



2017

Depressive Symptoms, Neuropsychological Functioning, and Self-Management in Youth with Spina Bifida: Direct, Mediating, and Reciprocal Pathways

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LOYOLA UNIVERSITY CHICAGO

DEPRESSIVE SYMPTOMS, NEUROPSYCHOLOGICAL FUNCTIONING, AND SELF-
MANAGEMENT IN YOUTH WITH SPINA BIFIDA: DIRECT, MEDIATING, AND
RECIPROCAL PATHWAYS

A THESIS SUBMITTED TO
THE FACULTY OF THE GRADUATE SCHOOL
IN CANDIDACY FOR THE DEGREE OF
MASTER OF ARTS
PROGRAM IN CLINICAL PSYCHOLOGY

BY
ALEXA R. STERN
CHICAGO, IL
DECEMBER 2017

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ACKNOWLEDGEMENTS

I would like to thank Dr. Grayson Holmbeck for his encouragement throughout this project. Thank you for your guidance and support during this process. I would also like to express my gratitude to Dr. Noni Gaylord-Harden, for her thoughtful input. Finally, I would like to thank the families of youth with spina bifida who have participated in the CHATS study. I am inspired by your resilience. Without you, our research would not be possible.

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ABSTRACT

Although successful self-management of health care responsibilities is critical to meeting the developmental demands associated with the transition to adulthood in youth with spina bifida (SB), research on individual factors impacting medical responsibility in this population is sparse. Given the increased risk for cognitive deficits and development of depressive symptoms in this population, this study aimed to examine two pathways through which depressive symptoms and neuropsychological dysfunction may be associated with medical autonomy in youth with SB. First, it was hypothesized that neuropsychological functioning would mediate the relationship between depression and self-management. Second, an alternative model was tested whereby it was expected that depressive symptoms would mediate the relationship between neuropsychological dysfunction and self-management.

Participants were recruited as part of a larger, longitudinal study. The study's sample included 114 youth with SB (M age = 10.96 at Time 1). Data were collected at three time points, each spaced approximately two years apart. Youth, their parents, and their teachers completed questionnaires on child depressive symptoms, child neuropsychological functioning, and child self-management behaviors. Youth also completed a brief test battery assessing executive functioning.

Greater deficits in attention and working memory, and more severe depressive symptoms predicted lower levels of medical responsibility over time. Unique relationships were found

among depressive symptoms and individual cognitive deficits. Bootstrapped mediation analyses revealed that teacher-reported depressive symptoms significantly mediated the respective relationships between attention and working memory, and medical responsibility (all p 's < .05), but that neuropsychological dysfunction did not mediate the relationship between depressive symptoms and medical responsibility. It is hoped that this research will inform the development of evidence-based interventions aimed at improving and fostering the development of self-management in youth with SB.

CHAPTER ONE

INTRODUCTION

Spina bifida (SB) is a congenital birth defect that results in several medical complications, including orthopedic, urinary, bowel, and neurological difficulties. To manage these medical problems and minimize the risk of developing secondary health complications, children with SB must adhere to a complex and demanding medical regimen (Copp et al., 2015). Research shows that SB health care responsibilities are gradually transferred from family- to youth-management across adolescence (Stepansky, Roache, Holmbeck & Schultz, 2010). Successful transition of these health care responsibilities from family- to self-management enables the child to function independently at home and within the community (Beacham & Deatrck, 2013; Dicianno et al., 2008). However, research specifically investigating self-management behaviors in youth with SB remains sparse.

Modi et al.'s (2012) conceptual model of pediatric self-management postulates that several individual and contextual factors contribute to self-management outcomes. One potentially important modifiable individual factor that may impact SB self-management is child depressive symptoms. Youth with SB are at a significantly greater risk for developing depressive symptoms compared to healthy peers (Appleton et al. 1997; Holmbeck et al., 2003). This increased likelihood may be attributed to the social and academic difficulties youth with SB experience at school, as well as negative self-perceptions of physical appearance and lower self

-esteem associated with the stigma of having a physical disability (Holmbeck et al., 2003; Oddson, Clancy, & McGrath, 2006).

While the relationship between depressive symptomology and self-management has not been explored in youth with SB, depressive symptoms in adults with SB and in youth with other chronic illnesses have been associated with poorer self-management (Guo et al., 2013; Kennard et al., 2004). Evidence suggests that neuropsychological dysfunction may be one mechanism through which depressive symptoms negatively impact self-management. Higher order attention and executive functioning skills allow a child or adolescent to plan, problem-solve, engage in goal-directed behavior, and regulate cognitions and emotions in order to meet the multifaceted demands of their medical regimen (Lansing & Berg, 2014). Depressive symptoms have been shown to negatively impact attention, concentration, working memory, inhibitory control, planning abilities, and mental flexibility in typically developing individuals (Crocker, 2013). Associations between depressive symptomology and executive functioning have been similarly demonstrated in youth with SB and youth with pediatric multiple sclerosis (Kelly et al., 2012; Holland, Graves, Greenberg, & Harder, 2014).

Neuropsychological functions are particularly important to examine when studying self-management outcomes in youth with SB, as they are already prone to multiple cognitive impairments. SB is associated with lower average IQs, as well as difficulties with executive functioning and attention (Copp et al., 2015). These deficits are related to lower treatment adherence and medical autonomy in youth with SB (Psihogios, Murray, Zebracki, Acevedo, & Holmbeck, 2016; O'Hara & Holmbeck, 2013), which are components of self-management. Interestingly, despite the high rate of depressive symptoms and neuropsychological dysfunction

associated with SB, research has yet to examine these variables in relation to self-management outcomes.

Furthermore, there is an ongoing debate in the current literature with regard to causal relationships between depressive symptoms and neuropsychological functioning. Accompanying findings that demonstrate how depressive symptoms adversely impact cognitive functioning (Ahern & Semkowska, 2016) is a body of research that shows deficits in executive functioning and attention can put individuals with spina bifida at risk for the development of future depressive symptoms (Lennon, Klages, Amaro, Murray, & Holmbeck, 2015). Thus, it is also possible that one pathway through which neuropsychological impairment can have adverse consequences on the development of medical autonomy and medical adherence in youth with spina bifida is via increased depressive symptoms.

The current study seeks to address gaps in our understanding by testing longitudinal, multi-method, and multi-informant models of these individual factors (see Figures 1a-1b). The following sections provide an overview of the current research on self-management behaviors, depressive symptomology, and neuropsychological functioning in youth with spina bifida. Additionally, past research supporting the hypothesized mediational pathways among these constructs is presented. Weaknesses and gaps in the current literature are identified. Lastly, a detailed description of the current study is provided. It is hoped that the proposed research will inform the development of evidence-based interventions aimed at improving and fostering the development of self-management in this population.

Figure 1a. Mediational Model of Depressive Symptoms, Neuropsychological Functioning, and Self-Management

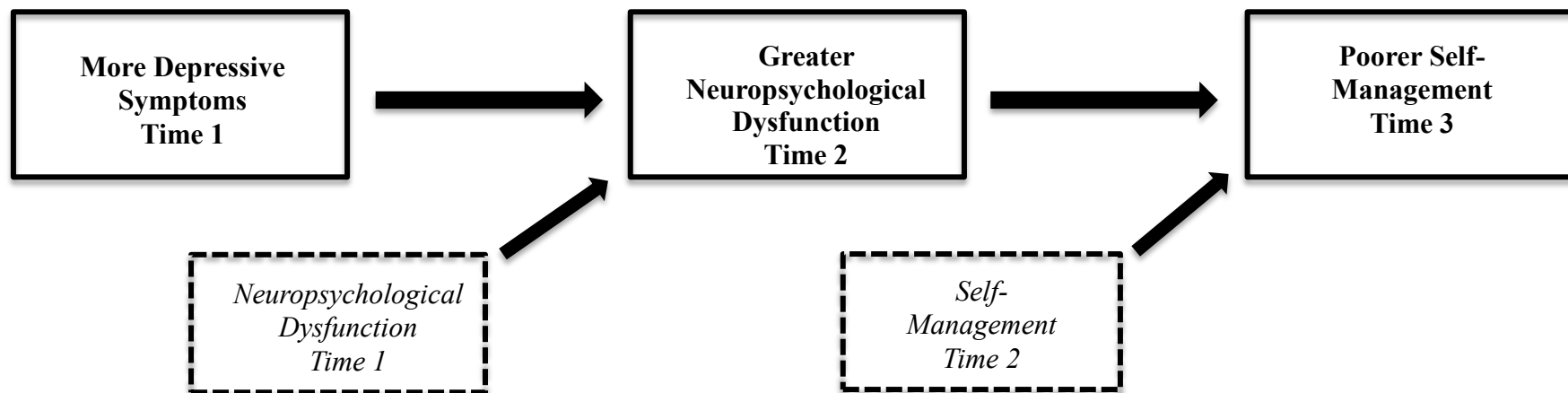
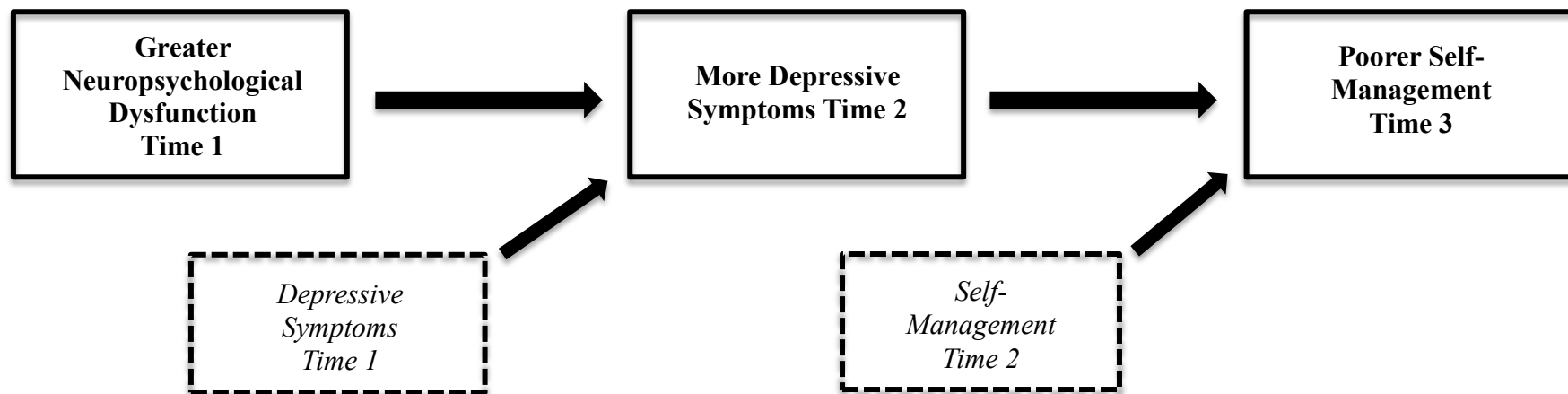


Figure 1b. Mediational Model of Neuropsychological Functioning, Depressive Symptoms, and Self-Management



CHAPTER TWO
REVIEW OF THE RELEVANT LITERATURE

Overview of Spina Bifida

Spina bifida is a relatively common congenital birth defect that results from failure of the neural tube to close during embryonic development, affecting one of every 1400-1500 births (American Association of Neurological Surgeons [AANS], 2015; Adzick et al., 2011; Mahmood, Dicianno, & Bellin, 2011). The most frequent and severe form of spina bifida, which accounts for approximately 75% of all cases, is myelomeningocele spina bifida (SBM); with SBM, there is a protrusion of the spinal cord and meninges into a sac filled with cerebrospinal fluid. Other complex congenital disorders, such as Arnold-Chiari II malformation (i.e., a structural defect of the cerebellum and hindbrain) and hydrocephalus (i.e., swelling of the brain due to excess cerebrospinal fluid, resulting in increased intracranial pressure) are present in up to 80-90% of children born with SBM (AANS, 2015; Dennis et al., 2006; Vick, Maassen, Mullaart, & Rotteveel, 2006). Spina bifida is a heterogeneous disorder, with the spinal lesion level affecting condition severity and individual functioning across several functional domains (Copp et al., 2015; Fletcher & Brei, 2010).

Spina bifida is associated with multiple medical complications. The most visible complications are linked to motor and orthopedic difficulties. Depending on lesion level, children can have loss of motor and sensory function and musculoskeletal anomalies in various

parts of their feet, legs, and pelvic region (Copp et al., 2015; Fletcher & Brei, 2010). Disruption of these motor and sensory nerves can require the use of assistive devices for ambulation, such as braces, crutches, or wheelchairs (Sandler, 2010). Many children with spina bifida (50% of SBM) are born with a foot deformity, which can worsen over time from muscle imbalance, muscle weakness, and growth processes. Surgery and bracing may be necessary to improve mobility. Spina bifida can also cause asymmetric hips and muscle imbalance, leading to scoliosis and pressure sores as the child grows (Sandler, 2010). Between 15 and 25% of children with spina bifida are born with scoliosis, but this secondary condition is also caused by or exacerbated in spina bifida due to tethered cord syndrome (i.e. where the spinal cord becomes attached or fixed to the spinal column, causing abnormal stretching and restricted movement of the spinal cord) or hip instability (AANS, 2015).

Spina bifida also impacts the nerves related to the bladder, urethra, and rectum, causing neurogenic bladder, bowel dysfunction, and sexual dysfunction in many individuals (Copp et al., 2015; Sandler, 2010). With a neurogenic bladder, children are unable to sense bladder fullness, which leads to issues with continence and kidney problems. Most children with spina bifida are also born with a neurogenic bowel, leading to difficulties with bowel mobility and constipation (Sandler, 2010).

Children with spina bifida are typically confronted with multiple neurological insults. Often, infants are born with Chiari II malformation, a complex brain malformation that consists of the displacement of the cerebellum, compression of the medulla, elongation of the fourth ventricle, and dysgenesis of the corpus callosum. The compression on the brainstem caused by the Chiari II malformation frequently causes hydrocephalus, which is associated with multiple

cognitive complications (Sandler, 2010). The Chiari II malformation and resulting hydrocephalus are typically accompanied by and can exacerbate the presentation of oculomotor disorders and fine motor dysfunction, leading to further complications.

Among individuals with Chiari II malformations, 90% receive ventriculoperitoneal shunts to relieve excess cerebrospinal fluid pressure and control ventricular volume (Sandler, 2010). Shunt placement is another factor associated with neurologic difficulties. Moreover, fifteen to twenty percent of children with spina bifida have seizures in childhood, which is exacerbated by the presence of a shunt placement. Additionally, seizures may signify a shunt malfunction. Shunt failure can cause headaches, changes in mood, lethargy, vomiting, impaired attention, and coordination. Unfortunately, approximately 40% of newly placed shunts fail within one year, and 80% fail within ten years, usually requiring multiple surgeries to revise or replace the shunt. Spina bifida is also associated with tethered cord syndrome (AANS, 2015). The spinal cord can become stretched and strained, leading to difficulties walking, back and leg pain, spasticity, worsening of scoliosis or foot deformity, and deterioration in bladder and bowel function (Sandler, 2010).

Due to neurologic dysfunction, children with spina bifida frequently have cognitive impairments, such as difficulties with abstract reasoning, visual perceptual abilities, and visual motor integration (Fletcher & Brei, 2010). Spina bifida is associated with below-average IQ, as well as difficulties with executive functioning (EF), attention, and organization (Copp et al., 2015; Sandler, 2010). A more detailed description of the neuropsychological functioning in youth with spina bifida will be provided in the following sections.

In addition to these primary complications, individuals with spina bifida are at risk for secondary health complications. These can include obesity, short stature, latex allergy, urinary tract infections (UTIs), and gastrointestinal disorders (Mayo Foundation for Medical Education and Research, 2014). Children with spina bifida are also at risk for developing pressure sores due to reduced sensation in lower extremities (Sandler, 2010). Given these pervasive health complications, children with spina bifida must adhere to a complex medical regimen often prescribed by a multidisciplinary team of physicians, nurses, social workers, and psychologists (Copp et al., 2015). They are required to manage a variety of tasks on a daily basis, including clean intermittent catheterization, bowel management programs, administration of medications, routine skin checks, and identifying shunt malfunctions or infections (O'Hara & Holmbeck, 2013). As these tasks are essential to maintaining the health of individuals with spina bifida, disease management is an extremely important part of their daily lives and care.

Self-Management in Spina Bifida

As advances in medicine are enabling children with spina bifida to survive into adulthood, and thus utilize adult health care services, successful transition of health care responsibilities from family to youth has become a critical component of pediatric development (Beacham & Deatrick, 2013; Dicianno et al., 2008). According to Modi et al.'s (2012) comprehensive conceptual model of pediatric self-management, self-management is “the interaction of health behaviors and related processes that patients and families engage in to care for a chronic condition.” Self-management processes include treatment adherence behaviors, responsibility for health-related tasks, and knowledge of disease-specific skills. Using this conceptualization, a child or adolescent must not only understand his or her medical condition

and the activities required to manage this condition on a daily basis, but also learn the specific health skills and undertake responsibility for enacting those skills (Binks, Barden, Burke & Young, 2007). The extent to which a child or adolescent with a chronic medical condition masters these tasks of self-management affects both individual and systemic health-related outcomes, such as secondary complications, quality of life, symptoms control, treatment efficacy, and financial healthcare costs (Modi et al., 2012).

Self-management behaviors specific to youth with spina bifida include: appointment keeping, self-advocacy (e.g. explaining spina bifida to peers in school), managing medication regimens and related medical supplies, taking preventative action for secondary health complications, and effectively adhering to a prescribed bladder and bowel program. Pediatric self-management takes into account developmental and contextual factors (e.g., influence of siblings, peers at school), and occurs across individual, family, community, and healthcare system domains. Within each domain, modifiable (e.g., disease knowledge) and nonmodifiable factors (e.g., IQ, insurance coverage) influence self-management outcomes. As this study focuses specifically on individual factors related to self-management (i.e., depressive symptoms and cognitive functioning), a discussion of each of the four domains is beyond the scope of this review; thus, this review of literature will focus only on the relevant influences within the individual domain.

Factors Associated With Self-Management

Self-management is a dynamic and fluid process that unfolds over a period of years. A successful transition from family- to of self-management likely depends on a number of child factors. Developmental stage and age play a pivotal role in self-management, as young children

may not have the capacity to care for themselves. Self-management also relies on physical, cognitive, and psychosocial abilities. A child with physical disabilities may not have the strength or dexterity to maintain and manage his or her healthcare regimen. Children must have the cognitive abilities necessary to perform complicated medical tasks, monitor their health and recognize changes in symptoms or functioning that may indicate improvement or worsening in health, and make informed decisions concerning their condition (e.g., knowing when to contact the doctor). They must also have the emotional maturity, self-regulation, and executive functioning skills to maintain their treatment in various settings (e.g., home, school, social situations; Beacham & Deatrick, 2013; Modi et al., 2012).

Autonomy is a major normative developmental goal for adolescence, and individuals with spina bifida are often interested in becoming autonomous with respect to their various self-management tasks (e.g., bladder and bowel care, skin checks), seeking to function independently and autonomously at home and within the community (Holmbeck & Devine, 2010). However, some children and young adults with spina bifida have developmental delays in self-help skills, resulting in lower levels or a delay in the acquisition of independent functioning (Andren & Grimby, 2004; Greenley, Holmbeck, Zukerman, & Buck, 2006; Varni & Wallander, 1984). This increased dependency on caregivers and delayed independence in activities of daily living are often associated with higher lesion levels and lower cognitive functioning (Sirzai et al., 2014).

While individuals with the most severe forms of spina bifida may not be able to self-manage all of the skills related to their healthcare due to cognitive or physical limitations, health professionals report that individuals with moderate and mild forms of spina bifida should be able to independently manage most of these tasks before adulthood (Greenley, 2010). Healthcare

providers believe that mildly to moderately impaired individuals with spina bifida should be able to master critical tasks such as self-catheterization and skin care checks during the elementary school years, and independently manage their bowel program by the middle school years. Severely impaired individuals with spina bifida may be expected to manage some of these tasks by high school or during post-high school years (Greenley, 2010).

Despite these clinical recommendations, adolescents with spina bifida may encounter difficulties when attempting to develop autonomy and assume self-care responsibility across contexts. Findings from a study comparing preadolescents with spina bifida and typically developing peers established that children with spina bifida were more passive, more dependent on adults for direction and guidance, less likely to make independent decisions, and responsible for fewer tasks at home, suggesting that developing autonomy poses a significant challenge for this population (Holmbeck et al., 2003). While children with spina bifida show development in behavioral and emotional autonomy during adolescence, they continue to lag behind their typically developing peers. Intrinsic motivation (i.e., behavior driven by internal rewards) may be particularly difficult for children with spina bifida as compared to their peers. Indeed, their level of intrinsic motivation during school at preadolescence has been shown to be lower and tends not to increase with age (Friedman et al., 2009).

These challenges in assuming responsibility apply to medical and health related tasks as well. A study of adolescents with spina bifida and cerebral palsy showed that approximately one-quarter of adolescents felt their parents infantilized them, and they perceived the constant reminders regarding self-management as parental overprotection. Additionally, one-third of participants with spina bifida in this sample were highly dependent on parental involvement in

their bowel programs (Blum, Resnick, Nelson, & Germaine, 1991). Longitudinal findings support a developmental trajectory where youth with spina bifida gradually gain responsibility for medical tasks such as catheterization and bowel program management over time. On the other hand, their adherence is linked to family functioning, such that family conflict is associated with a decrease in adherence (Stepansky, Roache, Holmbeck, & Schultz, 2010).

While responsibility for medical tasks in youth with chronic health conditions usually transitions from family to child during adolescence, treatment adherence rates tend to decrease during this developmental period, with rates as low as 50% in some pediatric populations (La Greca & Mackey, 2009). This finding aligns with the developmental literature, which asserts that adolescents do not completely develop the cognitive, emotional, and behavioral self-regulation skills that underlie the foundation of successful self-management until early adulthood (Lansing & Berg, 2014). A recent study of self-management behaviors in youth with spina bifida confirmed that, while children gained responsibility for medical care over time, rates of nonadherence remained high across late childhood and adolescence, with rates approaching 50% for some tasks (i.e. skin checks) in 12-13 year olds (Psihogios, Kolbuck, & Holmbeck, 2015). Thus, it is essential to study processes influencing self-management in youth with spina bifida, as increased understanding of these factors will help inform clinical interventions that support self-management and autonomy across development in this population.

Depressive Symptoms and Self-Management

Few studies have been conducted to isolate individual modifiable risk factors that are associated with poor self-management in youth with spina bifida. One potentially important modifiable factor to explore is depressive symptomology. High levels of depressive symptoms in

adolescence are related to poorer objective and subjective ratings of health in early adulthood, even in the absence of a chronic illness (Keenan-Miller, Hammen, & Brennan, 2007). Depressive symptoms are significantly more prevalent among chronically ill children and adolescents, as compared to their healthy peers (Turkel & Pao, 2007; Kline-Simon, Weisner, Sterling, 2016), and are predictive of increased medical complications in adulthood (Katon, Lin, & Kroenke, 2007).

Depressive symptoms may compromise self-management by decreasing an individual's motivation, self-efficacy, decision-making, problem-solving, attention, and concentration abilities that are required to complete healthcare-related tasks on a daily basis (Modi et al., 2012). A meta-analysis found that depressive symptoms were a risk factor for noncompliance with medical treatment across a variety of chronic illnesses in adults, with depressed individuals being three times as likely as non-depressed individuals to be nonadherent (DiMatteo, Lepper, & Croghan, 2000). Higher rates of adverse health behaviors have also been found among depressed adults with a chronic illness; depressive symptoms are associated with poor diet and exercise, as well as an increase in harmful behaviors such as drinking alcohol and smoking among those with heart disease and diabetes (Katon, 2003).

The literature on the relationship between depressive symptoms and health behaviors in individuals with spina bifida focuses primarily on young adults. Past research indicates that depressive symptoms are associated with poor self-rated health and unhealthy behaviors in young adults with spina bifida, including alcohol abuse, poor physical activity, and poor diet (Soe et al., 2012). Similarly, a longitudinal study demonstrated that a decrease in depressive

symptoms over time was associated with an increase in spina bifida management competencies in adults with spina bifida (Bellin et al., 2010).

Further support for the deleterious impact of depressive symptoms on pediatric self-management has been found in other chronic illness populations. Depression in diabetes has been associated with non-adherence, greater hospitalization rates, and more medical complications (Snoek & Skinner, 2006). In youth with type 1 diabetes, depressive symptoms were associated with a decrease in energy and motivation to complete complex diabetes care-related tasks (Guo et al., 2013). During adolescence, the risk for deterioration in metabolic control increases, and is associated with affective and social problems (Leonard, Jang, Savik, & Plumbo, 2005).

Depressive symptoms in children predicted an increase in parent responsibility for child diabetes management over time, suggesting that parents may compensate for a child's mental health difficulties (Helgeson, Reynolds, Siminerio, Escobar, & Becker, 2008). Interestingly, a study of pediatric oncology patients found a relationship between depressive symptoms, self-esteem, and adherence even though the sample scored within the normative range on the Beck Depressive Symptoms Inventory, suggesting that mood influences self-management at a sub-clinical level (Kennard et al., 2004). Thus, the relationship between depressive symptoms and self-management behaviors warrants more exploration in a spina bifida youth population.

Depressive Symptoms in Youth with Spina Bifida

Research has shown that youth with spina bifida, especially adolescents, are at a significantly greater risk for developing depressive symptoms compared to healthy peers (Appleton et al. 1997; Holmbeck et al., 2003; Pit-ten, Kennedy, & Stevenson, 2002). They may also have more depressive symptoms compared to youth with similar physical disabilities, such

as early onset spinal cord injury (Flanagan, Kelly, & Vogel, 2013). Possible predictors of elevated depressive symptoms in children with spina bifida include difficulties with social acceptance and social support, poorer family functioning, negative perceptions of physical appearance, lower self-worth, and higher levels of pain (Oddson, Clancy, & McGrath, 2006; Holmbeck, et al., 2010). There is also evidence for a mediational role of self-worth in the relationship between self-evaluations of physical appearance and depressed mood; youth with spina bifida may experience daily challenges with mobility, self-management, and toileting difficulties, thus creating more negative body-related perceptions (Appleton et al., 1997).

Developmental factors may influence the onset of depressive symptoms in youth with spina bifida, given that preadolescents (8-9 years) with spina bifida do not differ significantly from typically developing peers in internalizing symptoms (Holmbeck et al., 2003). Social difficulties in children with spina bifida at this age may stem from poor social engagement and social maturity, increasing the likelihood that depressive symptoms will develop during adolescence (Holmbeck et al., 2003). Adolescents with spina bifida have fewer positive experiences across social (i.e. school and peer) contexts than typically developing youth which, in turn, are related to poorer psychological adjustment and greater depressive symptoms (Essner, Holmbeck, & Elliot, 2010). Spina bifida-management tasks may contribute to lower levels of positive experiences, as many adolescents are concerned with navigating the social consequences of incontinence, catheterization, and bowel management in a peer setting, as well as the time demands that bladder and bowel programs place on leisure activities and social interactions (Lindsay, 2014).

In addition to school and peer settings, parenting behaviors have a cumulative effect on depressive symptoms in youth with spina bifida. Pre-adolescents with mothers who demonstrate less acceptance, greater psychological control, and greater behavioral control are at risk for developing more depressive symptoms. Once a child enters adolescence, maternal depressive symptoms pose an additional risk for the development of depressive symptoms, suggesting that parenting impacts a child's mental health differentially across development (Schellinger, Holmbeck, Essner, & Alvarez, 2012). Consistent with this line of research, perceived lack of parental support is significantly associated with depressed mood and low global self-worth (Appleton, 1997). In a study of adolescents with spina bifida and typically developing youth, parental warmth was negatively associated with adolescent depressive symptoms and maternal criticism was positively associated with depressive symptoms. However, maternal criticism at mid-adolescence was only predictive of depressive symptoms in late adolescence in those with spina bifida, suggesting that, within the context of depressive symptoms, an increased dependence on parents makes youth with spina bifida particularly vulnerable to the effects of parenting (Kelly, Holmbeck, and O'Mahar, 2011).

Depressive symptoms in children with spina bifida are strongly associated with negative outcomes such as poor quality of life (Leger, 2005; Oddson, Clancy, & McGrath, 2006). Disability status may interact with psychological adjustment to impact quality of life, as severely disabled adolescents with spina bifida have greater self-esteem and perceive themselves as having a higher emotional quality of life than their less disabled counterparts, while less disabled adolescents have higher emotional distress and, as a result, greater difficulty in daily activities (Padua et al., 2002). Padua et al. (2002) hypothesized that this finding may have been due to

individuals with a lower disability engaging in more social activity among healthy peers, and feeling more motivated to mask their physical differences. There appears to be a complex relationship among these variables, as research does not support a relationship between severity of physical disability and future internalizing problems (Hommeyer, Holmbeck, Wills, & Coers, 1999).

Pain management is another area of functioning in youth with spina bifida that is closely tied to depressive symptoms. Children with spina bifida may experience more frequent and severe pain than their healthy peers, which may take a toll on their emotional health and lead to a decrease in quality of life. Specifically, frequency of pain and severity of worst pain are linked to greater depressive symptoms (Oddson, Clancy, McGrath, 2006). This pain may present as joint and muscle pain in the lower extremities due to spasticity or utilizing assistive ambulatory devices (Rimmer, Rowland, & Yamaki, 2007). Interventions have targeted self-management in spina bifida to improve strategies in managing pain symptoms and encouraging lifestyle changes to alleviate pain, but depressive symptoms may interfere with the effective learning and application of these skills (Froehlich-Grobe, Driver, Sanches, 2016).

The prevalence of depressive symptoms among youth with spina bifida endures into adulthood. In one study of adults with spina bifida, over 50% reported experiencing depressive symptoms, and 87% of this subsample perceived their symptoms to “somewhat” or “greatly” impact their daily lives (Wagner et al., 2015). This finding further emphasizes that depressive symptoms are an important variable to be included in studies of self-management in this population, given the detrimental impact depressive symptoms likely have on self-management. The negative relationship between depressive symptomology and self-management may be

compounded by the adverse effects that depressive symptoms have on other areas of independence, including workplace functioning and maintaining a healthy relationship with a spouse or partner (Judd et al., 2000).

Neuropsychological Functioning in Youth with Spina Bifida

Based on prior findings, which assert a relationship between depressive symptoms and self-management issues, research needs to examine the process through which depression may influence a child's self-management of spina bifida. One possible mechanism is that depressive symptoms may exacerbate pre-existing neuropsychological deficits, which affects the higher order cognitive skills required to complete complex spina bifida self-management tasks. The neurodevelopmental effects of spina bifida may complicate this relationship, as spina bifida is associated with deficits in various dimensions of IQ, attention, and executive functioning. These congenital neurocognitive differences in children with spina bifida stem from hydrocephalus and the Chiari II malformation, two neural insults that produce other structural and functional abnormalities in the developing brain.

The following section discusses the neuropsychological profile of youth with spina bifida, with a focus on intelligence, attention, and executive functions. It should be noted that although these studies depict a prototypical description of a child with spina bifida's neuropsychological capabilities, a large degree of variability exists within the spina bifida population. Medical factors, such as the presence of hydrocephalus, Chiari II malformation, shunt complications, and higher lesion level, as well as demographic (e.g., lower socioeconomic status, child age) and familial factors (e.g., higher levels of parental stress) can impact the degree of neuropsychological impairment (Copp et al., 2015; Erickson, Baron, & Fantie, 2001; Rose &

Holmbeck, 2007; Barf et al., 2003; Brown et al., 2008; Bier et al., 1997; Holmbeck et al., 2003; Swartout, Garnaat, Myszka, Fletcher, & Dennis, 2010). The relative deficits and strengths reported below are meant to provide a framework for better understanding youth with spina bifida and some of the cognitive challenges they may face.

Intelligence

Intellectual disability affects roughly 20-25% of individuals with spina bifida myelomeningocele (SBM; Copp et al., 2015). The majority of children with spina bifida tend to obtain IQ scores in the average to low-average range (Crawley et al., 2014; Ramsundhar & Donald, 2014). In general, children with spina bifida exhibit relatively preserved verbal intelligence (VIQ), but weakened non-verbal or performance-based intelligence (PIQ) (Iddon et al., 2004; Fletcher et al., 1992). However, some studies have shown evidence for poorer performance on tests of verbal ability and reasoning skills, often used as a proxy for general intellectual functioning in youth with spina bifida compared to typically developing children (Tuminello, Holmbeck, & Olson, 2012; Burmeister et al., 2005). Those with SBM or spina bifida with hydrocephalus may be at a particular disadvantage, as they tend to display greater deficits in visual or abstract reasoning, verbal learning, and FSIQ scores than their spina bifida counterparts without hydrocephalus (Erickson, Baron, & Fantie, 2001; Burmeister et al., 2005). In children with hydrocephalus, increased cerebrospinal fluid pressure and ventricle size in the posterior brain regions, as well as malformations in the corpus callosum (i.e., the major white matter tract that connects the left and right cerebral hemispheres), were associated with the observed lower IQ scores and poorer information processing (Crawley et al., 2014; Fletcher et al., 1992; Fletcher et al., 1996; Erickson, Baron, & Fantie, 2001).

Attention

Studies show that children and adolescents with spina bifida also consistently perform worse than typically developing peers on measures of specific types of attention, and that these deficits persist after controlling for differences in intellectual functioning (Vinck, Mullaart, Rotteveel, & Maassen, 2009; Rose & Holmbeck, 2007). Results from traditional attention tests in this population may be confounded by the tests' dependency on visual-motor skills, as individuals with spina bifida often have deficits in visual-motor and fine motor domains (Vinck, Mullaart, Rotteveel, & Maassen, 2009).

Youth with spina bifida exhibit clinically significant deficits in focused attention, which reflects the ability to select specific stimuli from a broad array (Rose & Holmbeck, 2007; Vinck, Mullaart, Rotteveel, & Maassen, 2009). This population often exhibits difficulties with selective attention (i.e., the ability to restrict concentration to a target stimulus in the face of distracting or competing stimuli), and distractibility, especially when there is a history of hydrocephalus (Ou et al., 2013; Caspersen & Habekost, 2013; Vinck, Mullaart, Rotteveel, & Maassen, 2009; Fletcher et al., 1996; Erickson, Baron & Fantie, 2001). The neural correlates of attentional deficits in individuals with SBM include structural and functional abnormalities in the posterior attention network, corpus striatal and inferior parietal regions, superior parietal and frontal lobes, cerebellum, midbrain, and corpus callosum, which can be caused by hydrocephalus and the Chiari-II malformation (Dennis & Barnes, 2010; Ramsundhar & Donald, 2014; Out et al., 2013; Rose & Holmbeck, 2007). Data on sustained attention, the ability to maintain concentration over time, are mixed with some studies showing preserved function in youth with spina bifida (Rose

& Holmbeck, 2007; Swartwout et al., 2008), and others demonstrating deficits in this area (Caspersen & Habekost, 2013; Brewer et al., 2001; Erickson, Baron, & Fantie, 2001).

Moreover, youth with spina bifida are more at risk for being diagnosed with ADHD (Rose & Holmbeck, 2007). Studies have found that almost one-third of children and adolescents with spina bifida presented with ADHD-Inattentive type symptoms, far exceeding the population rate of 8% (Burmeister et al., 2005; Ammerman et al., 1998). Rates of ADHD-Combined type and ADHD-Hyperactive type are comparable to normative rates in youth with spina bifida, suggesting that issues related to distractibility, lack of focus, and disorganization are particularly problematic as compared with the impulsiveness and hyperactivity that characterize typically developing children with ADHD (Burmeister et al., 2005; Ammerman et al., 1998). Individuals with spina bifida and ADHD display a different pathophysiology than typically developing children with ADHD, who tend to have more problems with sustained attention (Ramsundhar & Donald, 2014). Attentional abilities are tied to executive functioning, as children with spina bifida and hydrocephalus classified with ADHD show greater executive dysfunction than children with spina bifida without ADHD (Burmeister et al., 2005). While attention and executive functioning are distinct constructs, executive functioning skills are often implicated in the top-down processes that control voluntary attention (Snyder, Miyake, & Hankin, 2015).

Executive Functioning

As previous studies have demonstrated, executive functioning (EF) is another domain in which many individuals with spina bifida tend to have deficits. Executive functioning abilities are a constellation of processes related to 1) goal formulation, 2) planning, 3) carrying out goal-directed plans, and 4) engaging in effective performance (Lezak, Howieson & Loring, 1995).

Executive functions fall into two categories: behavioral regulation, which includes impulse control, cognitive shifting, and emotional control; and metacognition, which includes skills related to problem-solving, initiation, working memory, planning, organization, and self-monitoring. While EF abilities vary among youth with spina bifida, these individuals tend to demonstrate low-average EF, with scores on performance-based measures falling below one standard deviation of the normative mean (Heffelfinger et al., 2008). These deficits appear to persist after controlling for possible confounding factors such as IQ and motor impairment (Rose & Holmbeck, 2007; Lindquist, Persson, Uvebrant, & Carlsson, 2008; Dennis & Barnes, 2010).

The executive dysfunction demonstrated in performance-based assessments is corroborated by parent, teacher, and self-report (Tuminello, Holmbeck, & Olson, 2012; Mahone et al., 2002; Zukerman, Devine, & Holmbeck, 2011). While typically developing children tend to exhibit maturation in EF abilities with age (Xu et al., 2013), children with spina bifida do not share the same age-expected gains in behavioral and cognitive control across adolescence (Tarazi, Zabel, & Mahone, 2008). Beyond statistical significance, a greater proportion of youth with spina bifida myelomeningocele and hydrocephalus exhibit *clinically* significant problems in certain types of EF, with rates of clinically-elevated scores reaching 50% in metacognitive abilities such as initiation, working memory, planning, and organizing (Tarazi, Zabel, & Mahone, 2008).

With regard to behavioral regulation, inhibition abilities may be intact (Tarazi, Zabel, & Mahone, 2008; Rose & Holmbeck, 2007), but youth with spina bifida have impairments in flexible thinking and cognitive shifting, which allow one to think flexibly in order to respond appropriately to a situation (Tarazi, Zabel, & Mahone, 2008; Iddon et al., 2004; Rose &

Holmbeck, 2007; Tuminello, Holmbeck, & Olson, 2012). Those with SBM and hydrocephalus may have difficulties with emotional control as well (Iddon et al., 2004). Structural anomalies in the caudate and thalamus due to hydrocephalus are associated with behavioral regulation difficulties in SBM (Ware et al., 2016).

Research examining EF in youth with spina bifida has found deficits not only in behavioral and parent-report measures of behavioral regulation, but also metacognition. Results suggest that such youth have problems with initiating and generating ideas, responses, or problem-solving strategies, and initiating tasks independently. Spatial and visual working memory skills, specifically visuospatial sequencing, encoding, and memory span, are impaired in individuals with SBM and hydrocephalus (Mammarella, Cornoldi, & Donadello, 2003), as are planning, organizing, and goal-directed abilities, and self-monitoring behaviors (Rose & Holmbeck, 2007; Tarazi, Zabel, & Mahone, 2008; Burmeister, 2005; Iddon et al., 2004; Tuminello, Holmbeck, & Olson, 2012; Erickson, Baron, & Fantie, 2001).

Again, these deficits in executive functioning are related to structural anomalies in the brain caused by hydrocephalus and the Chiari-II malformation. Intracranial pressure due to hydrocephalus can stretch the various pathways connecting the hippocampus, temporal lobes, cortex, and basal ganglia, negatively impacting memory encoding and retrieval processes (Erickson, Baron, & Fantie, 2001). Hydrocephalus can cause damage to the white matter tracts that deliver and send information to and from the prefrontal cortex, which is heavily implicated in executive functioning (Fletcher et al., 1996). The damaged posterior attention systems and cerebellum present in youth with spina bifida and hydrocephalus also affect EF (Dennis & Barnes, 2010; Burmeister, 2005). The presence of hydrocephalus or Chiari-II malformation in

youth with spina bifida may cause additional deficits in abilities related to abstract thinking and the formation of concepts (Heaton et al., 1993).

Other Cognitive Processing Deficits

In general, youth with spina bifida have relative strengths and weaknesses in the different types of information processing systems that undergird various content-specific domains (e.g., math, reading, science). *Associative processing* is a relative strength for individuals with SBM. This system reflects the ability to generate information that has been linked to material in long-term memory (Fletcher, Ostermaier, Cirino, & Dennis, 2008; Copp et al., 2015). On the contrary, *assembled processing* poses a relative challenge for this population. This type of processing involves constructing and assimilating information across content domains. The difference in ability between these two processing systems likely reflects the relative preservation of verbal IQ compared to nonverbal IQ (Fletcher, Ostermaier, Cirino, & Dennis, 2008; Copp et al., 2015). Children with SBM exhibit difficulties in the perception of time and space, as well as impairments in specific mathematics skills such as estimation, problem solving, mental calculation, and manipulation of numbers, with difficulties emerging in the preschool years (Dennis & Barnes, 2010).

Certain aspects of language development and literacy can also be problematic for youth with spina bifida with hydrocephalus. Challenging areas can include impaired processing of constructed meaning during conversation (Dennis & Barnes, 2010), phonological awareness, semantics, fluency, and word retrieval (Brookshire et al., 1995; Erickson, Baron, & Fantie, 2001). They tend to have difficulties with abstract language comprehension (Barnes & Dennis, 1998; Erickson, Baron, & Fantie, 2001) and correct contextual use of language (Ramsundhar &

Donald, 2014), as well as with narrating coherent and cohesive stories (Dennis, Jacennik, & Barnes, 1994; Erickson, Baron, & Fantie, 2001). Some individuals with spina bifida may struggle with explaining, analyzing, and drawing contextually appropriate inferences from text (Ramsundhar & Donald, 2014; Dennis & Barnes, 2010). While these relative assets and deficits constitute the modal neurocognitive profile of youth with spina bifida, variability exists within the population. Differences in the nuanced presentation of these neuropsychological domains stem from a host of individual, familial, and contextual factors.

Association of Neuropsychological Functioning with Self-Management

Attention and executive abilities are employed consistently in tasks related to daily living. Regarding youth with spina bifida, better functioning in these higher order cognitive domains has been predictive of greater psychosocial adjustment, functional independence skills, and social competence (Coakley, Holmbeck, & Bryant, 2006; Heffelfinger et al., 2008; Jacobson et al., 2013; Lennon, Klages, Amaro, Murray, & Holmbeck, 2015). Greater executive functioning was also associated with an increased likelihood of youth with spina bifida achieving certain developmental milestones in young adulthood, such as leaving home and attending college (Zukerman, Devine, & Holmbeck, 2011). Research with pediatric chronic illness populations has extended this association between neuropsychological functioning and general adaptive outcomes to health-specific outcomes, including treatment self-management (Lansing & Berg, 2014; Dunbar-Jacob et al., 2000). The complexity of managing a chronic illness requires extensive cognitive demands, and can be compromised by cognitive immaturity or dysfunction. Indeed, a systematic review identified issues related to cognitive functioning, including forgetfulness, poor organizational skills, and difficulties with problem solving, as consistent

barriers to self-management of medications in adolescents across multiple chronic illness populations (Hanghøj & Boisen, 2014). Most research in this area has focused on type 1 diabetes (Bagner et al., 2007; Graziano et al., 2011, McNally et al., 2010), where EF has been found to be related to medical autonomy and adherence. Executive functioning impacts cognitive, emotional, and behavioral self-regulation skills, which can extend to interpersonal processes implicated in self-management, such as an adolescent's ability to draw on support from parents, peers, and healthcare providers in the context of their illness (Lansing & Berg, 2014). Similarly, attention and concentration problems have been shown to interfere with diabetes regimen compliance in adolescents, adversely impacting their efforts to manage their illness independently (Sanchez, Chronis, & Hunter, 2006).

Poor executive functioning skills have emerged as barriers to adherence and medical autonomy in youth with spina bifida. Executive dysfunction was predictive of non-adherence and less medical responsibility in bowel management with youth with spina bifida (Psihogios, Murray, Zebracki, Acevedo, & Holmbeck, 2016). Time processing ability, which is a component of EF, was associated with independence in clean intermittent catheterization in adolescents with spina bifida (Donlau et al., 2011). In one study, parental control appeared to buffer against the deleterious effects of poor executive functioning on adherence and medical autonomy (O'Hara & Holmbeck, 2013). Despite its close ties to executive functioning, deficits in attention have not yet been examined as a predictor of self-management in this population. These preliminary findings provide evidence for a relationship between higher order cognitive skills and self-management in spina bifida.

Relationship among Depressive Symptoms, Attention, and Executive Functioning

Research supports a robust association between depressive symptoms and neuropsychological deficits. Impairments in attention and executive functions have been well documented in depressive episodes (Snyder, 2013; Rock, Roiser, Riedel, & Backwell, 2014; McClintock et al., 2010). Specifically, mild to moderate cognitive deficits have been demonstrated in depressive episodes among adolescents and young adults, including: selective and sustained attention, working and episodic memory, inhibition, cognitive flexibility, problem-solving, planning, processing speed, and self-monitoring (Castaneda, Tuulio-Henriksson, Marttunen, Suvisaari, & Lonnqvist, 2008; Han et al., 2012; Kyte, Goodyer, & Sahakian, 2005). Interestingly, the relationship between depressive symptoms and neurocognitive impairment has been shown to vary across cognitive domains, implying that cognitive skills should be examined individually in relation to depressive symptomology (Gotlib & Joormann, 2010; McDermott & Ebmeier, 2009). Importantly, the severity of depressive symptoms at sub-clinical, dysphoric levels has been negatively associated with attention and executive functioning, indicating cognitive impairment can occur with depressive symptoms even in the absence of a diagnosable disorder (Gotlib & Joormann, 2010). Additionally, residual impairments in executive functions and attention have been shown to persist after reduction and remission of depressive symptoms (Hammar & Ardal, 2009), suggesting a lasting negative effect over time. Thus, it is clear that our emotional and cognitive systems are linked, and deficits in one domain may leave one vulnerable to deficits in the other.

However, it is unclear if these remaining deficits represent a pre-existing cognitive vulnerability to depressive symptoms or are a direct consequence of depressive symptoms.

Research on the directionality of the relationship between depressive symptoms and neuropsychological dysfunction has been debated and thus far is inconclusive (Snyder, Miyake, & Hankin, 2015). The cognitive “scarring” model posits that experiencing depressive symptoms impairs cognitive functioning and can lead to persistent deficits (McClintock et al., 2010; Maalouf et al., 2011). In support of this view, the number and severity of previous depressive episodes predicted residual cognitive deficits in remitted adults, even after controlling for residual depressive symptoms (Bhardwaj, Wilkinson, Srivastava, & Sharma, 2010). Impairments in executive functions, memory, and attention were detected in adolescents with acute depressive symptoms, but not in high-risk children of mothers with major depressive disorder, suggesting that these cognitive deficits could be conceptualized as a consequence of, rather than a risk factor for, the development of depressive symptoms (Wilkinson & Goodyer, 2006; Klimes-Dougan et al., 2006). It is suggested that these persistent deficits may be a result of neural changes that occur during a depressive episode (Ahern & Semkovska, 2016). However, it is difficult to make causal inferences, as the majority of studies to date have utilized cross-sectional designs and lacked prospective data.

Alternatively, the cognitive vulnerability hypothesis argues that neuropsychological deficits reduce one’s ability to cope with stressors, which puts the individual at increased risk for developing depressive symptoms (Lee et al., 2012). Further, children with cognitive problems may compare their poorer performance in school or other areas of achievement to the abilities of children without cognitive deficits, which may result in lowered self-esteem and depressive symptoms (Blechman, McEnroe, Carella, & Audette, 1986). Compared to monozygotic twin-pairs with no history of depression, unaffected individuals in monozygotic twin-pairs discordant

for a lifetime history of depression had greater impairments in attention and working memory, which lends support to the cognitive vulnerability account (Hsu, Young-Wolff, Kendler, Halberstadt, & Prescott, 2014). Difficulties with certain aspects of executive functioning (e.g., working memory) have predicted higher levels of depressive symptoms longitudinally in both typically developing adolescents and youth with spina bifida, even after accounting for severity of depressive symptoms at baseline (Evans, Kouros, Samanez-Larkin, & Garber, 2016; Lennon, Klages, Amaro, Murray, & Holmbeck, 2015). Given the high comorbidity between neurocognitive dysfunction and internalizing symptoms in youth with spina bifida, this relationship warrants more a fine-tuned investigation.

Examining this relationship from a developmental perspective is important, as child and adolescent patterns of depressive symptoms and neuropsychological functioning may differ from that of adults. The onset of adolescence coincides with rapid changes in emotional responses to social stimuli, as well as alterations in motivation and rewards systems (Giedd, Keshavan, & Paus, 2008). Meanwhile, executive functions, which underlie emotional and behavioral self-regulation, are not fully developed, and continue to mature into adulthood. The prevalence of depressive symptoms increases during adolescence, as teenagers must navigate novel and challenging environments with immature executive skills (Wagner, Alloy, & Abramson, 2015). These changes may underlie the onset of depressive and other affective disorders during adolescence (Giedd, Keshavan, & Paus, 2008). While existing knowledge points to a strong relationship between psychological and cognitive functioning in youth, it is not fully understood. This study seeks to further clarify this relationship.

Neural Correlates of Depressive Symptoms and Neuropsychological Dysfunction

The developmental neurobiology of depressive symptoms provides a deeper understanding of its relationship with neuropsychological functioning in youth. From a neurological standpoint, depressive symptomology may disrupt and dysregulate the brain processes responsible for a child's developing executive functions, and these deleterious effects may continue to affect a child after symptom remission, negatively impacting future executive functioning and self-management behaviors.

Adolescence signifies a time of profound transformations in the brain, when both social-emotional and cognitive faculties mature (Weir, Zakama, Rao, 2012). Development across these domains, including self-control and executive functions, is intertwined with the development of connections within the prefrontal cortex-limbic and synaptic pruning process. The mesostriatal and mesocorticolimbic systems, which are connected to the development of reward processing and reward-directed behavior, also undergo maturation during this time. The presence of depressive symptoms at this vulnerable stage may alter the trajectory of typical neurodevelopment (Luby et al., 2016; Beauchaine, 2015), rendering the brain especially susceptible to changes in cognitive functioning.

Neuroimaging and fMRI studies reveal structural and functional changes within the corticolimbic and corticostriatal systems in children and adolescents with depressive symptoms, which involve the hippocampus, amygdala, prefrontal lobes, and striatum (Weir, Zakama, Rao, 2012; Beauchaine, 2015; Kessler, Traue, & Wiswede, 2011). Depressive symptoms are associated with a reduction in hippocampal volume, a region involved in memory and emotional processing. Increased ratio of amygdala to hippocampal volume has been found in youth with

depressive symptoms. In youth with a history of familial depressive symptoms, left prefrontal cortex volume is correlated with severity of depressive symptoms. Finally, gray matter deficits in the caudate nucleus have been observed in adolescents with depressive symptoms, a structure that is responsible for information processing and inhibition (Weir, Zakama, Rao, 2012).

Disruption and reduction of the caudate can result in distractibility, inattention, and forgetfulness of daily activities, and hyperactivity in children (Schrimsher, Billingsley, Jackson, & Moore, 2002).

Depressive symptoms in children may lead to neural changes throughout development. Preschoolers exhibiting depressive symptoms at a young age have demonstrated a reduction in cortical gray matter volume and thinning across the cortex in middle childhood and early adolescence at almost twice the rate of their emotionally healthy peers. These structural changes may reflect early experience-based synaptic pruning, which is maladaptive given that the child's developing brain may be shaped by these early negative experiences (Luby et al., 2016). Given the developmental challenges already facing a child or adolescent with spina bifida, depressive symptoms may pose an additional threat to their neuropsychological functioning.

Limitations of the Current Literature

The current literature reveals the importance of self-management in children with a chronic health condition (Pai & Drotar, 2010). Proper self-management of a child or adolescent's chronic illness is related to higher confidence levels, better school attendance, greater opportunities for socialization with friends, increased family functioning, greater ability to navigate life independently within one's community, more engagement with employment, and improved health outcomes (Conn, Fisher, & Rhee, 2016; Sabaté, 2003; Sawin, Bellin, Roux,

Buran, & Brei, 2009; Suris, Michaud, & Viner, 2004; Eilander et al., 2015; Ridosh, Braun, Roux, Bellin & Sawin, 2011; Van Mechelen, Verhoef, Van Asbeck, & Post, 2008).

Yet, despite the importance of self-management in pediatric chronic conditions, little research exists on self-management in spina bifida. While Psihogios and colleagues (2013; 2015; 2016) have examined parent-child discrepancies in perceptions of medical autonomy and adherence, empirical evidence is lacking on the individual factors that impact these self-management behaviors in spina bifida. Neuropsychological deficits are particularly important to investigate as a predictor of self-management outcomes in youth with spina bifida, as difficulties with attention and executive dysfunction are frequently present in this population. While this relationship has been previously documented, it is unclear what factors may exacerbate cognitive deficits, thus contributing to greater difficulties with medical management. In addition, although research has documented an association between depressive symptoms and self-management, only one study to date has considered potential mood-related effects on self-management in spina bifida (Bellin et al., 2013). This study was limited to emerging adults (i.e., 18-25 years) and was underpowered to permit use of more sophisticated statistical analyses. Given the connection between depressive symptoms and cognitive dysfunction, examining these factors in a single model would elucidate how emotional and cognitive functioning impact the development of self-management in spina bifida.

Previous research makes a strong argument for attentional and executive dysfunction as a mechanism explaining depressive symptoms' effect on self-management. Indeed, this model has been partially explored in other chronic illnesses (Guo et al., 2013; Cameron et al., 2010). However, few studies have applied this model to a pediatric population, and no research to date

has examined this model in youth with spina bifida, despite the fact that they are at increased risk for depressive symptoms and attentional/executive functioning deficits (Modi et al., 2012; Holmbeck et al., 2003; Rose & Holmbeck, 2007). Furthermore, given the ambiguous causal relationship between depressive symptoms and cognitive deficits, it is possible that the relationship between these two factors is bidirectional. That is, depressive symptoms may be one potential mechanism through which cognitive dysfunction negatively impacts self-management. To date, no such studies have examined competing models of cognitive dysfunction and depressive symptoms in relation to pediatric chronic health condition outcomes. Understanding pathways from which individual differences in psychological and cognitive functioning influence health-related behaviors is essential to developing and refining clinical interventions for strengthening self-management capacities in youth with spina bifida.

A review of relevant literature regarding self-management in spina bifida reveals significant methodological concerns. Most past studies were underpowered due to small sample size, were unable to infer directionality due to the cross-sectional nature of the design, and were vulnerable to common method variance effects due to a single-informant or single-method approach. The current study will address these critical gaps in the literature and methodology.

The Current Study

The goals of this study are to enhance the understanding of self-management in youth with spina bifida by examining potential individual factors impacting these behaviors. Specifically, this study aims to clarify the relationship between depressed mood and self-management by investigating neuropsychological dysfunction as a potential mediator (see Figure 1).

The study adopts a longitudinal, multi-informant, multi-method approach to address some of the methodological limitations of past research. A unique aspect of this study is its use of depressive symptoms as an independent variable, as it is more commonly studied as an outcome in youth with spina bifida (Appleton et al., 1997; Oddson, Clancy, & McGrath, 2006; Friedman et al., 2004; Müller-Goddefroy et al., 2008). The first goal of this study is to determine whether depressive symptoms and higher order cognitive functions are related to self-management behaviors in youth with spina bifida. It is expected that greater depressive symptoms will result in less child responsibility for spina bifida management tasks and lower adherence rates for spina bifida tasks that are managed primarily by the child.

The second goal of this study is to understand *how* depressive symptoms and neuropsychological deficits impact self-management. It is expected that neuropsychological dysfunction will mediate the relationship between depressive symptoms and self-management. That is, for children who present with more depressive symptoms, these symptoms will exacerbate attentional and executive dysfunction, leaving them with fewer cognitive resources to allocate towards their complicated medical regimen. Due to the potentially bidirectional relationship between cognitive functioning and depressive symptoms, it is also expected that depressive symptoms will mediate the relationship between neuropsychological deficits and self-management. In this pathway, children with poorer self-regulatory capacities due to attention and executive functioning deficits will be at greater risk of developing depressive symptoms, which will lead to a decrease in motivation to manage their medical regimen independently.

The utilization of three time points will allow for the investigation of a more complex hypothesis (i.e., mediational model), which will help broaden the field's knowledge of how

mood impacts disease management. Additionally, the longitudinal nature of this study will allow for an examination of how responsibility for disease management in youth with spina bifida develops across late childhood and adolescence, which will further deepen the understanding of self-management trajectories in this population.

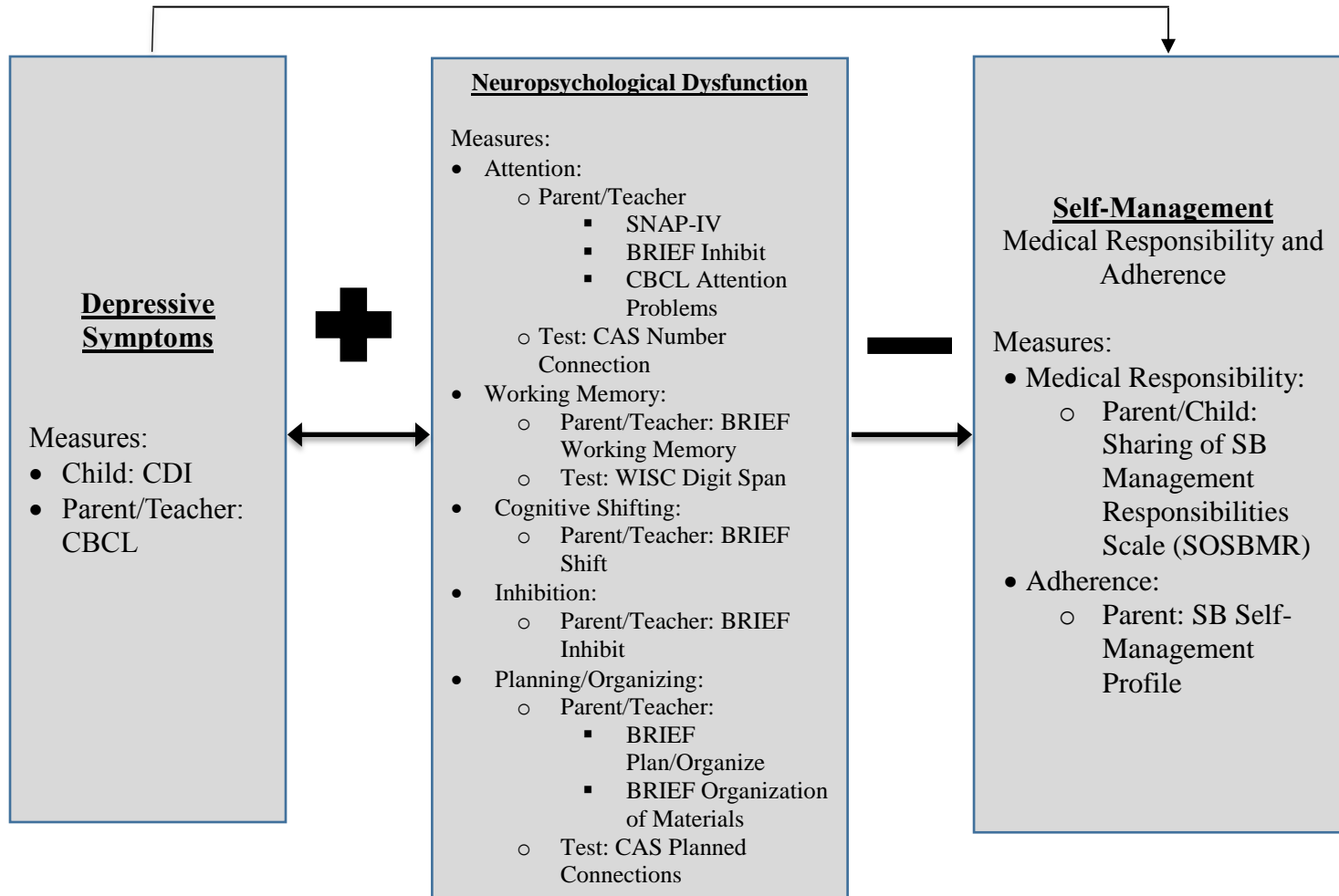
Study Hypotheses

The current study had two objectives. The first aim of the study was to examine the impact of depressive symptoms on self-management, as mediated by attention and executive dysfunction in youth with spina bifida (see Figure 1a). It was hypothesized that neuropsychological functioning would mediate the relationship between depression and self-management, such that greater depressive symptoms would predict worse neuropsychological functioning (**Hypothesis 1**), which in turn would predict poorer self-management (**Hypothesis 2**). Although greater depressive symptoms were also expected to be directly associated with poorer self-management outcomes, it was expected that the relation between depressive symptomology and self-management would be significantly reduced when controlling for attentional and executive dysfunction (**Hypothesis 3**).

To investigate an alternative direction of mediation, the second aim of the study was to examine the impact of attention and executive functioning on self-management, as mediated by depressive symptoms (see Figure 1b). It was hypothesized that depressive symptoms would mediate the relationship between neuropsychological functioning and self-management, such that worse neuropsychological deficits would predict greater depressive symptoms (**Hypothesis 4**), which would in turn predict poorer self-management (**Hypothesis 5**). Although poorer neuropsychological functioning was expected to be directly associated with worse self-

management outcomes, it was expected that the relation between neuropsychological functioning and self-management would be significantly reduced when controlling for depressive symptoms (**Hypothesis 6**). Figure 2 displays the specific measures used to assess each construct in the current study.

Figure 2. Mediational Model of Depression, Neuropsychological Functioning, and Self-Management



CHAPTER THREE

METHODS

Participants

Participants were recruited from an ongoing, larger longitudinal study examining family, neuropsychological, and psychological functioning among children and adolescents with spina bifida (e.g., Devine et al., 2012). The present study examined three waves of data that were collected every 2 years (ages 8-15 at Time 1), and focused on data regarding depressive symptoms, neuropsychological functioning, and disease self-management in youth with spina bifida.

Families of youth with spina bifida were recruited from four hospitals and a statewide spina bifida association in the Midwest. Families were sent recruitment letters and were also approached during regularly scheduled clinic visits. Interested families were screened by phone or in-person by a member of the research team, and were invited to participate if their child met the following criteria: (a) diagnosis of spina bifida (types included myelomeningocele, lipomeningocele, myelocystocele); (b) age 8–15 years at Time 1; (c) ability to speak and read English or Spanish; (d) involvement of at least one primary caregiver; and (e) residence within 300 miles of laboratory (to allow for home visits to collect data).

Two-hundred and forty-six families were approached during recruitment, of which 163 initially agreed to participate. After this initial recruitment, 21 families could not be contacted or later declined, and 2 families did not meet all of the inclusion criteria. The final sample of

participants included 140 families of children with spina bifida (53.6% female; 53.5% Caucasian; M age = 11.40). Children of families who declined participation did not differ from those who agreed to participate with respect to type of spina bifida (e.g., myelomeningocele vs. other), $\chi^2(1) = 0.0002, p > .05$, shunt status, $\chi^2(1) = 0.003, p > .05$, or occurrence of shunt infections $\chi^2(1) = 1.08, p > .05$.

Additionally, because self-management tasks necessitate a certain cognitive capacity, the present study excluded participants who functioned intellectually at two or more standard deviations below the population mean. This criterion was met if a child obtained an estimated intelligence quotient (IQ) score below 70 (American Psychiatric Association, 2013). At Time 1, 26 out of 140 (19%) individuals had an estimated IQ < 70 or did not complete the brief neuropsychological battery due to low comprehension, and as a result were not included in the study. The final sample used in analyses included 114 children and adolescents with spina bifida (56.1% female; $M_{age} = 10.96(2.43)$; 60.0% Caucasian). Data regarding child demographic characteristics is provided below in Table 1.

Out of the 114 participants that were included at Time 1, 92 (81%) participated at Time 2, and 84 (74%) participated at Time 3. Youth who did not participate at Time 2 or Time 3 ($n = 38$, 33%) did not significantly differ from youth who did with respect to gender, socioeconomic status, type of spina bifida, lesion level, shunt status, IQ, severity of depressive symptoms, degree of medical autonomy, attention, working memory, father-reported cognitive shifting, teacher-reporting cognitive shifting, inhibition, or planning and organizing abilities. However, youth who did not participate at Times 2 or 3 were significantly older at Time 1 [$M = 11.74$

compared to 10.61; $t(106) = -2.28, p = .03$] and had more problems with cognitive shifting per mother report [$M = 1.80$ compared to 1.62; $t(97) = -2.01, p = .05$].

Child medical information was gathered from their medical chart and maternal report via questionnaire. Of the 114 participants included in the current study, medical chart review at Time 1 indicated that 83.3% had a diagnosis of myelomeningocele, 8.8% lipomeningocele, and 7.8% other. The majority of children had spinal lesions in the lumbosacral or lumbar spinal regions (68.5%), while 21.3% had sacral lesions, and 10.2% had thoracic lesions. Most children (73.2%) had a shunt. Mothers reported that 81.7% of the children used braces to ambulate and 58.7% used a wheelchair.

Table 1. Child Demographic Information at Time 1

Characteristic	Child with Spina Bifida (N=114)
Age M (SD)	10.96 (2.43)
Gender:	
% Male	43.9%
% Female	56.1%
Ethnicity:	
% White	60.0%
% Hispanic	21.9%
% African American	13.3%
% Other	4.8%
Hollingshead SES, M (SD)	42.32 (14.99)

Procedure

The current study was approved by university and hospital Institutional Review Boards and utilized a multi-method, multi-informant longitudinal research design. Data were collected by trained undergraduate and graduate student research assistants during home visits that each lasted approximately three hours. Home visits at Time 1 consisted of two 3-hour home visits and only one 3-hour home visit at Time 2 and Time 3. Informed consent from parents and assent from youth were obtained prior to the start of the first visit. At least one bilingual research assistant was present with families who primarily spoke Spanish in the home. After obtaining consent from families, children completed questionnaires regarding psychological adjustment, executive functioning, and responsibility for spina bifida self-management tasks. Parents completed identical questionnaires separately pertaining to their child's medical and health history, psychological adjustment, executive and attentional functions, and spina bifida self-management behaviors. Demographic information was collected via a parent questionnaire. Questionnaires that were only available in English were adapted for Spanish speakers using forward and back translation by a translation team.

Children also participated in a brief neuropsychological battery assessing various domains of neuropsychological functioning (e.g., intelligence, attention, executive functioning, etc.). The neuropsychological assessments were administered by trained research assistants. The battery was conducted in English, but task instructions were clarified in Spanish if needed. Neuropsychological measures were scored by another trained research assistant after the home visit. Parents completed releases of information to allow for data collection from healthcare providers and teachers via mail, as well as obtainment of medical data from the medical chart.

The larger study involved youth, parent, teacher, healthcare provider, and peer questionnaires; youth, parent, and peer audiotaped interviews; youth neuropsychological testing; videotaped family interaction tasks of the child and his/her parent(s); and videotaped peer interaction tasks of the youth with a best friend. The current study used youth-, parent-, and teacher -reported questionnaire data and neuropsychological assessment data. Families received \$150, a t-shirt, and a pen as compensation for participation at each time point.

Measures

Demographics and Medical Information. Parents reported on youth and family demographic information through questionnaires at Time 1. Parents reported on child age, gender, race, and ethnicity. Parents also reported on their gender, ethnicity, education, employment, and income. The Hollingshead Index of socioeconomic status (SES) was computed to assess SES based on parents' education and occupation, with higher scores indicating higher SES (Hollingshead, 1975). To assess medical information, mothers completed the Medical History Questionnaire (MHQ; Holmbeck et al., 2003), and data were abstracted from hospital medical records. Specifically, information regarding the type of spina bifida (i.e., myelomeningocele, meningocele, or lipomeningocele), shunt status, lesion level (i.e., sacral, lumbar, or thoracic) and ambulation method (i.e., ankle-foot orthoses [AFOs], knee-ankle-foot orthoses [KAFOs] or hip-knee-ankle-foot orthoses [HKAGOs] wheelchair, or no assistance) was collected. Lesion level (sacral = 1, lumbar = 2, thoracic = 3) was used as a proxy indicator of illness severity, with higher scores indicating higher levels of severity (Hommeyer, Holmbeck, Wills, & Coers, 1999). Child age, socioeconomic status, and level were included as covariates in analyses.

Intelligence. Intellectual functioning was measured via child performance on two subtests of the *Wechsler Abbreviated Scale of Intelligence* (WASI; Wechsler, 1999). Intelligence at Time 1 was used as part of the inclusion criteria for this study, as intellectual ability may preclude children from being able to carry out certain tasks related to self-management. The Vocabulary subtest assesses verbal intellectual ability and consists of a 42-item task similar to the Vocabulary subtests of the Wechsler Intelligence Scale for Children (WISC-III) and the Wechsler Adult Intelligence Scale (WAIS-III), except that the WASI subtest includes low-end picture items. Participants are presented pictures in items 1-4 and are instructed to name them, while they are asked to define words that are presented orally and visually in items 5-42. The WASI Vocabulary subtest measures an individual's verbal concept formation and verbal knowledge, and is an acceptable measure of crystallized intelligence and general intelligence. The average internal consistency reliability coefficient for children ages 6-16 years old was .89 (Wechsler, 1999). The Matrix Reasoning subtest measures non-verbal intellectual ability, visual intelligence, and fluid intelligence. It is similar to the Matrix Reasoning subtest in the WAIS-II. The Matrix Reasoning subtest consists of 35 items; for each item, participants are presented with an incomplete matrix of shapes and must select one of five potential shape options to correctly complete the pattern. The average internal reliability coefficient for children ages 6-16 years old was .92 (Wechsler, 1999). A Full Scale IQ score was estimated using the scaled scores on the Vocabulary and Matrix Reasoning subtests.

Predictors

Depressive symptoms. Depressive symptoms were measured via child-, parent-, and teacher-report. Children completed the Child Depressive Inventory (CDI) at Time 1 and Time 2

(Kovacs, 1992). The CDI is a 27-item self-rated measure of depressive symptoms for children and adolescents. Children rate items that assess five factors of depressive symptoms (i.e., negative mood, interpersonal problems, ineffectiveness, anhedonia, and negative self-esteem) over the past two weeks. Each item consists of three choices, keyed 0, 1, or 2, with higher scores indicating increased symptomatic severity. While the CDI yields five subscales corresponding to the five factors of depressive symptoms, the total score was utilized for this study. This measure of depressive symptoms is well-validated for the general population and has also been used with samples of youth with spina bifida. The CDI demonstrated acceptable levels of internal consistency at both Time 1 and Time 2 ($\alpha = .82$; $\alpha = .78$).

Parents completed the Child Behavior Checklist (CBCL) and teachers completed the Teacher Report Form (TRF) at Time 1 and Time 2 (Achenbach, 1991; Achenbach & Rescorla, 2001). The CBCL and TRF are comprised of 118 items that assess behavioral and emotional problems over the past six and two months, respectively. Parents and teachers rate each item on a three-point Likert scale (0=not true, 1=somewhat or sometimes true, 2=very true or often true) as it pertains to the child in question. The CBCL and TRF yield T-scores for eight problem subscales (Anxious/Depressed, Withdrawn/Depressed, Somatic Complaints, Social Problems, Thought Problems, Attention Problems, Rule-Breaking Behavior, and Aggressive Behavior). T-scores above 65 are considered to be borderline or clinically significant. In a previous study of children with spina bifida and a matched comparison sample (Holmbeck, et al., 2003), 23.5% and 7.4% of the spina bifida sample had mean T-scores of 60 or above on the Internalizing and Externalizing scales, respectively. Percentages for the comparison sample were 7.4% and 7.4%, respectively.

For this study, a subscale of depressive symptoms were derived from 15 items included in the Anxious/Depressed and Withdrawn/Depressed subscales to form the CBCL-Depression Scale (CBCL-D; Clarke, Lewinsohn, Hops, & Seeley, 1992; see Table 2). Specifically, items 8, 14, 18, 24, 35, 52, 54, 76, 77, 91, 100, 102, 103, 111, and 112 will be used. This subscale was rationally derived to yield specific information on core depressive symptomology, without contamination from items related to unrelated affective symptoms (e.g., anxiety; Clarke, Lewinsohn, Hops, and Seeley, 1992). In this study, the CBCL-D demonstrated adequate levels of internal consistency across reporters and time points ($\alpha = .69-.84$). As this adapted scale has not been normed, raw mean total scores were calculated in lieu of T-scores.

Table 2. The Child Behavior Checklist Depression Scale Items

CBCL Item	Parent/Teacher Scale Item Number
Can't concentrate, can't pay attention	8
Cries a lot	14
Harms self or attempts suicide	18
Doesn't eat well	24
Feels worthless or inferior	35
Feels too guilty	52
Overtired	54
Sleeps less than most children	76
Sleeps more than most children	77
Talks about killing self	91
Trouble sleeping	100
Underactive, slow moving, lacks energy	102
Unhappy, sad, or depressed	103
Withdrawn, uninvolved with others	111
Worrying	112

Neuropsychological functions. Child attention and executive functions were assessed via performance-based measures, as well as parent- and teacher-report, at Time 1 and Time 2. This study aimed to evaluate different domains of higher order cognitive skills that could be disrupted by depressive symptoms and negatively impact self-management behaviors. In particular, the following areas of neuropsychological functioning will be examined: 1) attention, 2) working memory, 3) cognitive flexibility (i.e., cognitive shifting), 4) inhibition, and 5) planning and organizational skills. Depressive symptoms may cause varying levels of disturbance across the different neuropsychological domains, thus they will be evaluated separately and not as a global attentional/executive functioning construct. Multiple reporters, measures, or subscales tapped into each of the five domains. To maintain clinically relevant categories of neuropsychological functioning, a rational as opposed to an empirical or purely statistical approach was taken when clustering these measures into distinct constructs. Table 3 displays the measures included in each neuropsychological construct. The measures included in each domain of neuropsychological functioning are described below:

Table 3. Measures Included in Constructs of Neuropsychological Functioning

Construct/Measure	Source of Data		
	Parent Report	Teacher Report	Performance Based
Attention			
SNAP-IV	X	X	
CAS Number Detection Subtest			X
CBCL Attention Problems Subscale	X	X	
Working Memory			
WISC-IV Digit Span Subtest			X
BRIEF Working Memory Subscale	X	X	
Cognitive Flexibility			
BRIEF Shift Subscale	X	X	
Inhibition			
BRIEF Inhibit Subscale	X	X	
Planning/Organizing			
CAS Planned Connections Subtest			X
BRIEF Plan/Organize Subscale	X	X	
BRIEF Organization of Materials Subscale	X	X	

Attention. Attentional ability and control were measured via parent- and teacher-report, as well as child performance. *The Swanson, Nolan, and Pelham – Fourth Edition*. Parents and teachers completed the Swanson, Nolan, and Pelham – Fourth Edition (SNAP-IV; Swanson, 1992), a questionnaire which assesses ratings of ADHD symptoms based on DSM-IV diagnostic criteria. The measure consists of eighteen items and yields an inattention subscale (e.g., “Can’t pay attention,”) and a hyperactivity/impulsivity subscale (e.g., “Often fidgets with hands or feet or squirms in seat”), both of which are derived from nine items. Parents and teachers were asked to rate the degree to which a child endorses each item using a 0 to 3 Likert rating scale: Not at All = 0, Just A Little = 1, Quite A Bit = 2, and Very Much = 3. Higher scores indicate greater severity of symptoms within each subscale. Mean subscale scores were calculated for inattention

only. Previous research shows that mother-, father-, and teacher- report total item mean scores were sufficiently correlated on the inattentive ($r = .41$ to $.72$) subscale in families of children with spina bifida (O'Hara, 2012). The inattentive subscale demonstrated excellent internal consistency in this study ($\alpha = .92-95$).

Cognitive Assessment System (CAS) Number Detection Subtest. The CAS (Naglieri & Das, 1997) is an assessment battery of tests designed to measure non-verbal cognitive processing in children ages 5 to 17 years. The Number Detection (ND) subtest is a stimuli attention task which assesses selectivity, ability to shift attention, and resistance to distraction. Examinees were presented with a page of numbers and were required to locate and underline a particular stimulus (i.e., specific numbers) on a page containing several distractors (i.e., the same numbers in a different font). Each item within the subtest is scored for accuracy and timed to provide an estimate of task efficiency. Raw scores were converted into age scaled scores, with higher scores indicating greater levels of attentional ability. Internal reliability ($\alpha = .77$) for the ND subtest are high across age groups (Naglieri & Das, 1997).

CBCL Attention Problems. Parents and teachers completed the Attention Problems subscale of the CBCL and TRF, respectively (Achenbach & Rescorla, 2001). On the CBCL, this subscale includes eleven items assessing child problematic behaviors related to inattention (e.g., "Can't concentrate"), hyperactivity (e.g., "Can't sit still"), and impulsivity (e.g., "Acts without thinking"). The Attention Problems subscale of the TRF is comprised of twenty items that assess similar behaviors. However, extra items are included in the TRF to reflect classroom-specific behaviors (e.g., "Messy work"). T-scores from the Attention Problems subscale will be used in analyses, with higher scores indicating more severe attention problems. Previous research with

children with spina bifida (Wasserman, Stoner, Stern, & Holmbeck, 2016), demonstrated that 62% of the sample had clinically elevated T-scores of 65 or above on the Attention Problems subscales. This subscale demonstrated acceptable levels of internal consistency in this study ($\alpha = .73-.82$). For further description of the CBCL and TRF, see the “Depressive Symptoms” section above.

Working Memory. Working memory was measured via parent- and teacher-report, as well as child performance. *The Wechsler Intelligence Scale for Children-Fourth Edition (WISC-IV), Digit Span Subtest.* The WISC-IV (Wechsler, 2003) is a battery of assessments designed to measure the cognitive ability of children ages 6 to 16 years. The subtests yield index scores across several domains of cognitive functioning, including verbal comprehension, visual spatial processing, fluid reasoning, working memory, and processing speed. This study used the Digit Span subtest, which falls within the Working Memory scale. The Digit Span subtest is comprised of two tasks: Digit Span Forward (DSF) and Digit Span Backward. In Digit Span Forward, the child is instructed to listen to and repeats a sequence of numbers spoken aloud by the interviewer. In Digit Span Backward, the child listens to a sequence of numbers and repeats them in reverse order. Raw scores were converted into age scaled scores, with higher scores indicating greater working memory function. The Digit Span subtest has good internal consistency ($r = .87$) and test-retest reliability ($r = .83$; Williams, Weiss, & Rolfhus, 2003).

Behavior Rating Inventory of Executive Function (BRIEF) Working Memory Subscale. Parents and teachers completed the BRIEF (Gioia et al., 2000a, 2000b), a questionnaire that measures several domains of executive functions of children. It is composed of eight clinical subtests including Inhibit (i.e., the ability to resist or not act on an impulse; e.g., “Interrupts

others”), Shift (i.e., the ability to move freely from one situation, activity or aspect of a problem to another demand; e.g., “Becomes upset with new situations”), Emotional Control (i.e., the capacity to modulate emotional responses; e.g., “Overreacts to small problems”), Initiate (i.e., the capacity to begin a task or activity or independently generate ideas, responses, or problems solving strategies; e.g., “Does not take initiative”), Working Memory (i.e., the ability to hold information in mind for the purpose of completing a task; e.g., “Has trouble remembering things, even for a few minutes”), Plan/Organize (i.e., the ability to manage current and future-oriented task demands; e.g., “Has good ideas but cannot get them on paper”), Organization of Materials (i.e., orderliness of work, play, and storage spaces; e.g., “Keeps room messy”), and Monitor (i.e., work-checking habits; e.g., “Makes careless errors”). These subtests fall within two broad indices, Behavioral Regulation and Metacognition, which yield the overall Global Executive Composite Score. Mothers, fathers, and teachers completed the 86 items that comprise the BRIEF subtests. On each item, parents and teachers were instructed to circle whether their child has never, sometimes, or often demonstrated a particular behavior during the past six months. Higher scores on the BRIEF represent higher levels of executive dysfunction. Across clinical subscales, the BRIEF has high internal consistency ($\alpha = .80-.98$) for parent and teacher reports, strong test-retest reliability ($r = .81$), and moderate interrater agreement ($r = .32$). Parents and teachers completed the Working Memory subscale of the BRIEF (Gioia et al., 2000a, 2000b), which assesses ratings of a child’s behaviors related to working memory over the past six months. Items related to this subscale include, “Forgets what he/she was doing” and “Has trouble remembering things, even for a few minutes”. Higher scores indicate more reported problems

with working memory, or poorer working memory ability. In this study, the Working Memory subscale demonstrated good internal consistency ($\alpha = .86-.91$).

Cognitive Shifting. Parents and teachers completed the *Shift subscale* of the BRIEF (Gioia et al., 2000a, 2000b), which assesses ratings of a child's ability to make transitions, problem-solve flexibly, and adjust focus among different thoughts or activities as necessary. Items related to this subscale include, "Acts upset by a change in plans" and "Thinks too much about the same topic". This subscale reflects the ability to adapt to deviations from a usual, consistent routine, as well as the ability to think creatively or try new approaches to problem-solve. Higher scores indicate issues with mental rigidity. The Cognitive Shifting subscale demonstrated acceptable levels of internal consistency in this study ($\alpha = .74-.86$). For further description of the BRIEF, see the above "Working Memory" section.

Inhibition. Parents and teachers completed the *Inhibit subscale* of the BRIEF (Gioia et al., 2000a, 2000b), which assesses ratings of a child's ability to control impulses and stop engaging in non-goal oriented behavior. Higher scores indicate poorer inhibitory control. This subscale demonstrated good levels of internal consistency ($\alpha = .86-.92$). For further description of the BRIEF, see the above "Working Memory" section.

Planning and Organizational Skills. Planning and organizational ability were assessed via parent-report, teacher-report, and child performance. The *Planned Connections (PCn) subtest* of the Cognitive Assessment System (CAS) is one of three subtests addressing a child's ability to generate a plan of behavior, sufficiently apply the plan, and modify the plan as needed to achieve a certain goal (Naglieri & Das, 1997). This subtest contains eight items. Examinees are required to connect numbers in sequential order in the first six items, and must connect both numbers and

letters in sequential order in an alternating fashion in the last two items. The total amount of time in seconds taken to complete the item sequence correctly is recorded, with lower scores (i.e., less seconds) indicating greater efficiency. The PCn subtest has high internal consistency ($\alpha = .77$) and test-retest reliability ($r = .73$; Naglieri & Das, 1997). For further description of the CAS, see the CAS Number Detection subtest section above, under the “Attention” section.

BRIEF. Parents and teachers completed the *Plan/Organize* and the *Organization of Materials* subscales of the BRIEF (Gioia et al., 2000a, 2000b). The *Plan/Organize* subscale measures both the ability to determine the most effective steps needed to achieve current and future-oriented goals, and the ability to think about and present information in an orderly and efficient manner. Items related to this subscale include, “Has trouble carrying out the actions needed to reach goals” and “Gets caught up in details and misses the big picture”. The *Organization of Materials* subscale measures the child’s tendency to keep his or her work, play, and storage spaces neat and orderly. Higher scores indicate greater difficulties with organizing one’s belongings. The *Plan/Organize* subscale and *Organization of Materials* subscale demonstrated good ($\alpha = .88-.92$) and acceptable ($\alpha = .78-.88$) levels of internal consistency, respectively. For further description of the BRIEF, see the above “Attention” section.

Self-Management of Spina Bifida. Self-management behaviors were measured via parent- and child-report at Time 1, Time 2, and Time 3. *Sharing of Spina Bifida Management Responsibilities Scale.* Parents and youth completed the *Sharing of Spina Bifida Management Responsibilities Scale* (SOSBMR), which is an adaptation of the *Diabetes Family Responsibility Questionnaire* (Anderson, Auslander, Jung, Miller, & Santiago, 1990). The SOSBMR assesses division of spina bifida responsibilities within the family, and is comprised of 34 items that

describe relevant spina bifida and health-related tasks (e.g., remembering to catheterize regularly). Participants rated who was primarily responsible for each task (e.g., parent, child, equal, or not applicable). For each item, a score of “1” was assigned to tasks where the parent is primarily responsible, “2” was assigned to tasks that shared equally between the parent and child, and “3” pertained to tasks for which the child was primarily responsible. In addition to a total responsibility scale, the SOSBMR includes several subscales: health-care appointments, communication about spina bifida with others, medications, general needs and self-care, ambulation, skin care, catheterization, bowel management, exercise, and diet. To reduce number of analyses, total mean scores were calculated for total responsibility scale only. In line with previous research, this study was unable to compute internal consistency scores for the total scale score of this measure, as reliability software uses listwise deletion when computing alpha coefficients, and several items include a “not applicable” response (Psyhogios, Kolbuck, & Holmbeck, 2015).

Spina Bifida Self-Management Profile. Parents of participants completed the Spina Bifida Self-Management Profile (SBSMP; Wysocki & Gavin, 2006). The SBSMP is a 14-item, structured interview which measures adherence to several different domains of spina bifida medical care. In this study, the interview was administered to mothers as a questionnaire rather than an interview. Specific areas of spina bifida medical care that are assessed include appointment keeping, bowel control program, skin care, exercise, medications, clean intermittent catheterization, and treatment of urinary tract infections. Given the heterogeneous needs of individuals with spina bifida, parents could respond “not applicable” for certain items which were not part of their child’s prescribed medical regimen. As reliability analyses rely on listwise

deletion, and a low number of parents completed every item, scale reliability could not be computed for this sample. Higher scores indicated higher levels of adherence within each medical domain. While this measurement alone does not assess self-management behaviors (e.g., child could be highly adherent to catheterization recommendations if parent is completing the task), it will be utilized after the SOSBMR has identified tasks for which the child is predominately responsible, thus allowing for the effective evaluation of self-management competencies. Thus, adherence levels will only be included in analyses for participants who obtain a mean total scale score above or equal to 2.1 on the SOSBMR (i.e., “child responsible”; see above for detailed description of the SOSBMR). For participants whom demonstrate a mean total scale score below 2.1 on the SOSBMR (i.e., “parent responsible”), it will be assumed that the child does not take responsibility for completing their spina bifida tasks the majority of the time. As a result, adherence levels using the SBSMP would not be included in analyses, as the SBSMP would then be measuring parent- or family-management of spina bifida care, as opposed to child-management.

Statistical Treatment

Preliminary Analyses

Prior to hypothesis testing, the psychometric properties (e.g., alphas) of all measures were evaluated. This included determining whether variables contained outliers or were skewed. Descriptive statistics were computed for all outcome measures to determine basic distributional properties. Different sources of data were used when possible to reduce the introduction of common method variance into the analyses. To reduce the number of analyses, data transformation techniques were used when appropriate.

Pearson correlation coefficients were used to calculate associations between measures assessed by two informants (e.g., mother-report, father-report) or methodologies. A criterion of $r \geq .40$ was used to determine which measures could be collapsed across reporters (Holmbeck et al., 2002). For constructs with three or more informants or methodologies (e.g., mother-report, father-report, teacher-report, child performance), total scale scores were treated as separate items in a single, global scale; thus, internal consistencies for the composite scales could be calculated using alpha coefficients. A criterion of $\alpha \geq .60$ was employed to determine which construct-specific measures could be aggregated into a composite score. Measures or subscales that were not able to be combined into a composite score were treated separately in the analyses.

Primary Analyses

All analyses included spina bifida severity, child age, and SES as covariates, as all three of these may contribute to depressive symptomology, attentional or executive functions, and/or spina bifida self-management behaviors. Furthermore, all longitudinal analyses included target variables at previous waves of data collection as covariates. Researchers of neurodevelopmental disorders argue that controlling for differences in IQ when examining specific neuropsychological deficits as outcomes in individuals with such disorders is “methodologically tenuous,” as overall cognitive deficits are intrinsic feature to these disorders (Dennis et al., 2009). Further, there are concerns about statistical overcorrection. Therefore, IQ was not included as a covariate in this study (Dennis et al., 2009).

Analytic Plan for Objective 1. Preacher and Hayes’ (2008) bootstrapping methods were employed to determine the impact of youth depressive symptoms at Time 1 on spina bifida self-management behaviors at Time 3, as mediated by neuropsychological functioning (i.e., attention,

working memory, cognitive flexibility, inhibition, and planning/organizing ability) at Time 2. Bootstrapping has been validated in the literature and is preferred over other methods, such as the Sobel Test (Sobel, 1982), as bootstrapping is less conservative and reduces the possibility of Type II errors (Preacher & Hayes, 2008). This procedure produces an empirical approximation of the product of the estimated coefficients' sampling distribution constituting the direct path and percentile-based bootstrap confidence intervals (confidence intervals and bootstrap measures of standard errors using 5000 resamples, with replacement, from the dataset (Preacher & Hayes, 2008). When zero is not between the upper and lower bounds of the confidence interval, it can be claimed, with 95% confidence, that the indirect effect is not zero, indicating a significant indirect effect.

Analytic Plan for Objective 2. To examine the alternative direction of mediation, Preacher and Hayes' (2008) bootstrapping methods were utilized to examine the impact of neuropsychological deficits at Time 1 on spina bifida self-management at Time 3, as mediated by depressive symptoms at Time 2. See Objective 1 for further explanation of this procedure.

For mediation models analyzed using percentile bootstrapping methods, assuming a power of .80, and an alpha of .05, a sample size of 36 is required to detect large effect sizes, a sample size of 78 is required to detect medium effect sizes, and a sample of 558 is required to detect small effect sizes (Fritz & MacKinnon, 2007). For the meditational analyses using child responsibility for spina bifida tasks as the outcome, the current study had enough power to detect medium or large effect sizes. The inclusion of medical adherence as an outcome was dependent upon the sample size of the subset of participants who were determined to be mostly responsible for their medical care (i.e., SOSBMR total responsibility mean score ≥ 2.1). The current study

did not have enough power to detect large effect sizes in models using medical adherence as the outcome ($n's < 36$). Thus, medical adherence as an outcome was dropped from the analyses.

Exploratory Analyses. Exploratory analyses were also conducted to examine the impact of depressive symptoms on self-management, as mediated by neuropsychological functioning, cross-sectionally. These mediation models only included medical responsibility as an outcome.

CHAPTER FOUR

RESULTS

Preliminary Analyses

Results from the preliminary analyses are displayed in Table 4. All variables were examined for outliers, but none were identified. In addition, all variables were tested for skewness. As recommended by Tabachnick & Fidell (2013), a conservative approach was utilized and variables were considered skewed and were transformed if skewness values were greater than or equal to 1.0. Results indicated that 14 variables were positively skewed. All positively skewed variables were transformed using the square root transformation. After the initial transformation, father-report of child depressive symptoms on the CBCL at T1 (skewness value = 1.22), mother-report of child depressive symptoms on the CBCL at T2 (skewness value = 1.18), father-report of child depressive symptoms on the CBCL at T2 (skewness value = 1.36), teacher-report of child depressive symptoms on the TRF at T2 (skewness value = 1.16), and father-report of the BRIEF Inhibition subscale at T2 (skewness value = 1.00) continued to be skewed. Therefore, log transformations were used for these variables.

To reduce the number of analyses and provide more stable measures of the participants' functioning, preliminary analyses also included an examination of the associations among multiple reporters and measures of variables within each construct. Variables were aggregated across three or more reporters and methodologies if they demonstrated adequate internal consistency ($\alpha > 0.60$). If the alpha coefficient for a particular cluster of three or more variables

was too low to meet the aggregation criterion, bivariate Pearson correlation coefficients were used to determine if variables could be aggregated across two reporters ($r \geq .40$). When aggregated with questionnaire measures, performance-based measures of neuropsychological functioning were reverse-scored so that higher scores in all cognitive constructs reflected greater deficits, or poorer functioning, in those areas.

Results indicated that global, composite variables could be created for child attention, working memory, and planning/organizing abilities at Times 1 and 2, overall child responsibility for medical care at Times 1, 2, and 3, and medical adherence at Times 1 and 3. Mother- and father-report could be aggregated to form a parent-report composite for child depressive symptoms at Times 1 and 2, child cognitive shifting abilities at Time 2, and child inhibition at Times 1 and 2. The remaining measures (self- and teacher-reported child depressive symptoms at Times 1 and 2, mother-, father-, and teacher-reported cognitive shifting abilities at Time 1, teacher-reported shifting abilities at Time 2, teacher-reported inhibition at Times 1 and 2, and mother- and father-reported medical adherence at Time 2) could not be aggregated, and were thus examined separately in subsequent analyses. Table 5 displays correlations among child depressive symptoms, neuropsychological functioning variables, self-management, and covariates at Time 1. Table 6 displays correlations among Time 1 child depressive symptoms and covariates, Time 2 neuropsychological functioning variables, and Time 3 medical self-management.

Table 4. Descriptions of Variables Transformed or Aggregated in Preliminary Analyses

Construct	Time Point	Method of Assessment	Skewness Values of Variables Needing Transformation	Variables which Met Aggregation Criteria	Variables Included in Analyses
Child Depressive Symptoms	T1	T1 CDI (C) T1 CBCL-Dep (M) T1 CBCL-Dep (F) T1 TRF-Dep (T)	T1 CDI (SK = 1.13) T1 CBCL-Dep (M; SK = 1.56) T1 CBCL-Dep (F; SK = 1.71)	T1 CBCL-Dep (M), T1 CBCL-Dep (F); ($r = .46, p < .01$)	T1 CDI T1 CBCL-P T1 TRF-T
	T2	T2 CDI (C) T2 CBCL-Dep (M) T2 CBCL-Dep (F) T2 TRF-Dep (T)	T2 CBCL-Dep (M; SK = 1.37) T2 TRF-Dep (SK = 1.97)	T2 CBCL-Dep (M), T2 CBCL-Dep (F); ($r = .42, p < .01$)	T2 CDI T2 CBCL-P T2 TRF-T
Attention	T1	T1 CBCL-Attn (M) T1 CBCL-Attn (F) T1 TRF-Attn (T) T1 SNAP (M) T1 SNAP (F) T1 SNAP (T) T1 CAS-ND	T1 CBCL-Attn (F; SK = 1.35) T1 TRF-Attn (SK = 1.00)	All included variables ($\alpha = .82$)	T1 Attention
	T2	T2 CBCL-Attn (M) T2 CBCL-Attn (F) T2 TRF-Attn (T) T2 SNAP (M) T2 SNAP (F) T2 SNAP (T) T2 CAS-ND	T2 CBCL-Attn (F; SK = 1.53) T2 CBCL-Attn (M; SK = 1.35) T2 TRF-Attn (SK = 1.24)	All included variables ($\alpha = .86$)	T2 Attention
Working Memory	T1	T1 BRIEF-WM (M) T1 BRIEF-WM (F) T1 BRIEF-WM (T) T1 WISC-DS	N/A	All included variables ($\alpha = .63$)	T1 Working Memory
	T2	T2 BRIEF-WM (M) T2 BRIEF-WM (F) T2 BRIEF-WM (T) T2 WISC-DS	T2 BRIEF-WM (T; SK = 1.06)	All included variables ($\alpha = .66$)	T2 Working Memory
Cognitive Shifting	T1	T1 BRIEF-Sh (M) T1 BRIEF-Sh (F) T1 BRIEF-Sh (T)	N/A	None	T1 Shift-M T1 Shift-F T1 Shift-T
	T2	T2 BRIEF-Sh (M) T2 BRIEF-Sh (F) T2 BRIEF-Sh (T)	N/A	T2 BRIEF-Sh (M), T2 BRIEF-Sh (F); ($r = .66, p < .01$)	T2 Shift-P T2 Shift-T

Inhibition	T1	T1 BRIEF-In (M) T1 BRIEF-In (F) T1 BRIEF-In (T)	T1 BRIEF-In (F; SK = 1.00)	T1 BRIEF-In (M), T1 BRIEF-In (F); ($r = .50; p < .01$)	T1 Inhibition-P T1 Inhibition-T
	T2	T2 BRIEF-In (M) T2 BRIEF-In (F) T2 BRIEF-In (T)	T2 BRIEF-In (M; SK = 1.22) T2 BRIEF-In (F; SK = 1.42)	T2 BRIEF-In (M), T2 BRIEF-In (F); ($r = .60; p < .01$)	T2 Inhibition-P T2 Inhibition-T
Planning/Organizing	T1	T1 BRIEF-PI (M) T1 BRIEF-PI (F) T1 BRIEF-PI (T) T1 BRIEF-Or (M) T1 BRIEF-Or (F) T1 BRIEF-Or (T) T1 CAS-PC	N/A	All included variables ($\alpha = .72$)	T1 Plan/Organize
	T2	T2 BRIEF-PI (M) T2 BRIEF-PI (F) T2 BRIEF-PI (T) T2 BRIEF-Or (M) T2 BRIEF-Or (F) T2 BRIEF-Or (T) T2 CAS-PC	N/A	All included variables ($\alpha = .78$)	T2 Plan/Organize
Child Medical Responsibility	T1	T1 SBMR (C) T1 SBMR (M) T1 SBMR (F)	N/A	All included variables ($\alpha = .88$)	T1 Medical Responsibility
	T2	T2 SBMR (C) T2 SBMR (M) T2 SBMR (F)	N/A	All included variables ($\alpha = .87$)	T2 Medical Responsibility
	T3	T3 SBMR (C) T3 SBMR (M) T3 SBMR (F)	N/A	All included variables ($\alpha = .83$)	T3 Medical Responsibility
Medical Adherence	T1	T1 SBSMP (M) T1 SBSMP (F)	T1 SBSMP-F (SK = -1.23)	All included variables ($r = .62; p = .01$)	T1 Adherence
	T2	T2 SBSMP (M) T2 SBSMP (F)	N/A	None	T2 Adherence-M T2 Adherence-F
	T3	T3 SBSMP (M) T3 SBSMP (F)	T3 SBSMP-M (SK = -1.13)	All included variables ($r = .53; p = .01$)	T3 Adherence

Note. CDI = Children's Depression Inventory; CBCL-Dep = Child Behavior Checklist Depression Subscale; TRF – Teacher Report Form; CBCL-Attn = Child Behavior Checklist Attention Problems Subscale; SNAP - Swanson, Nolan, and Pelham – Fourth Edition; CAS-ND – Cognitive Assessment System – Number Detection subtest; BRIEF-WM – Behavior Rating Inventory of Executive Function Working Memory Subscale; WISC-DS– Wechsler Intelligence Scale for Children – Fourth Edition – Digit Span subtest; BRIEF-Sh – Behavior Rating Inventory of Executive Function Cognitive Shifting Subscale; BRIEF-In – Behavior Rating Inventory of Executive Function Inhibition Subscale; BRIEF-PI – Behavior Rating Inventory of Executive Function Plan/Organize Subscale; BRIEF-Or – Behavior Rating Inventory of Executive Function Organization of Materials Subscale; CAS-PC – Cognitive Assessment System – Planned Connections subtest; SBMR – Sharing of Spina Bifida Medical Responsibilities Scale; SBSMP – Spina Bifida Self-Management Profile; C – child-report; M – mother-report; F – father-report; T – teacher-report; P – parent-report.

Table 5. Correlations among Depressive Symptoms, Neuropsychological Variables, Self-Management Variables, and Covariates at Time 1

Variable	1.	2.	3.	4.	5.	6.	7.	8.	9.	10.	11.	12.	13.	14.	15.	16.
Depressive Symptoms																
1. CDI	–	.12	.14	.33**	.33**	.24*	.23*	.09	.22*	.20	.23*	.02	-.13	-.03	-.18	-.16
2. CBCL-P		–	-.03	.30**	.24*	.43**	.48**	.06	.48**	-.06	.36**	-.01	-.16	.03	.15	-.02
3. TRF			–	.63**	.59**	.04	.14	.69**	.13	.64**	.40**	-.24*	-.15	-.16	-.09	.01
Cognitive Functioning^a																
4. Attention				–	.84**	.33**	.37**	.69**	.30**	.60**	.78**	-.21*	-.21*	-.09	-.07	-.07
5. Working Memory					–	.40**	.44**	.69**	.41**	.62**	.76**	.19*	.26**	-.12	-.08	-.09
6. Shifting-M						–	.32**	.11	.47**	.11	.42**	-.26*	-.11	-.09	.15	-.11
7. Shifting-F							–	.14	.58**	.16	.46**	-.15	-.07	-.12	.09	-.16
8. Shifting-T								–	.18	.77**	.68**	-.15	-.28**	-.13	-.16	-.07
9. Inhibition-P									–	.19	.39**	-.16	-.34**	-.24*	-.08	-.26*
10. Inhibition-T										–	.59**	-.24*	-.14	-.27**	-.14	-.01
11. Plan/Organizing											–	-.09	-.31**	-.06	.04	-.07
Self-Management																
12. Med. Responsibility												–	-.17	.53**	.06	-.13
13. Adherence													–	-.08	-.03	.33**
Covariates																
14. Age ^b														–	.05	.01
15. SES ^b															–	.03
16. Lesion Level ^b																–

Note. CDI = Children's Depression Inventory; CBCL = Child Behavior Checklist; TRF – Teacher Report Form; M – mother-report; F – father-report; T – teacher-report; P – parent-report SES = socioeconomic status measured by Hollingshead Four Factor Index.^aAll cognitive variables were scored such that higher scores represent greater neuropsychological deficits attention, working memory, cognitive shifting, inhibition, and planning/organizing abilities; ^bThese variables are covariates. * $p < .05$, ** $p < .01$.

Table 6. Correlations among Time 1 Depressive Symptoms, Time 2 Neuropsychological Variables, Time 3 Self-Management Variables, and Covariates

Variable	1.	2.	3.	4.	5.	6.	7.	8.	9.	10.	11.	12.	13.	14.	15.
T1 Depressive Symptoms															
1. CDI	–	.12	.14	.17	.31**	.33**	.16	.30**	.23	.27*	.15	-.22	-.03	-.18	-.16
2. CBCL-P		–	-.03	.30**	.12	.21	.15	.26*	.07	.23*	-.09	-.24	.03	.15	-.02
3. TRF			–	.51**	.50**	.18	.63**	.30**	.47**	.42**	-.40**	-.23	-.16	-.09	.01
T2 Cognitive Functioning^a															
4. Attention				–	.80**	.49**	.74**	.43**	.67**	.82**	-.41**	-.27*	-.17	.08	.03
5. Working Memory					–		.68**	.51**	.64**	.79**	-.38**	-.23	-.28**	-.11	-.13
6. Shifting-P						–	.30*	.56**	.23	.65**	-.21	-.18	-.18	.03	-.05
7. Shifting-T							–	.31*	.82**	.73**	-.38**	-.21	-.16	-.01	.02
8. Inhibition-P								–	.33**	.55**	-.21	-.25	-.40**	-.17	-.30*
9. Inhibition-T									–	.71**	-.21	-.27	-.23	-.08	-.04
10. Plan/Organizing										–	-.24*	-.32*	-.28**	.02	-.09
T3 Self-Management															
11. Med. Responsibility											–	.05	.54**	-.03	-.15
12. Adherence												–	.08	-.03	.27*
Covariates															
13. Age ^b													–	.05	.01
14. SES ^b														–	.03
15. Lesion Level															–

Note. CDI = Children's Depression Inventory; CBCL = Child Behavior Checklist; TRF = Teacher Report Form; P = parent-report; T = teacher-report. SES = socioeconomic status measured by Hollingshead Four Factor Index. ^aAll cognitive variables were scored such that higher scores represent greater neuropsychological deficits attention, working memory, cognitive shifting, inhibition, and planning/organizing abilities; ^bThese variables are covariates. * $p < .05$, ** $p < .01$.

Hypothesis Testing

Mediation analyses were conducted to examine 1) if depressive symptoms indirectly influenced medical responsibility via neuropsychological dysfunction, and 2) if neuropsychological functioning directly impacted medical responsibility via depressive symptoms. For all analyses, SES, age, and lesion level at Time 1 were entered as covariates. For longitudinal analyses, Time 1 mediators (neuropsychological factors or depressive symptoms) and Time 2 medical responsibility scores were also entered as covariates. Age was a consistent, positive predictor of child medical responsibility in cross-sectional analyses examining child depressive symptoms as the independent variable and neuropsychological factors as mediators (p 's > .05), such that older age at Time 1 predicted more child responsibility for medical care at Time 1. However, age did not consistently predict medical responsibility in longitudinal analyses. Higher lesion level negatively predicted child medical responsibility in longitudinal analyses (p 's > .05), but not in cross-sectional analyses.

Objective 1

The first objective of this study was to examine if neuropsychological functioning mediated the impact of child depressive symptoms on self-management in youth with spina bifida longitudinally. It was hypothesized that more severe depressive symptoms at Time 1 would predict greater deficits in neuropsychological factors two years later at Time 2, which would in turn predict lower levels of self-management four years later at Time 3. To maximize sample size and investigate differential effects of depression symptoms on cognition, each neuropsychological factor (i.e., attention, working memory, cognitive shifting, inhibition, and planning/organizing) was tested separately as a mediator. Each model was tested with self-

parent-, and teacher-report of child depressive symptoms as separate independent variables at Time 1, for a total of fifteen models. Significant, main effects are presented in Figures 3a-3c. Hypotheses were partially supported, in that results indicated no significant mediation effects, but demonstrated that several of the individual pathways in the model were significant.

Hypothesis 1. Greater parent- and teacher-reported depressive symptoms at Time 1 predicted more deficits respectively in parent-reported inhibition ($b = 1.29, SE = .52, t = 2.47, p = .02$); and teacher-reported inhibition at Time 2 ($b = 1.82, SE = .79, t = 2.32, p = .03$). Greater teacher-reported depressive symptoms also predicted more deficits in teacher-reported cognitive shifting at Time 2 ($b = 1.10, SE = .37, t = 2.98, p = .01$).

Hypothesis 2. In the model using self-reported depressive symptoms as the independent variable, greater dysfunction in working memory ($b = -0.12, SE = .05, t = -2.32, p = .02$) predicted less child medical responsibility at Time 3. Additionally, teacher-reported inhibition at Time 2 predicted less child medical responsibility at Time 3, and this relationship was significant regardless of which reporter of depressive symptoms was included in the model (self-reported depressive symptoms: $b = -0.10, SE = .03, t = -3.21, p < .01$; parent-reported depressive symptoms: $b = -.09, SE = .03, t = -2.68, p = .01$; teacher-reported depressive symptoms: $b = -.09, SE = .03, t = -2.71, p = .01$).

Hypothesis 3. There were no significant mediating effects (p 's $> .05$). Contrary to hypotheses, there was a significant direct, positive effect of child depressive symptoms at Time 1 on child medical responsibility at Time 3, such that greater parent-reported depressive symptoms at Time 1 predicted more child responsibility for healthcare at Time 3 ($b = .27, SE = .12, t = 2.20, p = .03$). This effect was only significant in the model examining attention as a mediator. The

lack of significant bivariate correlation between these variables indicates statistical suppression; as a result, this finding will be regarded as a statistical artifact and will be interpreted with caution (Pandey & Elliott, 2010).

Figures 3a-3c. Longitudinal Mediation Model of Child Depressive Symptoms, Neuropsychological Functioning, and Self-Management¹.

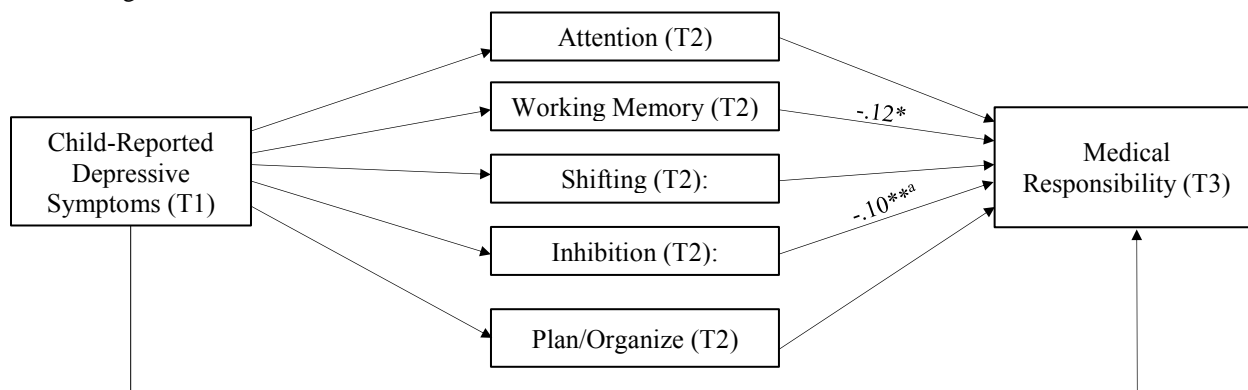
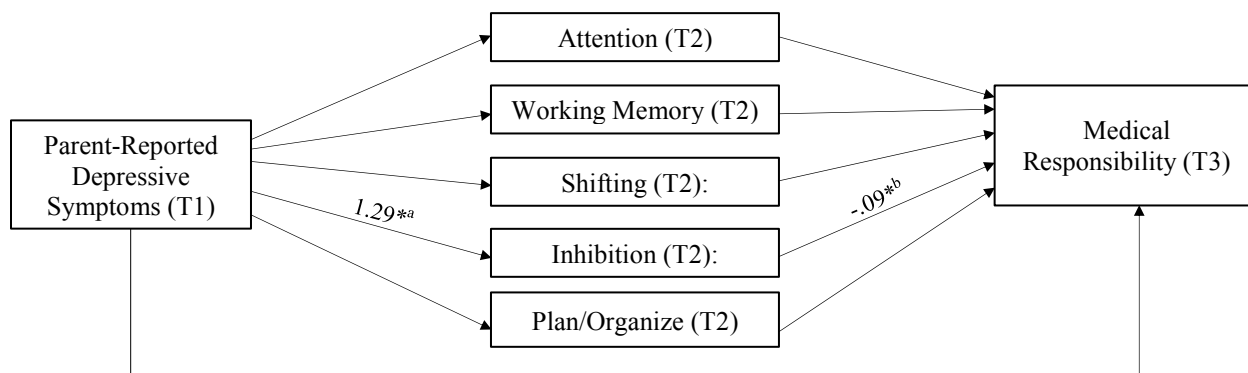


Figure 3a. Notes. ^aTeacher-reported child inhibition. * $p < .05$; ** $p < .01$.



Direct Effect^c: $b = .27$, $SE = .12$, $t = 2.20$, $p = .03$

Figure 3b. Notes. ^aParent-reported child inhibition; ^bTeacher-reported child inhibition; ^cDirect effect of parent-reported depressive symptoms on medical responsibility in model controlling for attention as a mediator. * $p < .05$; ** $p < .01$.

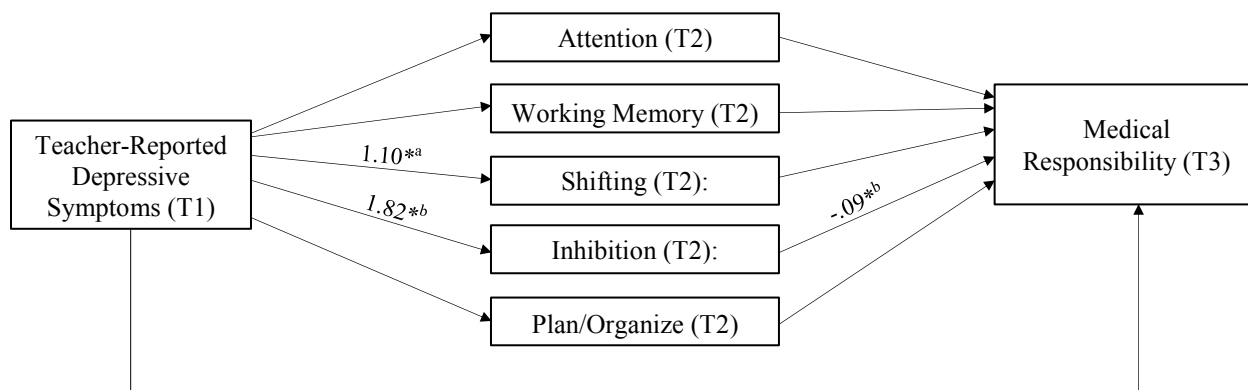


Figure 3c. Notes. ^aTeacher-reported cognitive shifting; ^bTeacher-reported inhibition. * $p < .05$; ** $p < .01$.

¹For Figures 3a-3c, analyses were tested separately for each of the five mediators and three independent variables. In all models, attention, working memory, and planning/organizing represent global, composite factors.

Objective 2

To test the alternative direction of causality (cognitive dysfunction predicting depressive symptoms), the second objective was to examine if child depressive symptoms mediated the impact of neuropsychological functioning on self-management in youth with spina bifida. This alternative pathway was also examined longitudinally. It was hypothesized that worse deficits in neuropsychological functioning at Time 1 would predict greater depressive symptoms at Time 2, which would predict less child responsibility for medical care at Time 3. Each neuropsychological factor as an independent variable was examined separately, and each of the three reports (self-, parent-, and teacher-report) of child depressive symptoms as the mediator were examined separately, for a total of twenty-four models (additional models were utilized for the separate reports of cognitive shifting and inhibition at Time 1 that were unable to be aggregated). Hypotheses were partially supported. Figures 4a-4h display significant, main effects.

Hypothesis 4. Greater deficits in attention ($b = .18$, $SE = .06$, $t = 2.83$, $p = .01$), working memory ($b = .13$, $SE = .06$, $t = 2.23$, $p = .03$), and planning and organizing abilities ($b = .20$, $SE = .07$, $t = 2.73$, $p = .01$) at Time 1 predicted more severe teacher-reported depressive symptoms at Time 2.

Hypothesis 5. Greater teacher-reported depressive symptoms at Time 2 predicted less child responsibility for medical care at Time 3. This relationship remained significant regardless of which neuropsychological factor was included in the model (attention: $b = -.22$, $SE = .11$, $t = -2.02$, $p = .05$; working memory: $b = -.23$, $SE = .10$, $t = -2.20$, $p = .03$; mother-reported cognitive shifting: $b = -.28$, $SE = .09$, $t = -3.10$, $p < .01$; father-reported cognitive shifting: $b = -.33$, $SE =$

.12, $t = -2.65$, $p = .01$; teacher-reported cognitive shifting: $b = -.23$, $SE = .10$, $t = -2.30$, $p = .03$; attention: $b = -.22$, $SE = .11$, $t = -2.02$, $p = .05$; parent-reported inhibition: $b = -.29$, $SE = .09$, $t = -3.06$, $p < .01$; teacher-reported inhibition: $b = -.24$, $SE = .10$, $t = -2.36$, $p = .02$; planning/organizing: $b = -.28$, $SE = .11$, $t = -2.60$, $p = .01$).

Hypothesis 6. When parent-reported depressive symptoms were included in the model, there was a significant, negative direct effect of attention ($b = -.09$, $SE = .03$, $t = -2.57$, $p = .01$) and teacher-reported cognitive shifting ($b = -.14$, $SE = .07$, $t = -2.03$, $p = .05$) at Time 1 on child medical responsibility at Time 3, such that greater attentional and shifting deficits predicted less child responsibility for medical care four years later. Contrary to hypotheses, there was a significant, positive direct effect of parent-reported inhibition at Time 1 on child medical responsibility at Time 3, such that greater inhibitory deficits predicted more child responsibility for medical care four years later ($b = .10$, $SE = .04$, $t = 2.55$, $p = .01$). This effect was found only when teacher-reported depressive symptoms were included as a mediator in the model.

Consistent with hypotheses, teacher-reported depressive symptoms at Time 2 significantly mediated the relationship between attention at Time 1 and child responsibility for medical care at Time 3 (estimated indirect effect = $-.04$, $SE = .02$, 95% LLCI to ULCI = $-.09$ to $-.01$). Teacher-reported depressive symptoms at Time 2 also significantly mediated the relationship between working memory at Time 1 and child medical responsibility at Time 3 (estimated indirect effect = $-.03$, $SE = .02$, 95% LLCI to ULCI = $-.09$ to $-.01$). Additionally, the indirect effect of planning and organizing abilities on self-management through teacher-reported depressive symptoms was significant (estimated indirect effect = $-.05$, $SE = .03$, 95% LLCI to ULCI = $-.14$ to $-.01$). However, because the magnitude of the direct effect of planning and

organizing skills when adjusting for depressive symptoms was greater than the total effect, results indicated statistical suppression as opposed to mediation (MacKinnon, Krull & Lockwood, 2000). Therefore, this finding will be regarded as a statistical artifact that will not be interpreted as mediation.

Figures 4a-4h. Alternative Direction Mediation Model of Child Neuropsychological Functioning, Depressive Symptoms, and Self-Management².

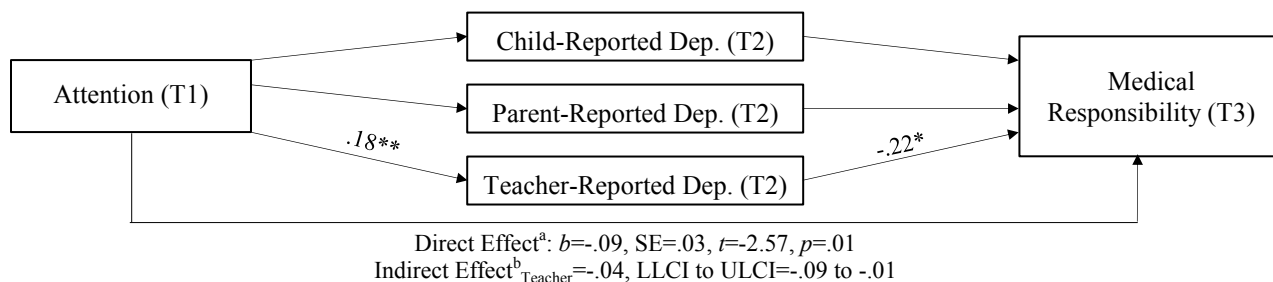


Figure 4a. Notes. ^aDirect effect of attention on medical responsibility in model controlling for parent-reported depressive symptoms as a mediator; ^bIndirect effect of attention on medical responsibility through teacher-reported depressive symptoms. Neither total effect nor direct effect was significant for the model with a significant indirect effect. * $p < .05$; ** $p < .01$.

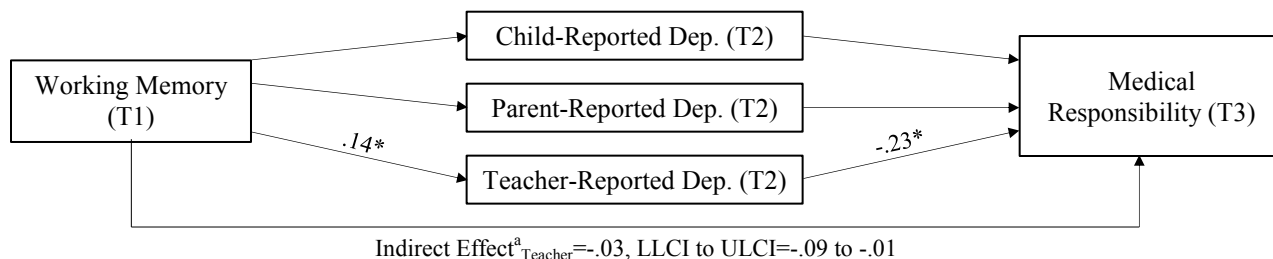


Figure 4b. Notes. ^aIndirect effect of working memory on medical responsibility through teacher-reported depressive symptoms. Neither total effect nor direct effect was significant for the model with a significant indirect effect. * $p < .05$.

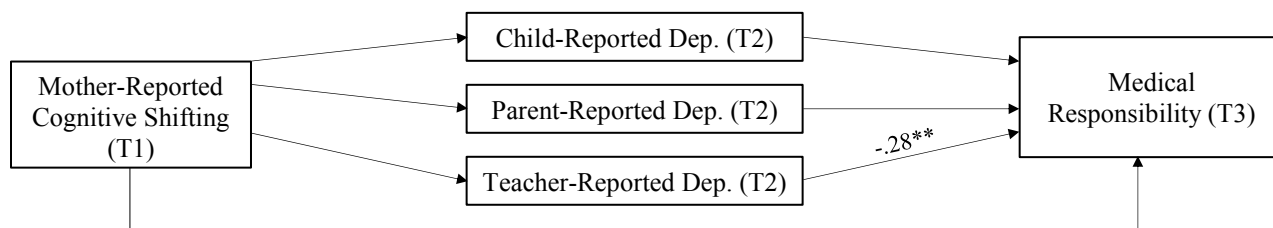


Figure 4c. Notes. ** $p < .01$.

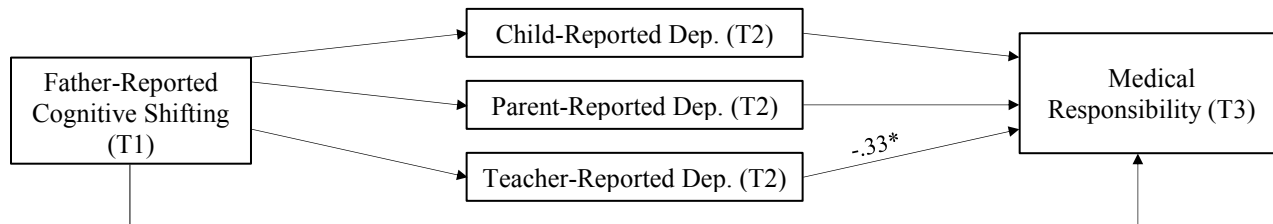


Figure 4d. Notes. * $p < .05$.

²For Figures 4a-4h, analyses were tested separately for each of the three mediators and eight independent variables. In all models, attention, working memory, and planning/organizing represent global, composite factors.

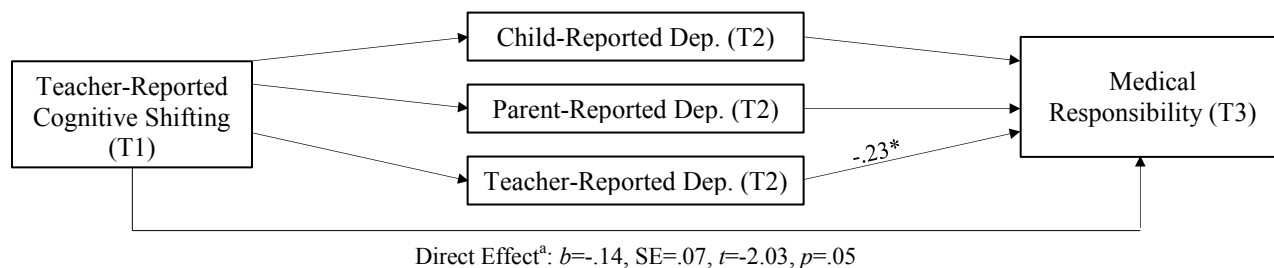


Figure 4e. Notes. ^aDirect effect of teacher-reported cognitive shifting on medical responsibility in model controlling for parent-reported depressive symptoms as a mediator. * $p < .05$.

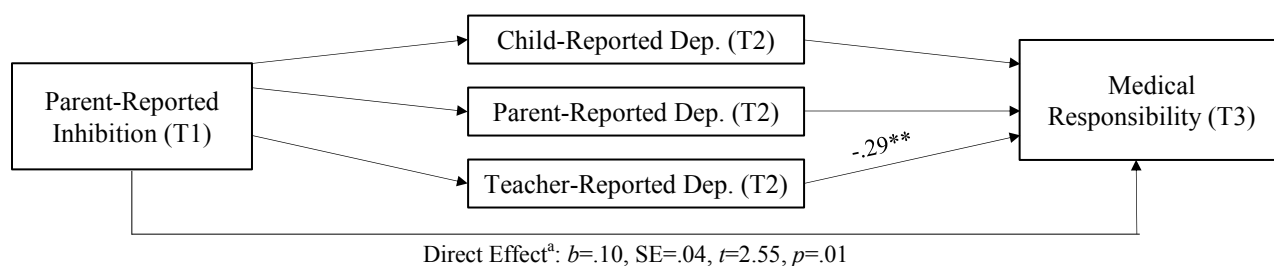


Figure 4f. Notes. ^aDirect effect of parent-reported inhibition on medical responsibility in model controlling for teacher-reported depressive symptoms as mediator. ** $p < .01$.

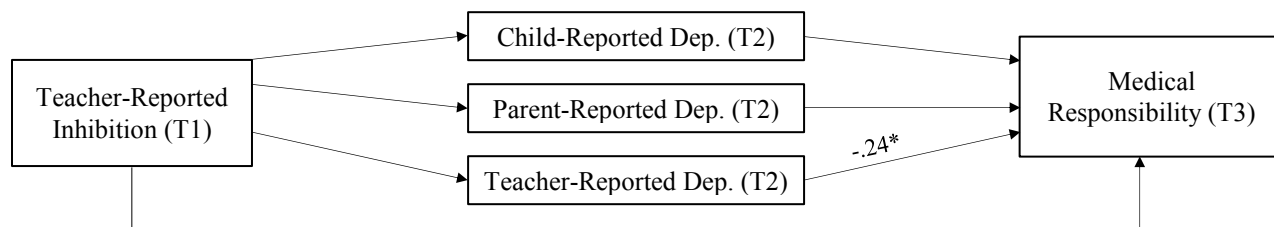


Figure 4g. Notes. * $p < .05$.

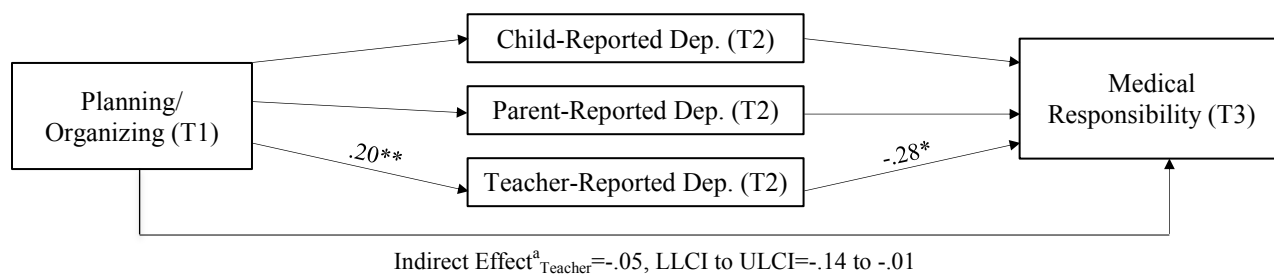


Figure 4h. Notes. ^aIndirect effect of planning/organizing on medical responsibility through teacher-reported depressive symptoms. Neither total effect nor direct effect was significant for the model with a significant indirect effect. * $p < .05$; ** $p < .01$.

²For Figures 4a-4h, analyses were tested separately for each of the three mediators and eight independent variables. In all models, attention, working memory, and planning/organizing represent global, composite factors.

Exploratory Analyses. Exploratory analyses were conducted to examine if neuropsychological functioning mediated the impact of child depressive symptoms on self-management in youth with spina bifida cross-sectionally at Time 1. Similarly to objective 1, neuropsychological factors as mediators and self-, parent-, and teacher-report of child depressive symptoms as the independent variable were tested separately, for a total of fifteen models. Figures 5a-5c display significant, main effects.

More severe self-reported depressive symptoms predicted greater deficits in attention ($b = 1.87, SE = .83, t = 2.25, p = .03$) and working memory ($b = 2.32, SE = .78, t = 2.96, p < .01$). More severe parent-reported child depressive symptoms also predicted greater deficits in attention ($b = 1.09, SE = .33, t = 3.34, p < .01$) and working memory ($b = 0.87, SE = .33, t = 2.65, p = .01$), as well as deficits in cognitive shifting (mother-report: $b = 0.87, SE = .25, t = 3.53, p < .01$; father-report: $b = 0.77, SE = .17, t = 4.46, p < .01$; teacher-report: $b = 0.61, SE = .27, t = 2.24, p = .03$), parent-reported inhibition ($b = 2.17, SE = .35, t = 6.26, p < .01$), and planning and organizing abilities ($b = 1.01, SE = .27, t = 3.75, p < .01$). Furthermore, more severe teacher-reported child depressive symptoms predicted greater deficits in attention ($b = 2.06, SE = .27, t = 7.76, p < .01$), working memory ($b = 1.84, SE = .27, t = 6.68, p < .01$), teacher-reported cognitive shifting ($b = 1.31, SE = .16, t = 8.24, p < .01$), teacher-reported inhibition ($b = 2.57, SE = .32, t = 7.95, p < .01$), and planning and organizing abilities ($b = 1.16, SE = .27, t = 4.32, p < .01$).

When self-reported depressive symptoms were included in the model, greater mother-reported deficits in cognitive shifting predicted less child responsibility for medical care ($b = -0.19, SE = .09, t = -2.28, p = .03$). When parent-reported depressive symptoms were included in

the model, greater deficits in attention ($b = -0.11$, $SE = .04$, $t = -2.57$, $p = .01$), as well as teacher-reported cognitive shifting ($b = -0.20$, $SE = .08$, $t = -2.44$, $p = .02$), predicted less child responsibility for medical care.

Results indicated a significant direct, negative effect of teacher-reported child depressive symptoms on child medical responsibility, such that greater child depressive symptoms predicted less child responsibility for medical care. The direct effect was significant only in models examining inhibition ($b = -0.35$, $SE = .16$, $t = -2.12$, $p = .04$) and planning and organizing abilities ($b = -0.29$, $SE = .13$, $t = -2.13$, $p = .04$) as mediators.

The indirect effects of self-reported depressive symptoms on child responsibility for medical care through attention (indirect estimated effect = $-.16$, $SE = .11$, 95% LLCI to ULCI = $-.48$ to $-.01$) and working memory (indirect estimated effect = $-.18$, $SE = .13$, 95% LLCI to ULCI = $-.54$ to $-.01$), respectively, were significant. Additionally, the indirect effects of parent-reported depressive symptoms on child medical responsibility through attention (indirect estimated effect = $-.12$, $SE = .07$, 95% LLCI to ULCI = $-.29$ to $-.02$), mother-reported cognitive shifting (indirect estimated effect = $-.16$, $SE = .08$, 95% LLCI to ULCI = $-.34$ to $-.03$), and teacher-reported cognitive shifting, respectively (indirect estimated effect = $-.13$, $SE = .08$, 95% LLCI to ULCI = $-.32$ to $-.01$), were significant. However, because the magnitude of the direct effect was greater than the total effect in these five instances, results indicated statistical suppression as opposed to mediation (MacKinnon, Krull & Lockwood, 2000). Therefore, these findings will be regarded as statistical artifacts that will not be interpreted as mediation.

Figures 5a-5c. Cross-Sectional Mediation Model of Child Depressive Symptoms, Neuropsychological Functioning, and Self-Management at Time 1³.

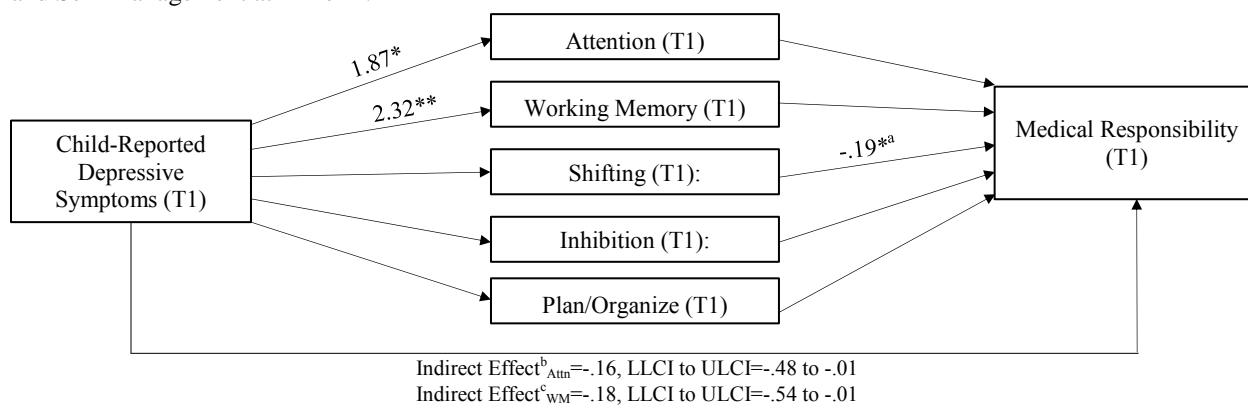


Figure 5a. Notes. ^aMother-reported cognitive shifting; ^bIndirect effect of child-reported depressive symptoms on medical responsibility through attention; ^cIndirect effect of child-reported depressive symptoms on medical responsibility through working memory. Neither total effects nor direct effects were significant for either model with significant indirect effects. * $p < .05$; ** $p < .01$.

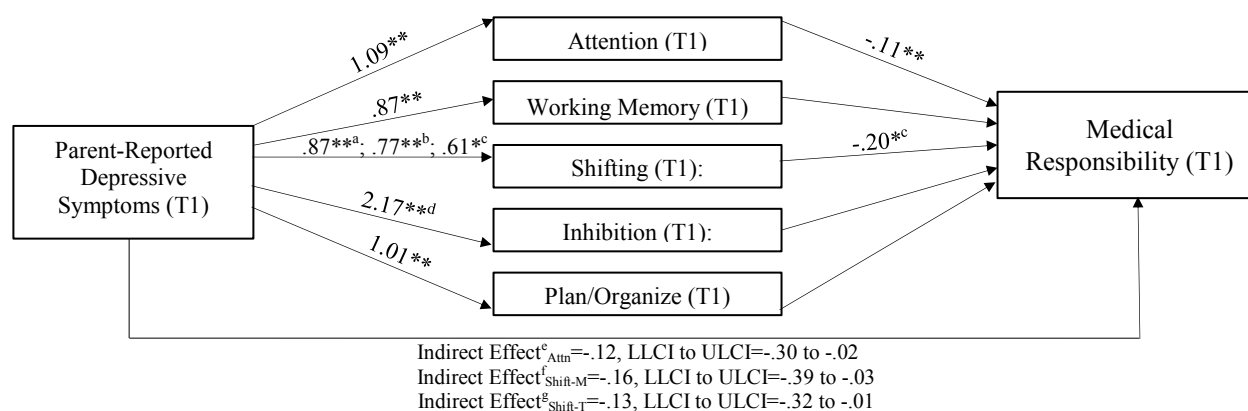


Figure 5b. Notes. ^aMother-reported cognitive shifting; ^bFather-reported cognitive shifting; ^cTeacher-reported cognitive shifting; ^dParent-reported inhibition; ^eIndirect effects of parent-reported depressive symptoms on medical responsibility through attention; ^fIndirect effects of parent-reported depressive symptoms on medical responsibility through mother-reported cognitive shifting; ^gIndirect effects of parent-reported depressive symptoms on medical responsibility through teacher-reported cognitive shifting. Neither total effects nor direct effects were significant for the three models with significant indirect effects. * $p < .05$; ** $p < .01$.

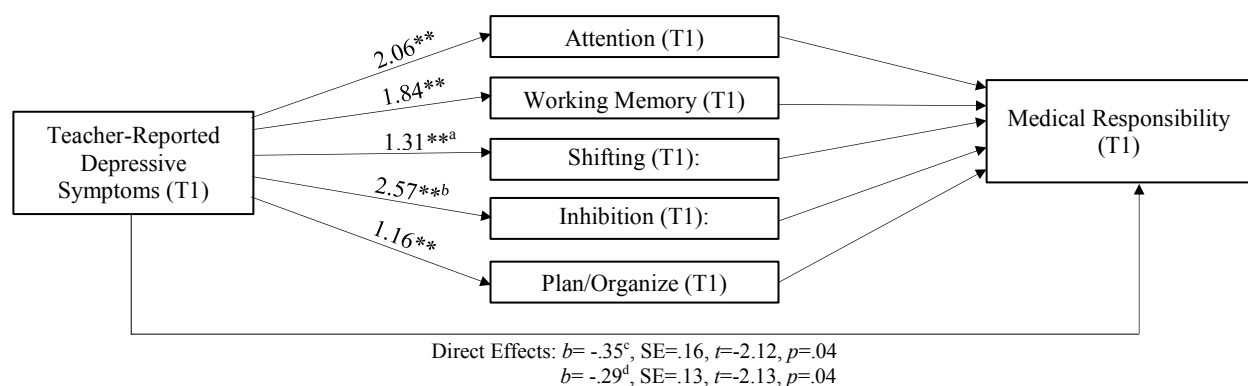


Figure 5c. Notes. ^aTeacher-reported cognitive shifting; ^bTeacher-reported inhibition; ^cDirect effect of teacher-reported depressive symptoms on medical responsibility in model controlling for inhibition as a mediator; ^dDirect effect of teacher-reported depressive symptoms on medical responsibility controlling for planning/organizing as a mediator. * $p < .05$; ** $p < .01$.

³For Figures 5a-5c, analyses were tested separately for each of the five mediators and three independent variables. In all models, attention, working memory, and planning/organizing represent global, composite factors.

CHAPTER FIVE

DISCUSSION

For adolescents with a chronic medical condition, learning to self-manage one's health is an essential prerequisite to achieving functional independence and preparing to transition to adult health care. While complete or excessive autonomy of medical care can compromise health outcomes in youth, constrained medical autonomy of a child with a chronic illness can stifle initiative and lead to difficulties with medical dependency later in young adulthood (Wysocki et al., 1996). Thus, adolescents with chronic medical conditions are encouraged to gradually gain more responsibility for their health care over the course of adolescence and emerging adulthood. The Pediatric Self-Management Model has identified depressive symptoms and neuropsychological deficits as two individual factors that influence medical self-management in children and adolescents with chronic illnesses (Modi et al., 2012). Less remains known about how these individual factors impact complex illness-specific health behaviors in youth with spina bifida, despite the increased risk for elevated symptoms of depression and pattern of cognitive dysfunction associated with the condition itself. Furthermore, past research suggests that depressive symptoms may hinder self-management via impaired executive and attentional abilities (Kichler, Moss, & Kaugars, 2012). However, this pathway has only partially been tested in youth with other chronic illnesses (e.g., diabetes; McGrady & Hood, 2010), and research examining how cognitive and psychosocial factors may be related to one another within the context of self-management remains scant.

The current study attempted to address this gap in the literature by examining the impact of depressive symptoms and neurocognitive deficits on two components of self-management, medical autonomy and adherence, over time in youth with spina bifida. Spina bifida-related health behaviors were examined during pre-adolescence and adolescence, a pivotal stage of development marked by the start of transfer of medical responsibilities from the parents to the child. This study sought to incorporate depressive symptoms, neuropsychological functioning, and spina bifida-specific health behaviors into a single, empirical model by examining cognitive impairment as a mediator through which depressive symptoms impacted self-management. Given the specific cognitive deficits associated with spina bifida and their negative effect on psychosocial functioning (Kelly et al., 2012; Lennon, Klages, Amaro, Murray, & Holmbeck, 2015), this study also sought to test an alternate direction, examining depressive symptoms as one pathway through which cognitive dysfunction hinders self-management.

It was hypothesized that more severe depressive symptoms would predict greater deficits in attention and executive functioning (**Hypothesis 1**), which in turn would predict lower medical responsibility and adherence (**Hypothesis 2**). While greater depressive symptoms were also expected to be related to spina bifida self-management, neurocognitive deficits were expected to mediate, or explain this relationship (**Hypothesis 3**). In other words, youth with greater depressive symptoms would demonstrate more profound deficits in attention and executive functioning than their better-adjusted peers, and these depleted cognitive resources would impair their ability to be medically autonomous and adherent.

To clarify the ambiguous causal relationship between depressive symptoms and neurocognitive deficits, an alternative direction was tested. It was hypothesized that greater

deficits in attention and executive functioning would predict more severe depressive symptoms (**Hypothesis 4**), which would in turn predict poorer medical responsibility and adherence (**Hypothesis 5**). While greater neurocognitive deficits were also expected to be related to poorer spina bifida self-management, depressive symptoms were expected to mediate this relationship (**Hypothesis 6**). Put another way, youth with worse attention and executive functioning abilities would be at increased risk for developing depressive symptoms, which would compromise their medical autonomy and adherence.

The results of the current study indicated that neuropsychological deficits did not significantly mediate the relationship between depressive symptoms and medical responsibility over time. However, depressive symptoms significantly mediated the relationship between attention and medical responsibility, as well as the relationship between working memory and medical responsibility, over time. Additionally, significant findings emerged for some of the individual pathways linking depressive symptoms, neurocognitive functioning, and medical autonomy. Child depressive symptoms were related to medical responsibility over time. The valence of the relationship was inconsistent, and dependent upon the reporter of depressive symptoms. Greater depressive symptoms as reported by the teacher were related to less medical autonomy over time, while greater depressive symptoms reported by parents were related to more medical autonomy over time. However, it should be noted that teacher depressive symptoms consistently predicted lower levels of medical responsibility across models, while parent depressive symptoms only predicted greater medical responsibility in one model. Further, this discrepancy may be due to statistical suppression (see Pandey & Elliott, 2010, for further explanation). Deficits in attention, working memory, cognitive shifting, and inhibition also

predicted less child responsibility for medical care over time. When examining the relationship between depressive symptoms and neuropsychological functioning, more severe depressive symptoms predicted greater deficits in cognitive shifting and inhibition over time, while deficits in attention, working memory, and planning and organizing abilities predicted greater depressive symptoms over time. It is important to note that findings varied across reporters; for example, some findings were significant for parent-report but not child- or teacher-report. Although the results are not completely consistent, several conclusions can be drawn. The discussion will focus mainly on the pathways in the conceptual model that were significant.

Associations between Depressive Symptoms and Self-Management

When controlling for age, disease severity, and socioeconomic status, greater teacher-reported depressive symptoms predicted less medical autonomy concurrently and prospectively. These results suggest that youth with more teacher-rated depressive symptoms struggled to develop independence with their spina bifida care. This finding parallels evidence linking depressive symptoms to poorer treatment adherence in youth with chronic health conditions (Hilliard, Wu, Rausch, Dolan, & Hood, 2013; McGrady & Hood, 2010), while extending the relationship beyond adherence to medical autonomy. Indeed, research has suggested that poor psychological adjustment in adolescents complicates the transition of health care responsibilities (Reed-Knight, Blount, & Gilleland, 2014). Although the current study did not assess parent-child beliefs surrounding health care transition or more general motivations towards autonomy, youth with more depressive symptoms may have been perceived to be not capable, not ready, or not willing to increase medical autonomy as compared to their less depressed peers.

Lower levels of concurrent and future medical autonomy may also have been related to a decline in child medical adherence due to depressive symptoms (i.e., responsibilities were relieved from the adolescent due to poor care), but this study was unable to test models that included adherence due to the low sample size of youth who were responsible for their medical tasks. Future studies should examine how motivational or dyad-level factors play a role in this relationship, as it is unclear if parents perceived their child to be less able to complete health care tasks due to increased psychological burden, or if youth with more depressive symptoms were less assertive about assuming more medical responsibilities. Future research should also examine the short and long-term adaptive function of reduced medical autonomy in youth with depressive symptoms, as physical health outcomes may be more favorable when parents take control over medical care for adolescents with elevated depressive symptoms.

Interestingly, more severe parent-reported depressive symptoms at Time 1 predicted more medical autonomy four years later at Time 3. This finding only occurred in one model, was unexpected, and is inconsistent with prior literature. It suggests that the temporal association between depressive symptoms and medical autonomy in spina bifida may be distinct from other chronic illnesses. Although this finding may seem counter-intuitive, it is speculated that the positive relationship between depressive symptoms and medical responsibility over time could reflect a “wraparound” effect. Specifically, youth with more depressive symptoms at baseline (Time 1) could have received additional support services and may have been responsible for managing their depressive symptoms early on (e.g., completing assignments for therapy), such that the family felt more comfortable with transitioning spina bifida-specific responsibilities later. Additionally, youth with greater depressive symptoms at baseline may not have

experienced increased psychological burden by taking on more responsibility for their spina bifida. An alternative explanation for these differences could lie in potential moderators of medical autonomy that were not assessed in this study, such as the role of family functioning or parent behaviors.

It is important to note that this positive relationship was only found with parent-reported depressive symptoms and that direct effects of parent-reported depressive symptoms were significant in only one out of five longitudinal models, while greater teacher-reported depressive symptoms predicted less medical responsibility and demonstrated consistent significant effects across all longitudinal models. It is likely that this singular finding represents a statistical suppression effect, as teacher-, but not parent-reported depressive symptoms were significantly associated with medical responsibility in bivariate correlations. Also, teachers may be more objective reporters of depressive symptoms in youth with spina bifida, as they are able to compare the adolescent to other same-aged peers. However, parents may be more in tune to subtle changes in behavior that indicate fluctuations in depressive symptoms in their children with spina bifida. These differences indicate that the association between depressive symptoms and medical autonomy in spina bifida warrants more attention.

Impact of Neuropsychological Functioning on Self-Management

Neuropsychological functioning was a robust predictor of medical autonomy in youth with spina bifida both concurrently and prospectively. This study replicates previous findings (Tarazi, Mahone, & Zabel, 2007; O'Hara & Holmbeck, 2013) and lends further support to the bio-neuropsychosocial model of medical autonomy and adherence in youth with spina bifida (Psihogios, Murray, Zebracki, Acevedo, & Holmbeck, 2016). Managing the complex

symptomology of spina bifida requires the coordination of multi-step, complicated tasks on a daily basis. Neurocognitive deficits, especially in attention and executive functioning, may complicate one's ability to manage spina bifida tasks independently. Furthermore, the lack of expected maturation in executive abilities across adolescence in spina bifida can complicate how an adolescent with spina bifida assumes medical responsibilities relative to youth with other chronic medical conditions (Tarazi, Zabel, & Mahone, 2008). Children with greater cognitive deficits may be more reluctant to take on new responsibilities related to their spina bifida, reflecting a difficulty with initiation; or, parents and health providers may perceive their children with greater executive and attentional deficits as less capable of maintaining responsibility for their medical care. O'Hara and Holmbeck (2013) found support for the prior interpretation, in that medical autonomy in youth with spina bifida was more influenced by child functioning than parenting behaviors. However, as their findings were based on cross-sectional rather than longitudinal data, parents may change their parenting style in reaction to their children's neurocognitive challenges, which may impact the development of medical autonomy over time.

In this study, different neuropsychological factors appeared to have varying effects on medical independence. Greater attentional problems and poorer cognitive shifting predicted less medical autonomy both cross-sectionally and longitudinally, while poorer working memory and inhibition (at Time 2) predicted less medical autonomy longitudinally but not cross-sectionally. These findings highlight the importance of teasing apart the impact of higher order cognitive skills on autonomy development in adolescents with spina bifida. The difficulties that inattention and cognitive rigidity cause may be more noticeable in the short-term. Alternatively, deficits in these domains may have a more immediate impact on medical autonomy than working memory

or inhibition. Difficulties with attention and cognitive shifting may manifest as issues with selecting appropriate self-management goals and shifting attention to focus on completing those tasks, as well as thinking flexibly to problem-solve issues related to self-management. If medical tasks change over time (e.g., a change in preferred bowel program), poor inhibition may impact one's ability to inhibit reflexive responses, or constrain habitual behavior, in order to follow through with spina bifida medical tasks. Furthermore, working memory deficits can cause difficulty with multitasking and "remembering to remember" to complete tasks related to one's medical regimen, such as catheterizing, conducting skin checks, or taking medications. As the demands of daily living increase over adolescence, a child with spina bifida who has working memory problems may have difficulty integrating the responsibilities associated with typically developing youth (e.g., homework) with responsibilities related to his or her medical care.

While almost all significant pathways were in the expected direction, such that more deficits in executive functioning and attention predicted less medical autonomy, greater problems with inhibition at Time 1 predicted more medical responsibility four years later at Time 3. This finding may be due to differences in the reporter of inhibitory control, as greater parent-reported inhibition at Time 1 was related to more medical autonomy at Time 3, while greater teacher-reported inhibition at Time 2 was associated with less medical autonomy at Time 3. However, this relationship requires further investigation to parse apart the differential impact of reporter versus time point on the relationship between inhibition and medical responsibility.

It is evident that difficulties with attention and executive functioning can lead to increased dependency and delays in other domains of development (e.g., the process of transitioning to adult care; Tuminello, Holmbeck, & Olson, 2012; Sawin et al., 2003). Despite

the increased risk in medical independence associated with cognitive deficits, these results do not indicate that self-management is an unattainable goal for all individuals with spina bifida who have attention problems or executive dysfunction. Rather, future research could examine these skills as a target for intervention. Cognitive training and goal management programs designed to strengthen or remediate executive and attentional functioning in individuals with spina bifida may be tailored to focus on medical independence as a secondary outcome (Stubberud, Langenbahn, Levine, Stanghelle, & Schanke, 2014). Specialized technologies or environmental modifications could also be implemented to support areas of executive or attentional weakness in individuals with spina bifida as they develop medical autonomy. For example, organizational applications for smart phones can help those with working memory deficits remember long-term events, such as doctor appointments, and visual schedules can prompt those with poor attention and working memory to complete each step of a complicated medical task, such as clean intermittent catheterization. Additionally, providers may focus on individuals' cognitive strengths as a way to compensate for their weaknesses in promoting independence with their spina bifida.

Relationship between Depressive Symptoms and Neuropsychological Deficits

This study also sought to clarify the relationship between depressive symptoms and neuropsychological dysfunction in youth with spina bifida. Previous research has found support for the cognitive vulnerability hypothesis, where executive functioning deficits have predicted internalizing symptoms in children and adolescents with spina bifida (Kelly et al., 2012; Lennon, Klages, Amaro, Murray, & Holmbeck, 2015). This study extended the literature by also examining the potential impact of depressive symptoms on executive functioning, including

attentional problems as a neuropsychological factor that may be related to psychosocial adjustment, and by using a longitudinal design to investigate directionality.

Results indicated that the directionality of this relationship in youth with spina bifida depended on the cognitive domain. In support of the cognitive scarring hypothesis, greater depressive symptoms exacerbated deficits in cognitive shifting and inhibition over time. Depressive symptoms may contribute to difficulties in cognitive shifting by impairing the ability to flexibly interpret information or adapt to changing environmental demands (Joormann & Quinn, 2014). Depressive symptoms may also hinder inhibitory control by weakening one's ability to stop negative, intrusive thoughts, or leading to more impulsive patterns of information processing and behavior (Kyte, Goodyer, & Sahakian, 2004) further impacting emotional and cognitive well-being (Joormann & Quinn, 2014). Additionally, depressive symptoms significantly predicted greater deficits in all neuropsychological domains cross-sectionally. However, the methodological limitations of cross-sectional research prevent inferences about directionality from being made. Thus, interpretation was limited to longitudinal findings.

In contrast, deficits in attention, working memory, and planning and organizing abilities predicted more depressive symptoms over time. Kelly et al. (2012) similarly found that decreased working memory, planning and organizing abilities predicted more depressive symptoms, and suggested that difficulties in these areas may interfere with multitasking and planning for long term goals, which may elicit negative feedback from parents and teachers. Recent research revealed that executive and attentional dysfunction may also lead to more severe internalizing symptoms indirectly, via impaired social competence (Lennon, Klages, Amaro, Murray, & Holmbeck, 2015). One interpretation of the results is that depressive symptoms may

lead to exacerbation of pre-existing deficits or cognitive “scarring” in cognitive flexibility and inhibition, while deficits in attention, working memory, and planning and organizing abilities may be conceptualized as neuropsychological risk factors that leave an individual with spina bifida particularly vulnerable to developing depressive symptoms.

An alternative explanation for these results may lie in how the neuropsychological constructs were assessed. Attention, working memory, and planning and organizing abilities were assessed using performance-based measures in addition to both parents’ and teachers’ ratings on a questionnaire. Cognitive flexibility and inhibition were assessed only by parent and teacher ratings. Questionnaire-based measures reflect behavioral and social components of attentional and executive functioning (Miranda et al., 2015). They capture how the child is able to use his or her cognitive skills to pursue goals without explicit guidance, and meet the demands of real life (Toplak, West, & Stanovich, 2013). Conversely, performance-based measures may evaluate the cognitive component of these skills (Rose & Holmbeck, 2007), and the extent to which the individual can execute goals when the aims are explicitly laid out for him or her (Toplak, West, & Stanovich, 2013). Since attention, working memory, and planning and organizing included more objective, cognitive measures, these variables may have captured additional risk factors that were contributing to depressive symptoms over time, beyond behavioral manifestations of attentional and executive dysfunction. Youth with spina bifida may have been able to minimize the effects of deficits in inhibitory control and mental flexibility on real-life situations. Finally, developmental influences should be taken into account when interpreting these results. Executive functioning and attentional skills are not fully developed until the mid-twenties, and youth with spina bifida continue to experience delays in the growth of

these abilities through adolescence and emerging adulthood. Given these differing developmental trajectories, and the vulnerability to changes in cognitive functioning based on transient medical factors (e.g., shunt infections), it is possible that the relation between depressive symptoms and cognitive deficits in spina bifida changes over time.

Mediation Effects

Deficits in attention and working memory indirectly impacted responsibility via increased depressive symptoms. These findings suggest that one way in which certain neurocognitive deficits may prevent youth with spina bifida from gaining autonomy over their medical care is through an increased risk for developing depressive symptoms. From a clinical perspective, it is possible that youth with poor attention and working memory have difficulty following instructions and completing multi-step tasks. This may lead to increased challenges across multiple environments (e.g., home, school, community) followed by decreased self-esteem and greater depressive symptoms, which may act as a barrier to medical autonomy. Thus, when conceptualizing the growth of medical autonomy in spina bifida, it is important to consider not only the congenital neurocognitive impairments associated with spina bifida, but also the way in which these deficits leave youth vulnerable to increased depressive symptoms.

However, the current study did not find that depressive symptoms indirectly impacted self-management through neuropsychological dysfunction, and the exact mechanism through which depressive symptoms may influence future independence in medical care remains unclear. The lack of findings in the opposite direction may be due to the lack of adequate power to detect small effects in analyses. It may also be that depressive symptoms have an impact on medical responsibility that is not dependent on how neuropsychological functioning is impacted by

depressive symptoms. Given the developmental stage of participants, which spans from pre-adolescence to emerging adulthood, it is plausible that family or peer factors may explain the relationship between depressive symptoms and self-management more so than cognitive dysfunction. Indeed, depressive symptoms have demonstrated bidirectional relationships with higher levels of peer conflict in adolescents (Kochel, Ladd, & Rudolph, 2012), which have recently been identified as a barrier to medical autonomy in youth with spina bifida (Psihogios, Murray, Zebracki, Acevedo, & Holmbeck, 2016). Other individual factors, such as lowered intrinsic motivation or self-efficacy to manage one's medical condition, may explain this relationship as well.

Strengths, Limitations, and Future Research

This study had several strengths. First, the current study expanded the limited knowledge of self-management in youth with spina bifida by examining potential neurocognitive factors and depressive symptoms as predictors. Second, the current study used multiple methods and reporters, which allowed for more stable examinations of child functioning across environments. Importantly, when constructs could not be aggregated across reporters, they were examined separately, allowing for the examination of different perspectives. Administering performance- and questionnaire-based assessments of executive and attentional functioning have been encouraged in research, as they capture distinct components of these cognitive domains in structured and unstructured settings (Toplak, West, & Stanovich, 2013). Third, a longitudinal, mediational design was used to examine relationships over time, which allowed for consideration of *why* and *how* depressive symptoms and cognitive functions impacted the development of medical autonomy in adolescence.

However, there are several limitations of the current study that should be addressed in future work. To focus on individuals who were cognitively capable of achieving and maintaining self-management, this study excluded participants who scored two standard deviations or more below average on a full scale IQ measure (i.e., <70). Below this level, there may be significant cognitive and adaptive impairments that make completing self-management tasks autonomously an unrealistic goal (Harris, 2013). However, excluding participants with spina who have a lower IQ prevents these findings from being generalized to all youth with spina bifida. The understanding of how certain cognitive and emotional factors may impact medical autonomy was limited by excluding those with a lower IQ, as their development of medical responsibility may differ from those with a higher IQ. Future research may aim to include individuals with a lower IQ to gain a better understanding of the challenges they face in trying to self-manage their spina bifida.

Furthermore, this study found interesting relationships among executive functioning, attention, depressive symptoms, and medical autonomy. As cognitive deficits are a direct consequence of spina bifida itself, these findings may not be representative of youth with chronic illnesses that do not congenitally impact the central nervous system. For the same reason, the findings of how depressive symptoms and certain cognitive factors may uniquely influence one another may be unique to children with spina bifida, and may not be able to be generalized to typically developing youth.

Due to small sample size, this study was underpowered to detect small effects in analyses, and was unable to conduct analyses with medical adherence as an outcome. Medical adherence is distinct from medical autonomy, and is an important part of self-management. It is

possible that neuropsychological impairment may mediate the relationship between depressive symptoms and medical self-management in youth with spina bifida, but this study was unable to examine such relationships. Indeed, most research on depressive symptoms and pediatric health behaviors has focused on adherence to treatment regimen rather than medical autonomy (La Greca & Mackey, 2009). Future collaboration across multiple sites may provide researchers with a larger sample size to investigate variables that are associated with medical adherence in this population.

While a strength of this study was its multi-method assessment of cognitive variables, not all neuropsychological domains were measured uniformly. Some domains (i.e., attention, working memory, planning/organizing) incorporated performance-based and report-based measures, while others (i.e., cognitive flexibility, inhibition) were assessed only using a questionnaire. As previously stated, performance-based and questionnaire measures assess different components of attentional and executive functions, and it is possible that objectively measured components of cognitive shifting and inhibition may have had a different relationship with depressive symptoms or medical autonomy. Further, individuals who participated at Times 1, 2, and 3 had less difficulties in cognitive flexibility and were younger than those who did not participate at all three times points, and thus were excluded from analyses. These significant differences may have impacted findings. Additionally, gaps between time points spanned approximately two years. It is possible that depressive symptoms indirectly impacted a child's ability to take responsibility for his or her medical care via impaired cognitive functioning over a shorter time period (e.g., months). This study was unable to investigate potentially more subtle changes in medical autonomy. Finally, age was a significant predictor of medical autonomy, but

the current study did not examine how depressive symptoms or neurocognitive deficits may differentially impact health behaviors based on age. A wide range of ages was included in this study (i.e., ages 8-15 at Time 1), and future research should investigate if differences among these individual factors varied based on developmental stage.

Additionally, while this study aimed to investigate two pathways in depth, it did not include potentially important factors related to self-management processes, such as peer relationships or parenting influences (Modi et al., 2012; O'Hara & Holmbeck, 2013). Indeed, past research has shown that peer and family factors, such as peer conflict and family cohesion, have a unique impact on medical autonomy and adherence in youth with spina bifida (Psyhogios, Murray, Zebracki, Acevedo, & Holmbeck, 2016). To date, no studies have examined the influence of community or macro-level (e.g., health care system) factors on spina bifida self-management outcomes. Inclusion of these broader dyad- and community-level influences in future research would help build a more comprehensive picture of how cognitive and affective functioning impacts self-management over time in spina bifida.

Moreover, while these findings established that depressive symptoms are one pathway through attention and executive functioning influence medical autonomy, examining other potential mediators was beyond the scope of the present study. For example, cognitive functioning and depressive symptoms could be related to parent and child readiness to transfer medical responsibilities, or self-efficacy surrounding medical self-management, which may play an integral role in the development of medical autonomy.

Conclusions and Clinical Implications

The results of the current study have important implications for promoting the growth of medical autonomy in youth with spina bifida. First, building off of Modi et al.'s (2012) comprehensive model of pediatric self-management and Psihogios et al.'s (2016) bio-neuropsychosocial model for self-management in youth with spina bifida, it appears that depressive symptoms, attention, and executive functioning are intertwined and have a unique impact on medical autonomy in this population. Psychological screenings have been shown to predict disease management in adolescents with type 1 diabetes (Hilliard, Herzer, Dolan, & Hood, 2011). Results from this study suggest that regular psychological screenings could help clinicians identify depressive symptoms early on that may be negatively impacting health autonomy in preadolescents and adolescents with spina bifida. Clinical interventions aimed at facilitating the transfer of healthcare responsibilities to the child may maximize treatment success by taking into account an individual's level of depressive symptoms and executive and attentional skills. Given the robust association between these neurocognitive factors and medical autonomy, providers who want to encourage families to begin the transfer process may choose to incorporate cognitive training programs into a treatment plan for a preadolescent with spina bifida who is struggling in these areas (Stubberud, Langenbahn, Levine, Stanghelle, & Schanke, 2014), as executive and attention difficulties are commonly identified during this developmental stage (Rose & Holmbeck, 2007).

Second, rather than depressive symptoms hindering self-management outcomes by exacerbating cognitive deficits, depressive symptoms appear to be one pathway through which attention and executive impairment may hinder medical autonomy. This key finding paves the

way for further research on other pathways that may mediate the impact of neuropsychological functioning on medical autonomy in spina bifida. Further, given the increased prevalence of depressive symptoms in youth with spina bifida, this study serves as a call for research on other factors that may explain the relationship between depressive symptoms and medical autonomy (e.g, intrinsic motivation, self-efficacy). Given the demonstrated longitudinal patterns, it is evident that these individual factors should be monitored throughout the course of adolescence, as depressive symptoms, attention abilities, and executive functions may fluctuate across development.

Third, findings from this study have revealed that different executive and attentional skills have unique temporal relationships with depressive symptoms in youth with spina bifida. While the congenital neural impairment found in spina bifida myelomeningocele precludes these findings from being generalized to other populations, distinct patterns linking cognitive and emotional functioning emerged. More severe depressive symptoms appeared to weaken cognitive shifting and inhibitory control over time, while greater deficits in attention, working memory, and planning/organizing abilities were predictive of future increased depressive symptoms. These relationships may inform the development of evidence-based psychological interventions in youth with spina bifida, as providers may conceptualize weaknesses in certain cognitive areas (e.g. attention, working memory) as risk factors for future depressive symptoms, and challenges in other areas (i.e., inhibition, cognitive flexibility) as cognitive consequences of depressive symptoms in adolescents with spina bifida.

APPENDIX A
MEASURES

Questionnaire Measures (Alphabetized):

Behavior Rating Inventory of Executive Function (BRIEF)

Child Behavior Checklist (CBCL)

Children's Depression Inventory (CDI)

Medical History Questionnaire (MHQ)

Sharing of Spina Bifida Management Responsibilities Scale (SOSBMR)

Spina Bifida Self-Management Profile (SBSMP)

Swanson, Nolan, and Pelham – Fourth Edition (SNAP-IV)

Teacher Report Form (TRF)

Direct Assessment Measures:

Cognitive Assessment System (CAS)

Wechsler Abbreviated Scale of Intelligence (WASI)

Wechsler Intelligence Scale for Children-Fourth Edition (WISC-IV)

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VITA

Alexa is a doctoral student at Loyola University Chicago studying clinical psychology with a specialty in child, adolescent, and family issues. She received her B.A. in Psychology from the University of Pennsylvania in 2013, graduating summa cum laude. During her time as an undergraduate at the University of Pennsylvania, she conducted research on posttraumatic stress in ill and injured children and coping in pediatric cancer under the guidance of Dr. Meghan Marsac. After graduating, Alexa worked as a research coordinator for the Behavioral Diabetes Team at Children's National Health System, studying adjustment and adherence in children and adolescents with type 1 diabetes. Through her work on these projects, she presented research at multiple regional and national conferences, and co-authored several peer-reviewed journal articles. Since starting graduate school at Loyola, Alexa has been a member of Dr. Grayson Holmbeck's research lab, studying families of youth with spina bifida. As part of this lab, she has worked on multiple projects highlighting her different interests. These include projects examining the impact of parenting behavior on medical adherence in youth with spina bifida, and investigating the influence of executive functioning and self-advocacy on academic achievement in youth with spina bifida. Alexa's master's thesis examined the relationships among neuropsychological functioning, depressive symptoms, and self-management in youth with spina bifida. Work on these various projects has resulted in numerous presentations, peer-reviewed journal articles, and book chapters.