was associated with fewer symptoms of internalizing as reported by teachers ($r = -.49$ & $r = -.55$, respectively, $ps < .05$). More advanced pubertal status was also associated with fewer executive function problems as reported by teachers ($r = .42$, $p = .02$). Lower income level was associated with lower quality of life ($r = .36$, $p = .03$) and greater child report of internalizing symptoms ($r = -.37$, $p = .03$). Female gender was also associated with lower quality of life ($r = -.48$, $p < .01$).

Regarding SB medical covariates (Table 13), the presence of a shunt and lower IQ were associated with more symptoms of internalizing as reported by teachers ($r = .43$ & $r = -.58$, respectively, $ps < .05$). Lower IQ was also related to greater symptoms of executive dysfunction as reported by parents ($r = -.43$, $p = .02$). Counterintuitively, the presence of a shunt was associated with *less* family conflict ($r = -.35$, $p = .04$) and *greater* family cohesion ($r = .37$, $p = .03$). Higher lesion level was associated with more frequent and intense pain ($r = .40$ & $r = .36$, $ps < .05$) and lower parent report of scholastic competence ($r = -.40$, $p = .03$). Higher number of prescription medication categories/types was also associated with more intense pain ($r = .35$, $p = .04$) and higher grades ($r = .35$, $p = .04$). Greater limitations in gross motor function was

associated with more frequent pain ($r = .34, p = .04$), lower health-related quality of life ($r = -.59, p < .01$), and greater adolescent report of internalizing ($r = .45, p = .01$).

Sleep-Wake disturbances and bio-neuropsychosocial functioning. Several significant associations were found among actigraphic sleep-wake variables and bio-neuropsychosocial functioning. Greater variability in total sleep time (sleep variability) was significantly related to greater child- and parent-report of internalizing symptoms ($r = .42$ and $r = .53, ps < .05$) and lower quality of life ($r = .49, p < .01$). Counterintuitively, longer total sleep time was associated with *greater* teacher-reported symptoms of internalizing ($r = .44, p = .01$) and greater WASO was associated with *lower* adolescent report of internalizing symptoms ($r = -.34, p = .04$). Further, longer total sleep time was also associated with greater family cohesion ($r = .35, p = .04$). Similarly, more minutes awake after sleep onset (WASO) and lower sleep efficiency were associated with greater family conflict ($r = .36$ & $r = -.34, ps < .05$). In terms of academic and neuropsychological functioning, unexpectedly, longer total sleep time was associated with *greater* teacher report of executive dysfunction ($r = .41, p = .02$) and *lower* parent-report of scholastic competence ($r = -.39, p = .02$).

There were also several significant associations found among the subjective, questionnaire-reported sleep-wake variables and biopsychosocial variables. The total score of the Adolescent Sleep Hygiene Scale was the only sleep variable associated with physical functioning; poorer sleep habits was significantly correlated with higher zBMI ($r = -.41, p = .02$).

Table 12. Bivariate Correlations among Demographic and Bio-neuropsychosocial Variables in SB Group

Variables	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19
1. Gender ^a	--																		
2. Age	-.03	--																	
3. Income Level ^b	-.25	-.01	--																
4. Pubertal Status	.28	.52*	.21	--															
5. zBMI	.06	.26	-.14	.23	--														
6. Pain Frequency (C)	.18	-.21	-.14	.10	-.12	--													
7. Pain Intensity (C)	.11	-.10	-.18	-.11	-.10	.62*	--												
8. PedsQL (M, F, C)	.48*	-.07	.36*	-.05	-.16	-.41*	-.36*	--											
9. Internalizing (M, F)	.15	-.17	-.08	-.09	-.17	.22	.18	-.45*	--										
10. Internalizing (T)	.01	-.49*	-.27	-.55*	-.10	-.02	.14	.00	.05	--									
11. Internalizing (C)	.21	.06	-.37*	-.07	.17	.30	.22	-.76*	.29	.23	--								
12. Family Conflict (M, F)	-.16	-.01	.03	-.19	-.01	-.05	-.08	.12	.08	-.01	-.03	--							
13. Family Cohesion (M, F)	.17	.08	-.20	.14	-.04	.05	-.11	.10	-.28	.20	-.09	-.37*	--						
14. SNAP (M, F, T)	-.07	.06	.07	-.14	.16	-.03	.05	-.33	.23	.21	.18	.24	-.23	--					
15. BRIEF (M, F)	-.17	.20	.09	.07	.06	-.05	.02	-.38*	.55*	-.10	.20	.07	-.22	.71*	--				
16. BRIEF (T)	.01	-.28	-.28	-.42*	.11	.09	.27	-.38*	.06	.64*	.32	.09	-.14	.69*	.25	--			
17. Harter-SC (M, F)	.02	.06	-.01	.02	-.08	-.09	-.18	.44*	-.44*	-.31*	.39*	-.11	-.01	-.65*	-.68*	-.46*	--		
18. Harter-SC (T)	-.07	.42*	.13	.24	-.02	-.04	-.28	.15	-.09	-.59*	-.06	-.11	.22	-.27	-.01	-.62*	.14	--	
19. Grades (M, F, T)	.08	.16	.11	-.02	-.35*	.03	-.04	.27	-.09	-.34	-.18	-.06	.04	-.39*	-.23	-.42*	.36*	.54*	--

Notes. M = Mother-report; F = Father-report; C = Child-report; P = Combined parent-report; T = Teacher-report; zBMI = Body Mass Index z-score; PedsQL= Pediatric Quality of Life Inventory- Psychosocial Total Score; SNAP = Swanson, Nolan, and Pelham Teacher and Parent Rating Scale (Swanson et al., 2001) to measure inattention/hyperactivity; BRIEF = Behavioral Rating Inventory of Executive Function (Gioia, Isquith, Gray, & Kenworthy, 2000); Harter-SC = Harter Scholastic Competence scale.

^aGender coded as 0 = male, 1 = female

^bSpearman's rho

* $p < .05$

Table 13. Bivariate Correlations among Medical and Bio-neuropsychosocial Variables in SB Group

Variables	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	22	23	
1. Shunt Status ^a	-																							
2. Lesion Level ^b	-.13	-																						
3. Shunt Revisions ^c	.52*	-.03	-																					
4. Surgeries ^c	.24	-.30	.27	-																				
5. Hospitalizations ^c	.06	.04	.17	.58*	-																			
6. GMF	.08	.47*	.04	.06	.30	-																		
7. Rx Medication ^d	-.03	-.01	.23	.24	.34*	.10	-																	
8. WASI	-.03	-.40*	.07	.22	.04	-.04	.04	-																
9. zBMI	.08	-.17	.05	-.20	-.01	.07	.08	-.06	-															
10. Pain frequency (C)	-.04	.39*	-.11	-.12	.14	.34*	.04	-.01	-.12	-														
11. Pain intensity (C)	-.12	.36*	-.08	-.15	.13	.33	.35*	-.30	-.10	.62*	-													
12. PedsQL (M, F, C)	-.04	-.26	.08	.07	-.13	-.59*	-.25	.09	-.16	-.41*	-.36*	-												
13. Internalizing (M, F)	-.04	.32	-.06	.06	.06	.31	.03	-.01	-.17	.22	.18	-.45*	-											
14. Internalizing (T)	.43*	.27	.06	-.02	-.07	.10	-.25	-.58*	-.10	-.02	.14	.00	.05	-										
15. Internalizing (C)	.02	.32	-.19	-.09	.11	.45*	.16	-.16	.17	.30	.22	-.76*	.29	.23	-									
16. Family conflict (M, F)	-.35*	.10	-.13	-.07	-.10	-.08	-.14	.01	-.01	-.05	-.08	.12	.08	-.01	-.03	-								
17. Family cohesion (M, F)	.37*	.16	.15	-.33	-.32	-.05	-.20	-.02	-.04	.05	-.11	.10	-.28	.20	-.09	-.37*	-							
18. SNAP (M, F, T)	.22	-.04	.28	.22	.03	.10	.16	-.09	.16	-.03	.05	-.33	.23	.21	.18	.24	-.23	-						
19. BRIEF (M, F)	.31	-.03	.02	.09	.08	.25	.11	-.43*	.11	-.05	.02	-.38*	.55*	-.10	.20	.07	-.22	.71*	-					
20. BRIEF (T)	.11	.08	.20	.12	-.13	.08	.09	.09	.06	.09	.27	-.38*	.06	.64*	.32	.09	-.14	.69*	.25	-				
21. Harter-SC (M, F)	-.15	-.40*	-.24	-.09	-.11	-.32	-.19	.21	-.08	-.09	-.18	.44*	-.44*	-.31*	-.39*	-.11	-.01	-.65*	-.68*	-.46*	-			
22. Harter-SC (T)	-.13	.10	.07	.12	.02	-.09	.32	.36	-.02	-.04	-.28	.15	-.09	-.59*	-.06	-.11	.22	-.27	-.01	-.62*	.14	-		
23. Grades (M, F, T)	-.12	.07	.06	.01	-.13	-.27	.35*	.11	-.35*	.03	-.04	.27	-.09	-.34	-.18	-.06	.04	-.39*	-.23	-.42*	.36*	.54*	-	

Notes. GMF = Gross motor functioning (higher scores indicate higher level of impairment); WASI = Wechsler Abbreviated Scale of Intelligence; zBMI = Body Mass Index z-score; PedsQL= Pediatric Quality of Life Inventory- Psychosocial Total Score; SNAP = Swanson, Nolan, and Pelham Teacher and Parent Rating Scale to measure inattention/hyperactivity; BRIEF = Behavioral Rating Inventory of Executive Function; Harter-SC = Harter Scholastic Competence scale.

^aShunt status coded as 1 = present; 0 = absent/no shunt

^bLesion level coded such that sacral lesions levels had lowest scores and thoracic had highest scores

^cTotal number; Shunt revisions are total in lifetime, surgeries and hospitalizations are total in previous two years

^dRx medications represent the total number of types of prescription medications taken (e.g., antidepressants, anticonvulsants, etc.)

* $p < .05$; ** $p < .01$

In terms of psychosocial functioning, worse sleep quality, poorer sleep habits, higher levels of pre-sleep arousal, and greater daytime fatigue were associated with more symptoms of internalizing as reported by adolescents ($r_s = -.59 - .54, p_s < .05$) and lower psychosocial health-related quality of life ($r_s = -.54 - .76, p_s < .05$). Sleepiness was associated with greater parent report of youths' internalizing ($r = .44, p = .01$) and lower psychosocial quality of life ($r = -.36, p = .03$).

With regard to neuropsychological and academic functioning, fatigue and sleepiness were associated with greater executive function problems as reported by teachers and parents, respectively ($r = -.44$ & $r = .39; p_s < .05$). The sleepiness subscale was also associated with greater symptoms inattention/hyperactivity ($r = .36, p = .04$) and lower parent-report scholastic competence ($r = .39, p = .02$). Finally, greater fatigue and sleepiness was associated with lower school grades ($r = .39$ & $r = -.41, p_s < .05$).

None of the objective or subjective sleep variables were significantly associated with pain frequency or intensity. The PSQ snoring subscale was not significantly related to any of the bio-neuropsychosocial outcomes ($p_s > .05$).

Multivariate regression analyses: sleep-wake disturbances as predictors of bio-neuropsychosocial functioning in adolescents with SB (Hypothesis 4). Results of multivariate regression analyses predicting physical functioning after controlling for relevant covariates are presented in Table 14.

Physical functioning. Worse sleep habits were identified as the only significant predictor of increased body mass index (BMI) in adolescents with SB ($\beta = -.38, p = .02$). Subjective and objective sleep disturbances were not associated with pain frequency or intensity in adolescents

with SB. While higher lesion level did not significantly predict higher pain intensity using the standard alpha level of less than .05, the p-value was exactly .05 ($\beta = .36$) and therefore closely approached significance.

Because recent research has increasingly highlighted the impact of sleep timing (i.e., later bedtime/wake times) as a salient predictor of pediatric obesity (Horne, 2008), we sought to further explore the specific domains of the Adolescent Sleep Habits Questionnaire (ASHS) that may predict greater zBMI in our sample of adolescents with SB. Specifically, the four subscales of the ASHS (Cognitive, Emotional, Sleep Environment, and Sleep Stability) were entered into the regression model predicting zBMI in forward fashion. Results indicated that lower scores on the Sleep Stability subscale (including 4 items related to going to bed and waking up late on the weekends and weekdays) predicted zBMI ($r = -.25, p = .04$). Thus, greater sleep timing instability (going to bed late/sleeping in during the week) was related to higher zBMI in adolescents.

Psychological functioning. As hypothesized, poorer self-report of sleep quality and greater actigraphic sleep variability predicted lower psychosocial quality of life in our sample of adolescents with SB ($\beta = .51$ & $\beta = -.28$, respectively, $ps < .05$). Greater sleep variability and sleepiness predicted greater internalizing symptoms as measured by parent report ($\beta = .53$ & $\beta = .31, ps < .05$). In addition, sleep variability and greater pre-sleep arousal emerged as a significant predictors of adolescent report of internalizing symptoms ($\beta = .33$ & $\beta = .48, ps < .05$).

Table 14. Objective and Subjective Sleep Predicting Bio-neuropsychosocial Functioning in Adolescents with SB

Dependent Variables	Independent Variables	<i>b</i>	SE <i>b</i>	β
<u>Physical Functioning</u>				
Body Mass Index (BMI)	Sleep Habits	-0.72	0.31	-0.38*
Pain frequency (C)	<i>None Significant</i>	--	--	--
Pain intensity (C)	Lesion Level ^a	1.85	0.91	0.36+
<u>Psychological Functioning</u>				
PedsQL Psychosocial Total (M, F, C)	Gender ^b	-10.05	3.44	-0.38**
	Gross Motor Function	-6.48	1.63	-0.51**
	Sleep Variability (A)	-0.45	0.20	-0.28*
	Sleep Quality (Q)	9.34	2.14	0.50**
Internalizing (M, F)	Sleep Variability (A)	0.57	0.16	0.53**
	Sleepiness (Q)	9.46	4.41	0.31*
Internalizing (T)	Age	-2.79	0.97	-0.34**
	Pubertal Status	-5.52	2.63	-0.25*
	Shunt Status ^c	9.55	2.91	0.34**
Internalizing (C)	IQ ^d	-0.30	0.06	-0.55**
	Gross Motor Function	0.10	0.04	0.45**
	Sleep Variability (A)	0.01	0.01	0.33*
	Pre-sleep Arousal (Q)	0.74	0.21	0.48**
<u>Family Functioning</u>				
Family conflict (M, F)	Shunt Status ^c	-0.34	0.16	-0.35*
	WASO (A)	0.05	0.02	0.34*
Family cohesion (M, F)	Shunt Status ^c	0.29	0.13	0.37*
<u>Neuropsychological Functioning</u>				
Inattention/Hyperactivity ^e (M,F,T)	Sleepiness (Q)	0.43	0.21	0.34*
Executive Dysfunction ^f (M, F)	Sleepiness (Q)	0.43	0.17	0.41*
Executive Dysfunction ^f (T)	Pubertal Status	-0.23	0.11	-0.35*
	IQ ^d	-0.01	0.01	-0.37*
	Fatigue (Q) ^h	-0.01	0.01	-0.41*
<u>Academic Functioning</u>				
Scholastic Competence ^g (M,F)	Lesion Level ^a	-0.42	0.18	-0.40*
Scholastic Competence ^g (T)	Age	0.23	0.10	0.42*
Grades (M, F, T)	# Rx Medication Types	0.54	0.26	0.35*
	Fatigue (Q)	0.04	0.01	0.47**

Notes. Regression models were trimmed such that non-significant ($p > .05$) were effects removed). M = Mother-report; F = Father-report; C = Child-report; P = Combined parent-report; T = Teacher-report; A = Actigraphy/Objective Sleep Variable; Q = Questionnaire/Subjective Sleep Variable; BMI = Body Mass Index z-score; WASO = minutes awake after sleep onset; Sleep Variability = average variation in Total Sleep Time.

^aCoded such that sacral lesions levels had lowest scores and thoracic had highest scores;

^bCoded as 0 = male, 1 = female;

^cCoded as 1 = shunt present; 0 = absent/no shunt;

^dMeasured with the Wechsler Abbreviated Scale of Intelligence (WASI);

^eMeasured with Swanson, Nolan, and Pelham Teacher and Parent Rating Scale (SNAP);

^fMeasured with the Behavioral Rating Inventory of Executive Function (BRIEF).

^gMeasured with the Harter- Scholastic Competence Scale.

^hMeasured using the PedsQL Multidimensional Fatigue Scale, lower scores indicate greater problems with fatigue.

* $p < .05$, ** $p < .01$

Sleep-wake disturbances did not predict teacher report of adolescent internalizing after adjusting for covariates. Younger age, less advanced pubertal status, lower IQ, and presence of a shunt were significantly related to greater teacher-report of internalizing symptoms (β s = $-.55$ - $.34$, $ps < .05$). In addition, greater (i.e., more limited) gross motor function and female gender predicted lower quality of life ($\beta = -.51$ & $\beta = -.38$, $ps < .05$). More limited gross motor function also predicted more internalizing symptoms as reported by adolescents ($\beta = .48$ $p = .01$).

Family functioning. WASO emerged as the only significant sleep predictor of family functioning. As hypothesized, more minutes awake after sleep onset (WASO) predicted greater family conflict ($\beta = .34$, $p = .04$). Counterintuitively, the presence of a shunt predicted lower family conflict ($\beta = -.35$, $p = .04$) and greater family cohesion ($\beta = .37$, $p = .03$) in our sample.

Neuropsychological functioning. Parent report of adolescent sleepiness emerged as a significant predictor of inattention/hyperactivity ($\beta = .34$, $p = .04$) and parent-report of executive dysfunction ($\beta = -.41$, $p = .04$). Similarly, greater levels of fatigue were associated with greater executive dysfunction as reported by teachers ($\beta = .40$, $p = .01$). Less advanced pubertal status and lower IQ were also associated with greater problems with teacher-reported executive dysfunction ($\beta = -.37$ & $\beta = -.37$, $ps < .05$).

Academic functioning. Fatigue emerged as the only significant predictor of academic functioning; greater fatigue was associated with lower school grades in adolescents with SB ($\beta = .47$, $p = .004$). Sleep-wake disturbances did not predict scholastic competence. In terms of covariates, older age and lower lesion level were associated with better teacher- and parent-

report of scholastic competence, respectively ($\beta = .42$ & $\beta = -.40$, $ps < .05$). More types of prescription medication were associated with higher school grades ($\beta = .35$, $p = .04$).

CHAPTER FOUR

DISCUSSION

Sleep Disturbances in Adolescents with Spina Bifida

This study is the first to provide a case-controlled, multi-modal assessment of sleep-wake disturbances in pediatric SB using objective and subjective sleep instruments. Overall, study findings suggest that adolescents with SB experience higher rates of sleep-wake disturbances across actigraphy and questionnaire measures compared to typically developing youth.

Adolescents with SB were found to experience sleep deficits, more time awake after sleep onset, lower sleep efficiency, and greater sleep variability (inter-individual variation in sleep time) according to actigraphy and reported worse sleep quality, sleep habits, and higher levels of fatigue on questionnaire measures.

Our findings that adolescents with SB reported worse objectively and subjectively-measured sleep is consistent with prior research on sleep patterns among youth with chronic illnesses, developmental and physical disabilities, and neurological conditions (Hysing et al., 2009; Tham, Fales, & Palermo, 2015; Tieze et al., 2012). Notably, our data confirmed that adolescents with SB received slightly less total sleep compared to their peers. In particular, adolescents with SB slept a little over 6 hours, with typically developing youth receiving slightly more sleep (about 6 ½ hours) on average. Given National Sleep Foundation recommendations that adolescents should obtain between 8-10 hours of sleep for optimal daytime functioning (National Sleep Foundation, 2015), these results confirm the presence of significant sleep deficits

in adolescents with SB. Sleep deficits in this population are likely due to disrupted sleep continuity, as indicated by actigraphic results of greater time awake after sleep onset (WASO) and poorer sleep efficiency.

Subjective reports of sleep-wake disturbances substantiated actigraphy data on poor sleep continuity. In particular, this is the first study to highlight the presence of insomnia symptoms in pediatric SB. About one quarter (25.7%) of adolescents with SB reported clinically significant difficulties staying asleep (i.e., 60% or more of the time) compared to 8.1% of TD youth. Consistent with the typical clinical presentation of pediatric insomnia, the largest effect sizes for group comparisons on the sleep quality subscales were found for difficulties going to bed and reinitiating sleep (Meltzer & Crabtree, 2015). Difficulties with bedtime settling and staying sleep often co-occur, as adolescents that cannot easily settle to sleep may also be less likely to re-settle themselves after awakening during the night.

Results of this study also highlight the presence of more problematic bedtime sleep habits in adolescents with SB compared to the TD sample. In addition, adolescents with SB experienced more sleep variability (often referred to as night-to-night stability in sleep time/quantity) compared to their peers. As we found in our preliminary analyses, poorer bedtime habits are often associated with greater sleep variability (Becker et al., 2016). For adolescents with SB, less optimal sleep environments (e.g., noise, TV watching) and more disruptive/chaotic schedules (e.g., time spent in hospital settings/away from home) may impact the stability of night-to-night sleep time. It will be critical for clinicians working with adolescents with SB and their caregivers should not only inquire about overall or average sleep quantity, but also the stability of sleep quantity, as well as barriers to establishing consistent sleep schedules.

Efficacious preventative and treatment efforts may focus on promoting better sleep habits (Melzer & Crabtree, 2015) including the improvement of physical sleep environments (reduced electronics use, and cool, dark sleep environments) and establishing consistent bedtimes and wake times to stabilize sleep patterns in adolescents with SB.

Finally, levels of fatigue were alarmingly high in adolescents with SB compared to their TD peers. In fact, severity levels were similar to those reported by children and adolescents undergoing active treatment and chemotherapy for cancer (Erickson et al., 2011; Varni et al., 2002). Clinicians and parents commonly attribute fatigue to underlying disease-related mechanisms such as pain and weakness; indeed, significant associations between gross motor limitations and fatigue may implicate muscle weakness in the development of fatigue in pediatric SB. Intuitively, sleep disturbances may contribute to the development of fatigue in adolescents with and without chronic illnesses (Tham, Holley, Zhou, Clarke, & Palermo, 2013). Indeed, our preliminary analyses found that questionnaire-reported sleep quality and pre-sleep arousal were strongly correlated with fatigue in adolescents with SB (i.e., $r_s \geq .70$), with less robust associations found for TD adolescents (i.e., moderate $r_s = .50$ -.69). Further research is needed to understand the role of nighttime sleep disturbances and other psychosocial factors (e.g., depression) on the development and persistence of fatigue in youth with SB. Fatigue represents a significant clinical problem in adolescents with SB, and such work may help to identify salient treatment targets to prevent or ameliorate these symptoms.

While the majority of group comparison results showed that adolescents with SB are at-risk for sleep disturbances, descriptive analyses of diary-reported sleep quality found that the SB sample endorsed *better* sleep quality compared to TD adolescents. Although counterintuitive,

these findings may be partially attributed to the chronicity of sleep problems and co-morbidities in this condition; more positive ratings of sleep quality may reflect an adaption process to undetected, unremitting sleep difficulties rather than lack of sleep disruptions. In other words, it is possible that some adolescents with SB have not yet experienced a reprieve from sleep disturbances and, thus, do not perceive “good” sleep quality in the same way as their TD peers. It is also possible that adolescents with SB are less likely to notice daily fluctuations in sleep quality because noticeable behavioral and neuropsychological symptoms of poor sleep quality, such as inattention, may be misattributed or masked by underlying neuropsychological vulnerabilities present since birth. Clearly, a one-item rating of global sleep quality is not sufficient to identify sleep disturbances in youth with SB. In clinical and research settings, a comprehensive assessment of sleep-wake patterns may be critical to improve identification and treatment of sleep disturbances in adolescents with SB.

Associations between Sleep Disturbances and Bio-neuropsychosocial Functioning

Sleep is a critical developmental need for adolescents, and when disrupted, has been linked to a multitude of issues with physical status, psychological health, family functioning, neuropsychological symptoms, and academic performance (Alfano, Zakem, Costa, Taylor & Weems, 2009; Chen, Beydoun & Wang, 2008; Kheriandish & Gozal, 2006). Because adolescents with SB are already at-risk for difficulties in these areas (Appleton, 1997; Clancy, McGrath, & Oddson, Holmbeck et al., 2003; Holmbeck et al., 2010; Rose & Holmbeck, 2007), good sleep quantity and quality may be especially critical. Yet there are limited data to date on how sleep disturbances relate to key areas of health and functioning in this population.

As a first step toward understanding potential consequences of sleep disturbances in adolescents with SB, our study examined bio-neuropsychosocial correlates of sleep disturbances in this at-risk population. As part of a 10-day sleep assessment procedure, we collected daily actigraphy and diary information to examine temporal relationships between sleep, mood, and pain in the SB and comparison groups. We also identified associations between sleep and a broader array of adolescent physical, psychosocial, family, neuropsychological, and academic functioning in the SB group only. Overall, study findings indicated that sleep-wake disturbances were associated with every domain of adolescent functioning within the bio-neuropsychosocial model. Most consistently, our results uncovered associations between objective and subjective nighttime sleep disturbance and psychological maladjustment (internalizing, HRQOL). To a lesser extent, nighttime sleep disturbances were linked to worse physical health (pain, BMI) and family functioning (family conflict). Furthermore, daytime sleepiness and/or fatigue, but not nighttime sleep disturbances, was associated with worse neuropsychological and academic functioning, including inattention/hyperactivity, executive dysfunction, and lower school grades. For children and adolescents with SB, good sleep quality and quantity have far-reaching effects and may be critical to optimizing multiple aspects of health and well-being.

Sleep and Physical Functioning

Research on sleep and physical health has tended to focus on two important physical health parameters: pain and weight status/obesity. Similar to investigations of daily sleep-pain associations in children with JIA (Bromberg, Gil, & Schanberg, 2012) and adolescents with idiopathic chronic pain (Lewandowski, Palermo, De la Motte et al., 2010), pain intensity did not impact sleep disturbances, but more minutes awake after sleep onset (WASO) predicted higher

pain intensity the following day in adolescents with and without SB. It is possible that sleep disturbances may be a stronger predictor of pain than pain is of sleep disturbances. Experimental sleep modification research showing that sleep deprivation causes increased pain sensitivity and hyperalgesic pain response supports this notion (Lautenbacher, Kundermann & Krieg, 2006). Because overall levels of pain reported by adolescents with and without SB were low, it is also possible that pain was not severe enough to disrupt nighttime sleep. However, comparing our data with previous reports on young adults with SB suggests that pain may become more severe and frequent as individuals with this condition approach adulthood (Verhoef et al., 2004), potentially due to increases in secondary medical complications (e.g., shunt malfunctions) and surgical procedures. Interventions targeted toward sleep in this population may be effective at reducing sleep disturbances while also reducing pain intensity and, quite possibly, lowering the risk for developing chronic pain later in adulthood.

In addition to chronic pain, individuals with SB are also at-risk for obesity and overweight. Our study found that poorer sleep habits were associated with higher zBMI in adolescents with SB. This finding was somewhat surprising given the number of studies that have supported associations between total sleep time and pediatric obesity (Cappuccio et al., 2008; Marshall et al., 2008). On the other hand, recent research has revealed that *late sleep timing* (i.e., later bedtimes or wake times) may be a better predictor of pediatric obesity than total sleep time (Horne, 2008) per say. Indeed, our exploratory regression analyses found that later sleep timing (going to bed late, sleeping in) was driving this relationship. Later sleep timing behavior may provide more opportunities for adolescents with SB to engage in sedentary behaviors (e.g., TV watching) and nighttime snacking/overeating (Golly, Maher, Mattriciani, &

Olds, 2013). Health professionals working with adolescents with SB on issues pertaining to sleep and eating behavior should consider assessing typical bedtimes and wake times. Interventions and experimental home-based sleep extension protocols have successfully modified bedtimes (and extended sleep) in adolescents with asthma and T1DM (Meltzer, Faino, Szeffler, Strand, Geldand, & Beebe, 2015; Perfect, Beebe, Levine-Donnerstein, Frye, Bluez, & Quan, 2016), with observed positive effects on physical health outcomes. Similar research may be conducted in adolescents with SB to provide useful data on sleep timing as a treatment target for improved physical health

Sleep and Psychological Functioning

The current study also suggests that nighttime sleep patterns and behaviors are strongly related to adolescent psychological dysfunction. In support of a small but growing number of studies on the adverse psychosocial consequences of sleep variability (Bei Bei, Wiley, Trinder, Manber, 2016; Becker, Sidol, Van Dyk, Epstein, & Beebe, 2016), greater sleep variability was associated with poorer psychological well-being in adolescents with SB (internalizing, psychosocial HRQOL). While pediatric sleep is often measured across several days, previous studies have not systematically investigated night-to-night variability in sleep (Bei Bei, Wiley, Trinder, Manber, 2016). There is also a lack of existing framework for understanding the mechanisms through which sleep variability is associated with psychological functioning. Potentially, sleep variability may play a role in mood instability and difficulties coping with fluctuations in negative arousal. To better understand the nature and impact of sleep variability, there is an overarching need for clinical and research efforts to address both typical *and* night-to-night variation in sleep time. Working with parents to establish consistent bedtimes may improve

sleep and co-morbid internalizing symptoms in adolescents with SB. From a preventative standpoint, bedtime habits may be established early in childhood and monitored throughout development.

Further, our study results found that heightened pre-sleep arousal and daytime sleepiness was associated with internalizing symptoms in adolescents with SB. Cognitive and physiological arousal at bedtime may perpetuate or maintain negative sleep patterns (Palermo, Law, Churchill, & Walker, 2012), placing youth at-risk for developing depression. Dysfunctional cognitions about sleep and the daytime consequences of poor sleep are common in those experiencing difficulties initiating sleep (i.e., cognitive pre-sleep arousal), and this type of ruminative thinking style may be reinforced over time, further perpetuate sleep disturbance, and lead to the development or intensification of depression symptoms (Lovato & Gradisar, 2015). Daytime sleepiness may further intensify depressive symptoms by reducing adolescents' motivation to partake in social activities (Lovato & Gradisar, 2015). It will be important for longitudinal research to expand data on the temporal sequence of sleep and internalizing symptoms in vulnerable child and adolescent populations such as SB. Such work may test complex longitudinal pathways and include modifiable behavioral mechanisms (i.e., pre-sleep arousal) through which sleep confers risk for psychosocial maladjustment.

In support of these cross-sectional data, daily analyses found that poorer sleep efficiency, reduced total sleep time, and lower diary-reported sleep quality predicted worse next-day mood in our adolescent samples. These findings are unsurprising given growing evidence of the role of sleep in developing affective disorders such as depression in adolescents (Ivanko, 2015). Sleep has been implicated in the regulation of emotional brain networks, including facilitation of

inhibitory effects of the medial-prefrontal amygdala (Walker & van der Helm, 2009). Sleep difficulties may therefore disrupt emotional regulation processes and, if chronic, may lead to more frequent and intense symptoms of depression and anxiety (Ivanko, 2015).

Results of multilevel models on sleep predicting next-day pain and mood suggest that sleep disturbances may represent a common risk factor for internalizing and pain symptoms in pediatric SB. Thus, interventions that target sleep may lead to improvements in both pain and mood. It will also be important for future work to discern the temporal sequence of sleep, pain, and internalizing symptoms- deemed the “Unhappy Triad” (Koffel, Krebs, Arbisi, Erbes, & Polusny, 2016). Research on adult populations suggests that the temporal link between sleep and pain may be due, in part, to negative mood (Koffel et al., 2016). In other words, sleep disruptions may result in emotional dysregulation and impaired mood, which can impact pain perception and pain-related distress through increased physiologic or cognitive arousal. On the other hand, pain may also mediate the sleep-internalizing relationship, such that disrupted sleep reduces pain thresholds, directly resulting in increased pain severity, and ultimately increased negative cognitions and emotions (Koffel et al., 2016). Testing competing longitudinal models encompassing this symptom triad should be applied to pediatric populations including SB to broaden our understanding of the etiology and temporal unfolding of sleep, pain, and mood. s may increase susceptibility to psychological disorders such as anxiety and depression.

Finally, and counterintuitively, positive daytime mood predicted *reduced* total sleep time and *lower* diary-reported sleep quality. Thus, the daily dynamics of sleep and mood may be more complexly related than previously theorized. It is possible that typical adolescent activities associated with positive mood, such as socializing with friends, delay sleep onset and thereby

reduce sleep quantity and quality (Fuligni & Hardway, 2006). Additionally, while our study did not measure different types of positive and negative affect, certain emotions may be characterized by higher levels of cognitive and physiological arousal (e.g., excitement vs. calm) that are non-conducive to sleep. Furthermore, our study was limited to measuring mood once daily (in the morning). Future studies may utilize techniques such as electronic momentary assessment to capture fluctuations in mood throughout the day and prior to bed, along with co-occurring activities (e.g., studying, social/extra-curricular activities), to better understand the complex relations between mood, activities, and sleep in adolescents with and without chronic medical illnesses.

Sleep and Family Functioning

Consistent with previous research (Kelly & El-Sheikh, 2011), adolescent sleep disturbances were linked to family dysfunction. Specifically, more wake time after sleep onset (WASO) was associated with greater parent-reported family conflict. There are several possible explanations for why adolescent sleep disturbances may be associated with family conflict. Sleep disturbances may contribute to adolescent psychological maladjustment, resulting in negative and stressful parent-child and marital interactions (Kelly & El-Sheikh, 2011). Particularly for youth with chronic health conditions, sleep disturbances could also compromise parents' sleep (Meltzer & Mindell, 2007), which can impact parental psychological functioning (Bayer, Hiscock, Hampton, & Wake, 2007) and increase the likelihood of conflict (Kelly & El-Sheikh, 2011). Many parents of youth with SB continue to be involved in several aspects of medical management into late adolescence (Psyhogios, Kolbuck, & Holmbeck, 2015). Thus, adolescent and parent dyads may experience poor sleep quality as a result of parent-assisted nighttime

medical management behaviors that disrupt sleep, such as catheterization management.

Examining adolescent and parental psychological maladjustment as mediators of the sleep-family conflict relation will be an important next step towards understanding these processes.

Importantly, emerging evidence has broadened our understanding of reciprocal associations between sleep and family functioning. Just as adolescent sleep may impact family conflict, conflict may disrupt sleep (Kelly & El-Sheikh, 2011). Family conflict is broadly linked to higher perceived vulnerability and environmental stress (Jarrin, McGrath, & Quon, 2014), which may heighten cognitive and physiological arousal and disrupt sleep continuity. These processes may also exist in a cyclical pattern; exposure to conflict may lead to sleep disruptions which may, in turn, lead to increased family conflict. Previous research and current study findings highlight the importance of multimodal, longitudinal sleep research that examines reciprocal relations between sleep and family/parent functioning in the context of pediatric SB.

Sleep and Neuropsychological and Academic Functioning

While the origins of poor neuropsychological skills in pediatric SB are not fully understood, our study findings indicate that executive dysfunction and symptoms of inattention/hyperactivity may be partially attributed to daytime sleepiness. Higher levels of fatigue were also associated with executive dysfunction and lower school grades. These findings support a growing number of studies that have found that daytime sleepiness and fatigue, but not sleep quantity, may lead to decrements in neuropsychological and academic functioning (Anderson et al, 2009; Dewald et al., 2010). Overall, sleepiness and fatigue may have a more proximal influence on neuropsychological and school functioning than sleep disturbances per se. While insufficient or poor sleep quality may cause daytime sleepiness, reductions in

cognitive alertness may directly impair executive function, attention, and learning capacity (Dewald et al., 2010).

Moreover, inattention, executive dysfunction, and poor academic performance has been well documented in pediatric sleep disorders characterized by sleep fragmentation (i.e., increased night awakenings) and daytime sleepiness, including sleep disordered breathing (SDB) and delayed sleep phase syndrome (Beebe et al., 2004; Giannotti, Cortesi, Sebastiani, & Ottaviano, 2002; Blunden, Lushington, Kennedy, Martin, & Dawson, 2000; Cortese et al., 2005). Youth with SB are at-risk for physiological sleep disorders such as central and obstructive sleep disordered breathing (SDB; Waters et al., 1998; Kirk, Morielli & Brouillette 1999). Indeed, symptoms suggestive of SDB were found in one fifth (21.6%) of our SB sample, which is several times the prevalence found in large community samples of adolescents (e.g., 6.0%; Johnson & Roth, 2006). From a developmental psychopathology perspective, sleep disturbances and daytime sleepiness are also common in youth with ADHD (Chervin, Dillon, Bassetti, Ganoczy, & Pituch, 1997; Cortese, Konofal, Yateman, Mouren, & Lecendreaux, 2006; Gau & Chiang, 2009), and children and adolescents with SB have higher rates of ADHD-inattentive type compared to their peers (Burmeister et al., 2005). Thus, sleepiness or fatigue and other associated cognitive and academic impairments may be the result of underlying sleep or psychiatric diagnoses (ADHD, MDD) that were not assessed in the current study.

Limitations and Future Directions

The current study has a number of strengths, such as utilizing a multimodal sleep assessment (questionnaires, actigraphy, diary), multiple reporters of adolescent functioning (youth-, mother-, father-, and teacher- report), and a comparison group. However, there are

several limitations of this study that can be addressed in future work. As is common in pediatric samples, our sample size was small. This limited our statistical power, particularly for single-group analyses involving SB adolescents only. The negative sequelae of sleep may be compounded in adolescents with SB given their increased vulnerability to poor bio-neuropsychosocial outcomes secondary to their illness. Yet, because this study utilized a comparison group from a separate NIH-funded adolescent sleep study that had already completed study procedures, we could not administer bio-neuropsychosocial measures to the control group and test for the potential moderating effect of group (i.e., SB vs. TD groups) on sleep-functioning associations. On the other hand, our use of hierarchical linear modeling of daily sleep, pain, and mood allowed for increased power to detect within-individual variations in sleep-pain and sleep-mood associations. Previous work has also found greater sleep disturbances in minority compared with Caucasian children and families of lower socioeconomic status (Roberts, Roberts, & Chen, 2000). However, our sample was composed of predominately Caucasian (70%) and middle-income families, which limits the generalizability of study findings. At the same time, there was an extra effort to recruit Hispanic, Spanish-speaking families due to the higher incidence of SB in this ethnic group. Recruitment efforts resulted in a higher percentage of Hispanic families (21.6%), which better reflect the ethnic composition of pediatric SB. Further, researchers have advocated for specifying multiple indices of socioecological risk factors of sleep in children, such as community-level poverty (El-Sheikh et al., 2013). Yet, our study was limited to assessment of family income level and could not determine socioeconomic risk factors for sleep disturbances.

Various physiological and psychosocial mechanisms may help to explain the

development of nighttime sleep disruptions in adolescents with SB. However, pediatric SB is characterized by significant medical heterogeneity, which may explain why our study found limited support for condition-related risk factors. Sleep disruptions may be associated with physiological alterations of this condition, including respiratory impairments that increase the likelihood of SDB. Additionally, greater limitations in gross motor functioning were associated with higher levels of daytime fatigue, which may implicate muscle weakness in the development or persistence of fatigue in this population. Future research should examine other salient condition characteristics that were not assessed in the current study, including discomfort related to spasticity and constipation, nighttime enuresis, neurological characteristics (e.g., Chiari malformation), and the presence of comorbid sleep disorders (i.e., periodic limb movement) and medical conditions (e.g., epilepsy, asthma).

Moreover, it is likely that sleep and medical illness exist in a bidirectional relationship; while various aspects of a chronic illness may affect sleep, sleep disturbances may also exacerbate a chronic illness and disease-related symptomatology (Lewandowski et al., 2011b). In particular, it will be beneficial to identify specific medical management behaviors that may disrupt sleep in this population, such as nighttime catheterization and bed turning. Likewise, effective management of SB requires daily use of neuropsychological and emotional processes (e.g., decision-making, motivation, planning, memory) that may be significantly impaired by poor sleep quality and daytime sleepiness (Dewald, Meijer, Oort, Kerkhof, & Bögels, 2010; Perfect et al., 2014; Steenari et al., 2003). Future research may investigate whether sleep-wake disturbances exacerbate cognitive and emotional dysfunction with consequential impediments in youths' ability to perform the often complex array of medical management behaviors.

Our study also had limitations in measurement and design. While actigraphy and self-reports are important tools for assessing sleep-wake patterns in adolescents, the gold standard for quantifying sleep is polysomnography (PSG). Actigraphy is often used in studies due to its comparatively low cost and ecological validity in the home environment compared to PSG. Still, future work may use PSG to expand our understanding of sleep in adolescents with SB, including the incidence of undetected physiological sleep disorders. Finally, we used parent- and teacher- questionnaire report of inattention/hyperactivity and executive dysfunction. Questionnaire measures of neuropsychological functioning have the advantage of assessing “real-world” functioning, and attention and executive functioning are often difficult to assess in a testing environment (Beebe, 2011). However, performance-based measures of neuropsychological functioning may reduce common method variance and parse out specific cognitive deficits related to sleep (Beebe, 2011).

Finally, the cross-sectional nature of this study presents a major limitation in understanding the direction of associations sleep and adjustment in adolescents with SB. There is limited longitudinal data on sleep in SB and other pediatric populations. Our previous work employed longitudinal growth modeling to understand trajectories of sleep disturbances in children and adolescents with SB, yet utilized a limited measure of sleep disturbances drawn from a behavioral screening tool (the CBCL; Murray et al., 2015). Future longitudinal research may examine trajectories, predictors, and consequences of sleep-wake disturbances using multi-wave, multi-modal sleep assessments. Further, conducting sleep interventions and experimental sleep extension trials may increase our understanding of the mechanisms and consequences of poor sleep in this vulnerable medical condition.

Clinical Implications and Conclusions

Despite limitations, the results of this study have important clinical implications. Unfortunately, sleep disturbances in children with developmental and physical disabilities often go unrecognized in clinical settings (Mindell & Owens, 2003). Extending the limited data on sleep disturbances in individuals with SB, results of our study suggest that adolescents with SB are at-risk for significant nighttime sleep disturbances and fatigue that require early assessment and management. This finding is clinically important given evidence that sleep disturbances may persist or recur over time (Morrison, McGee, & Stanton, 1992; Roberts et al., 2008) and are connected to adverse outcomes highly relevant to individuals with SB (e.g., BMI, executive dysfunction). Early assessment and prevention efforts may be critical to prevent long-term sleep problems and optimize health and well-being in this population.

Currently, sleep disturbances among adolescents with SB may be under-identified and undertreated. Given the gap in literature on sleep in pediatric SB, parents and health professionals may be more likely to attend to other salient aspects of SB management (e.g., adherence to a complex medical regimen), and inadvertently neglect to discuss sleep-wake patterns. Yet, results of this study and our previous work (Murray et al., 2015) suggest that adolescents with SB are vulnerable to the development of sleep disturbances that requires early detection through regular screening. Quick and effective multidimensional screening tools may include the BEARS (B = Bedtime Issues, E = Excessive Daytime Sleepiness, A = Night Awakenings, R = Regularity and Duration of Sleep, S = Snoring (Owens & Dalzell, 2005). Furthermore, modification of existing sleep measures to include items geared towards identifying modifiable and non-modifiable condition-related issues that underlie delayed bedtime routines

and disrupted nighttime sleep (e.g., lengthy medical management regimens) will help tailor interventions and clinical recommendations for youth with complex medical conditions such as pediatric SB.

Given the risk for sleep disturbances in adolescents with SB and potential adverse effects of sleep on critical health outcomes, there is a clear need to increase prevention and intervention efforts in this population. Behavioral sleep interventions used in physically healthy populations have been used successfully in adolescents with autism and ADHD, and have been shown to ease daytime behavioral symptoms (e.g., Papadopoulos et al., 2015). Due to the complex medical presentation of SB, the intensity and specific components of behavioral sleep treatment will need to be modified to fit individual needs. Nevertheless, our results provide insight on potential treatment targets, including modification of nightly bedtime routines (i.e. preparing for bed and conducting nighttime medical management earlier), improving physical sleep environments (i.e., dim light, reduced noise, limiting TV or phone use), and increasing parental monitoring to establish earlier and consistent bedtimes/wake times. For youth with more significant insomnia issues (i.e., difficulties staying asleep), implementation of behavioral sleep techniques (e.g., stimulus control therapy) may also be appropriate. Identification of individual (e.g., developmental ability) and contextual barriers (e.g., socioeconomic status and family stress) impacting bedtime routines and nighttime sleep in this population will also be critical to help tailor sleep treatments in this population. Overall, prevention and intervention efforts aimed to ameliorate sleep disturbances may have a far-reaching impact on improving health and functioning in adolescents with SB. Increased recognition and treatment of sleep disturbances

represents a unique opportunity to improve overall health and well-being in individuals with SB and their families.

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VITA

Dr. Caitlin Murray earned her doctoral degree in clinical psychology from Loyola University Chicago. She received her B.A. in psychology from the University of North Carolina at Chapel Hill. Upon acceptance into Loyola's graduate program, Dr. Murray gained extensive research experience under the direction and supervision of Dr. Grayson Holmbeck. As part of Dr. Holmbeck's research lab, she helped to conduct longitudinal research examining family, social, emotional, physical and neuropsychological functioning in youth with spina bifida. During her graduate training, Dr. Murray realized a strong interest in project development and grant writing and was fortunate to be awarded the Ruth L. Kirschstein National Research Service Award (NRSA). As the basis for her dissertation, this pre-doctoral research grant enabled her to collect novel data on the prevalence and impact of sleep disturbances in an underserved population- adolescents with spina bifida. Currently, Dr. Murray is a Pediatric Psychology Intern at Nationwide Children's Hospital in Columbus, OH. She will continue her training in pediatric pain and behavioral sleep research as a Postdoctoral Fellow at Seattle Children's Research Institute in Seattle, WA.

