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Longitudinal Associations among Individual Factors, Parenting Behaviors, and Medical Responsibility in Youth With Spina Bifida: Mediation, Moderation, and Growth Analyses

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LONGITUDINAL ASSOCIATIONS AMONG INDIVIDUAL FACTORS, PARENTING BEHAVIORS, AND MEDICAL RESPONSIBILITY IN YOUTH WITH SPINA BIFIDA: MEDIATION, MODERATION, AND GROWTH ANALYSES

A DISSERTATION SUBMITTED TO
THE FACULTY OF THE GRADUATE SCHOOL
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BY
ALEXA R. STERN

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The objective of this research was to examine how youth individual factors (neuropsychological functioning and depressive symptoms) and maternal and paternal acceptance, behavioral control, and psychological control were associated with child medical responsibility among youth with spina bifida (SB). These longitudinal studies examined multimethod, multi-informant data from families of youth with SB, their parents, and teachers. The first study used bootstrapping methods to examine two competing, mediational pathways through which depressive symptoms, executive functioning, and attention were associated with medical responsibility over time. The second study used moderation analyses to examine how parenting behaviors moderated the relationship between these cognitive skills and medical responsibility over time. The third study used mixed methods growth analyses to explore how neuropsychological factors and parenting behaviors were related to trajectories of medical responsibility across adolescence and young adulthood, utilizing a task specific approach. Results are discussed within the context of broader social-ecological frameworks of pediatric self-management. These findings have implications for potential interventions targeted at helping families manage the transition from parent- to self-management of SB medical tasks. Further investigation of the impact that individual, family, and community factors have on the unfolding of medical responsibility among youth with SB is warranted.
CHAPTER ONE
INTRODUCTION

Overview of Spina Bifida

Spina Bifida (SB; “split spine”) is the most complex congenital medical condition that is compatible with life. It occurs in the first trimester of fetal development when the neural tube fails to close completely, leaving the spinal column exposed and resulting in damage to the spinal cord and brain (Copp et al., 2015). Consequently, SB is associated with varying degrees of motor impairment, paralysis, sensory loss, orthopedic abnormalities (e.g., scoliosis), bowel and bladder dysfunction, and neurodevelopmental difficulties (Fletcher & Brei, 2010). SB is considered heterogeneous in nature, with the extent of disability and impairment dependent on the lesion level and presence of anomalies in the brain and spinal cord. Higher lesions in the spine are associated with greater motor impairment and paralysis (Copp et al., 2015).

There are different forms of SB categorized by varying levels of severity, including myelomeningocele, meningocele, lipomyelomeningocele, and occulta. The most frequent and severe type of SB is myelomeningocele, which occurs in 80-90% of cases (Sandler, 2010). With myelomeningocele, the spinal cord and nerves protrude from the back through the spinal column. The majority of individuals with myelomeningocele are also born with a Chiari II malformation (i.e., the displacement of the brain stem and cerebellum into the spinal canal) and develop hydrocephalus (i.e., excess cerebrospinal fluid in the brain; Fletcher & Brei, 2010). Youth with SB and shunted hydrocephalus are at increased risk for developing seizures (Sandler, 2010).
SB is associated with other structural abnormalities in the brain, including dysgenesis of the corpus callosum. The modal cognitive profile of youth with SB is characterized by a variety of strengths and weaknesses, with associative processing (e.g., retrieving information learned from repetition) being relatively intact compared to impairments in assembled processing (e.g., integrating information across contexts; Fletcher, Ostermaier, Cirino, & Dennis, 2008). In other words, youth with SB may demonstrate relative strengths in tasks that involve simpler relations and categorizing stimuli, such as recognizing faces or decoding familiar words, and relative weaknesses in tasks that require the formation of more complex relationships, such as inferring meaning from sentences and mentally rotating objects (Dennis, Landry, Barnes, and Fletcher, 2006). Most individuals with SB perform within the average to low average range on tests of intellectual functioning. Basic word reading and verbal skills tend to be preserved with relative weaknesses in more complex language and verbal abilities, such as reading comprehension and pragmatic language (Dennis et al., 2010). Weaknesses in memory and nonverbal skills, including mathematics, processing speed, and visual-spatial perception, are common (Dennis et al., 2010).

Notable deficits have been documented in executive functioning skills and attention abilities (Burmeister et al., 2005; Iddon, Morgan, & Sahakian, 1996; Rose & Holmbeck, 2007; Snow, 1999). Executive functioning refers to a set of higher order cognitive skills that allow one to self-regulate and engage in goal-directed behavior (Gioia, Isquith, Kenworth, & Barton, 2002). Specifically, individuals with SB have demonstrated impairments in working memory, problem solving, cognitive flexibility, inhibitory control, task initiation, planning, and organization (Mahone, Zabel, Levey, Verda, & Kindsman, 2002; Rose & Holmbeck, 2007;
Snow, 1999). Further, children with SB do not exhibit the same age-expected maturation in executive functioning skills across adolescence that typically developing youth exhibit (Tarazi, Zabel, & Mahone, 2008). Youth with SB have shown difficulties with focused attention, selective attention, and with shifting attention to a new task (Capsersen & Habekost, 2013; Fletcher et al., 1996; Ou, Snow, Byerley, Hall, & Glasier, 2013; Rose & Holmbeck, 2007).

Individuals with SB also face a number of psychosocial functioning challenges. They are at increased risk for developing anxiety and depressive symptoms, particularly as they approach adolescence and emerging adulthood (Appleton et al., 1997; Holmbeck et al., 2003). Studies have linked several factors, including negative perceptions of physical appearance, lower self-worth, higher levels of pain, difficulties with social acceptance, lack of social support, poorer family functioning, maladaptive parenting, with internalizing symptoms among youth with SB (Holmbeck et al., 2010; Kelly, Holmbeck, & O’Mahar, 2011; Oddson, Clancy, & McGrath, 2006). Adolescents with SB may also encounter difficulties when attempting to develop autonomy and assume self-care responsibility across contexts. Specifically, studies have shown that youth with SB are more passive, more dependent on adults for direction and guidance, less likely to make independent decisions, and responsible for fewer tasks at home than their peers, suggesting that autonomy development poses a significant challenge for this population (Holmbeck et al., 2003). Indeed, adolescents with SB have exhibited delays across multiple indices of autonomy, including behavioral, emotional, and decision-making autonomy (Davis, Shurtleff, Walker, Seidel, & Dunguy, 2006; Devine, Wasserman, Gershenson, Holmbeck, & Essner, 2011; Friedman, Holmbeck, Delucia, Jandasek, & Zebracki, 2009). In summary, studies
show that the developmental transition during adolescence is a vulnerable time for youth with SB.

**Medical Responsibility in Spina Bifida**

Due to the pervasive impact of SB, management is complex and requires monitoring by a multidisciplinary team. Youth with SB are often followed by a urologist, orthopedic surgeon, neurosurgeon, specialized nurses, physical therapist, occupational therapist, social worker, and psychologist. Surgery is performed within 48 hours of birth or prenatally to close the spinal cord (Bowman, Boshnjaku, & McLone, 2009). Children with SB may be required to have additional surgeries throughout development to manage orthopedic, neurological, or urinary issues. Hydrocephalus may necessitate surgical placement of a shunt, which poses additional complications including further surgeries to correct shunt infections or malfunctions. Shunt surgery frequency tends to be associated with poorer adjustment and cognitive outcomes (Dennis et al., 2006). Youth with SB may need assistive devices, such as crutches, or a wheelchair to ambulate. Many individuals with SB take medication regularly and practice clean intermittent catheterization to manage urinary incontinence and prevent urinary tract infections. Bowel programs to manage constipation or bowel incontinence can include suppositories, medication, laxatives, or enemas (Mitchell et al., 2004). Youth are often asked to follow special dietary modifications, including increased fiber and fluid intake to assist with bowel management, and avoidance of certain foods to manage the increased risk for latex allergy (Wittenbrook, 2010). Routine skin checks are recommended to detect pressure wounds in areas impacted by reduced sensation. Children and parents must also be vigilant for signs of a shunt malfunction. Failure to adhere to their medical regimen can lead to potentially dangerous, but often preventable,
secondary complications, including urinary tract infections, renal problems, gastrointestinal problems, shunt malfunctions, pressure wounds and infections, undetected skin injuries, and obesity.

Due to advancements in the medical treatment for youth with SB over the last several decades, the rate of survival to adulthood has increased to 75-85% (Liptak et al., 2013). A new challenge that families of children with SB must face is the transition to adulthood. As part of this transition, adolescents and young adults with SB must learn how to independently self-manage their medical regimen. Self-management is an overarching, multifaceted concept that refers to the interaction of health behaviors and processes people engage in as part of living with a chronic health condition (Modi et al., 2012). For pediatric self-management, this concept also refers to the ways that health behaviors occur within different domains beyond the individual, such as one’s family, broader community, and healthcare system (Modi et al., 2012). One important component of self-management is medical autonomy (also known as medical responsibility), which accounts for who in the family is primarily responsible for carrying out these condition-related tasks. Other components that may be examined within the umbrella of self-management include adherence (i.e., the degree to which a medical regimen is being followed according to the doctor’s recommendations), self-efficacy regarding one’s ability to complete medical tasks, or knowledge of one’s condition and medical history.

The studies described in this dissertation focus on the unfolding of medical responsibility and factors that impact this process. Similar to other types of autonomy, medical responsibility is an interactive, developmental process that involves a negotiation of responsibilities between adolescents and their caregivers (Friedman et al., 2009). Successful transfer of responsibilities
from family- to self-management allows youth with SB to achieve self-sufficiency and can pave the way for a smooth transition from the pediatric to adult healthcare system. While individuals with the most severe forms of SB 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 mild to moderate forms of SB should be able to independently manage most of these tasks before adulthood (Greenley, 2010). Unfortunately, many individuals with SB enter young adulthood with preventable secondary complications, indicative of poor self-management behaviors, and struggle for independence from their parents (Ridosh et al., 2011; Wagner et al., 2015). As such, it is imperative to understand not only the nature of how medical responsibility develops among youth with SB, but also factors that are associated with this process.

Theoretical frameworks that articulate the unfolding of medical responsibility in youth with chronic health conditions have adopted a social-ecological approach within a developmental systems perspective, and emphasize the dynamic interrelationships among modifiable and nonmodifiable factors across individual, familial, community, and systemic contexts (Modi et al., 2012; Reed-Knight, Blount, Gilleland, 2014). Within these contexts, the child’s developmental level is paramount and influences the level of responsibility they can assume for their healthcare tasks. Models specifically designed for youth with SB have been developed (e.g., the Bio-Neuropsychosocial Model of Adjustment; Holmbeck & Devine, 2010) that highlight the importance of relevant disease-specific (e.g., SB severity, lesion level), neuropsychological (e.g., executive functioning), and social (e.g., family functioning) influences. Longitudinal findings support a developmental trajectory where youth with SB gradually gain responsibility for medical tasks such as catheterization and bowel program management over time (Psihogios,
Interestingly, however, adolescents with SB may acquire autonomy for many non-medical self-management skills later than their peers (Davis et al., 2006). Moreover, the literature on medical and non-medical autonomy in this population remains scant and more studies are needed to characterize how youth with SB assume increased levels of responsibility.

**Role of Attention/Executive Functioning**

Existing literature points to higher-order cognitive functioning skills as modifiable, individual-level factors that may be particularly influential for medical responsibility in youth with SB. Specifically, executive functioning skills appear to play a prominent role in the development of autonomy across adolescence more broadly (Zimmer-Gembeck & Collins, 2008). The development of executive functioning skills is thought to mirror the development of autonomy across adolescence and young adulthood. As these cognitive skills mature, adolescents develop the ability to self-regulate their thoughts, emotions, and behaviors, multitask, consider long-term consequences, and engage in planning and organizing, all of which are needed to gain and manage greater independence. Executive functioning also allows adolescents to complete tasks more efficiently, and thus meet increasingly higher demands within home, school, and community contexts. With regard to pediatric chronic illness, executive functioning and complex aspects of attention are thought to be a critical part of the foundation of condition self-management, as youth must organize and direct their actions towards the goal of assuming responsibility for their healthcare regimen (Lansing & Berg, 2014; Modi et al., 2012).

As the medical regimen for SB requires a high degree of coordination, problem-solving, organization, and planning, it is not surprising that executive functioning is associated with self-
management among youth with SB. For example, for adolescents with SB to successfully self-catheterize on their own, they must plan when and how they will perform this task during school and at home. They may also be required to problem-solve relatively quickly when faced with unexpected changes in routine that impede their plans to catheterize (e.g., important class lesson, impromptu activities after school). Finally, poor organization (e.g., losing catheterization materials) can further impede task completion. Indeed, higher executive functioning skills, as measured by parent-report and performance-based tests, have consistently been related to greater medical responsibility and self-care skills in children and adolescents with SB (Donlau et al., 2011; Heffelfinger et al., 2008; Jacobson et al., 2013; O’Hara & Holmbeck, 2013; Psihogios, Murray, Zebracki, Acevedo, & Holmbeck, 2016; Ries et al., 2003; Tuminello, Holmbeck, & Olsen, 2012; Stern et al., 2018). Executive functioning has also demonstrated relationships with related outcomes among children, adolescents, and adults with SB, including adaptive functioning, intrinsic motivation, and the acquisition of adult developmental milestones (Heffelfinger et al., 2008; Stubberud & Riemer, 2012; Warschausky, Kaufman, Evitts, Schutt & Hurvitz, 2017). Attention, while less studied, has also demonstrated links to medical responsibility and self-management outcomes in SB (Stern et al., 2018). It is particularly important to understand the relationship between attention/executive functioning and medical responsibility in SB, given the documented deficits and likely links across both of these areas.

**Role of Parenting Behaviors**

The successful development of medical responsibility also depends on the family environment, which includes supportive and nurturing parenting (Drotar; Ittenbach, Rohan, Gupta, Pendley, & Delamater, 2013). Specific parenting behaviors that have been examined in
relation to adolescent adjustment and autonomy outcomes among youth with spina bifida and other chronic medical conditions include parental acceptance, behavioral control, and psychological control (Butler et al., 2007; Holmbeck, Shapera, & Hommeyer, 2002). Acceptance involves being supportive, warm, validating, and understanding of a child’s experience (Steinberg, 1990). Acceptance can be expressed physically or verbally through indications of endearment, such as hugging, smiling, or praising. Behavioral control is exhibited as parenting that provides clear limits or restrictions on an adolescent’s behavior, and ensuring that the child complies with these expectations (Steinberg, 1990). Psychological control involves controlling an adolescent’s attitudes, feelings, and thoughts (Steinberg, 1990). Parents who exhibit high levels of psychological control may be perceived as intrusive, overprotective, and manipulative.

In general, adolescents tend to benefit from parents who are accepting, appropriately firm, and promoting of psychological autonomy (i.e., an authoritative parenting style; Steinberg & Silk, 2002). These behaviors translate into relatively higher levels of parental acceptance and behavioral control, and lower levels of psychological control. Parental acceptance and appropriate behavioral control have been tied to positive educational, social, mental health, and health-related outcomes in young adults with SB (Murray, Amaro, Devine, Psihogios, Murphy, & Holmbeck, 2015). However, an excessive level of behavioral control has also been related to externalizing problems among adolescents (Garber, Robinson, & Valentiner, 1997). Among both typically developing youth and individuals with SB, psychological control is consistently associated with multiple negative adjustment outcomes, including depression, anxiety, poorer academic achievement, decreased social competence, and behavior difficulties (Barber, 1996; Holmbeck et al., 2000; Holmbeck, Shaper, & Hommeyer, 2002; Murray et al., 2015).
Within pediatric populations, supportive parenting behaviors are important facilitators of healthcare behaviors (Beacham & Deatrick, 2013; Martire & Helgeson, 2017). However, this relationship is under-researched, and most literature has focused on the relationship between parenting and medical adherence. Across youth with various chronic illnesses, including type 1 diabetes, cystic fibrosis, and SB, higher levels of parental acceptance, positive reinforcement, positive parent-child relationships, and family cohesion have been associated with greater adherence and self-management behaviors (DeLambo et al., 2004; Jaser & Grey, 2010; O’Hara & Holmbeck, 2013; Psihogios et al., 2016; Stepansky et al., 2009). On the other hand, higher levels of critical, unsupportive parenting behaviors and negative family interactions have been negatively associated with self-efficacy for condition management and poorer adherence (Armstrong, Mackey, & Streisand, 2011; Duke et al., 2008; Hood, Butler, Anderson & Laffel, 2007; Jaser & Grey, 2010; Lewin et al., 2006). Moreover, while much of the research on parenting among pediatric chronic health conditions has utilized mothers, there is a dearth of research regarding father involvement (Taylor, Fredericks, Janisse, & Cousin, 2019).

Interestingly, existing evidence on fathers of children with chronic health conditions has yielded mixed findings, with some studies indicating positive effects of fathering on health outcomes (e.g., glycemic control in type 1 diabetes; Berg et al., 2008), while other studies suggesting that increased paternal involvement has negative effects on certain aspects of health (e.g., adherence; Hansen, Weissbrod, Schwartz, & Taylor, 2012). Thus, additional research is needed to explore the contributions of both mothers and fathers in pediatric chronic health conditions, including SB.
Regarding youth with SB, adaptive parenting behaviors and greater parental support are predictive of increased autonomy (Holmbeck, Coakley, Hommeyer, Shapera & Westhoven, 2002; Holmbeck, Johnson et al., 2002; Holmbeck et al., 2003). In contrast, maladaptive parenting, including excessive parental control and intrusiveness, can impede developing responsibility for medical care, which is related to negative adjustment outcomes and delays in autonomy (Antle, Montgomery, & Stapleford, 2009; Holmbeck et al., 2002; Tuminello et al., 2012; Zukerman, Devine, & Holmbeck, 2011). Further, adolescents with SB depend more on adults for completing basic self-care tasks than do typically developing peers (Friedman et al., 2009), which underscores parenting as an important modifiable factor to investigate in relation to the development of medical responsibility outcomes (Blum et al., 1991; Holmbeck et al., 2003).

**Associations among Parenting, Neuropsychological Functioning, and Self-Management**

While parenting behaviors may be related to SB self-management in their own right, they may also facilitate the development of medical responsibility through complex interactions with executive functioning and attention. Given the protracted development of attention and executive functioning, these skills are thought to be particularly sensitive to caregiver influences (Bernier, Carlson, Deschenes, Matte-Gagne, 2011). From a developmental standpoint, at younger ages, parents help children coregulate, which lays the foundation for greater self-regulation and autonomy in adolescence and young adulthood (Collins, Madsen, & Susman-Stillman, 2002). There is growing empirical support that positive parenting behaviors in early childhood are related to higher executive functioning and self-control among school-age children and adolescents, while maladaptive parenting behaviors in turn are related to poorer executive functioning skills (Berthelsen, Hayes, White, & Williams, 2017; Cuevas, Deater-Deckard, Kim-
Spoon, Watson, Morasch, & Bell, 2014; Sosic-Vasic, Kroner, Schneider, Vasic, Spitzer, & Streb, 2017). In SB, parental responsiveness has demonstrated longitudinal associations with stronger early cognitive skills and parental intrusiveness has been related to greater executive functioning deficits, indicative of the developmental and cognitive susceptibility to parenting factors (Dennis et al., 2006; Landry, Taylor, Swank, Barnes, & Juranek, 2013; Lomax-Bream et al., 2007; Tuminello et al., 2012).

Despite the limited research investigating associations among parenting, neuropsychological functioning, and medical responsibility in youth with SB, literature supports that parenting behaviors may moderate the impact of executive functioning and attention on child adjustment outcomes more broadly (Kawabata et al., 2012; Potter et al., 2011). Interestingly, one study (O’Hara & Holmbeck, 2013) did not find support for buffering effect of adaptive parenting behaviors on the relationship between executive functioning deficits and health-related autonomy among youth with SB. However, these findings may have been limited by the cross-sectional study design. Indeed, maladaptive parenting behaviors, such as parental intrusiveness, have been linked to reduced autonomy and poorer executive functioning skills among youth with SB (Tuminello et al., 2012). Authoritative parenting has been found to shape cognitive development by moderating the impact of traumatic brain injury (TBI) on youth’s executive functioning post-injury (Potter et al., 2011). Parental warmth and responsiveness also moderated the effect of TBI on ADHD symptoms and behavior problems in youth post-injury (Treble-Barna et al., 2016). Further, parenting style moderated the relationship between TBI severity and behavioral adjustment in children (Yeates et al., 2010). Additionally, maternal warmth and parental autonomy support moderated the impact of attention and executive
functioning problems on social adjustment and task perseverance among youth with ADHD (Kawabata et al., 2012; Thomassin & Suveg, 2012). Finally, familial support moderated the relationship between neuropsychological deficits and academic achievements in youth with epilepsy (Fastenau et al., 2004), providing further evidence for the importance of family context in shaping adjustment outcomes in neurodevelopmental populations.

**Gaps in the Literature**

While the importance of self-management has been identified among pediatric populations, self-management behaviors among youth with SB remain understudied (Psihogios et al., 2016; Stepansky et al., 2009). There is preliminary support that individual- and family-level factors impact medical responsibility among preadolescents and adolescents with SB (Holmbeck & Devine, 2010; O’Hara & Holmbeck, 2013; Psihogios et al., 2016). These findings need to be extended using more complex mediational and moderational models in order to explore how these factors influence each other and how they affect medical responsibility. Only two studies could be identified that investigated relations among executive functioning, parenting behaviors, and autonomy outcomes (both medical and non-medical) in youth with SB (O’Hara & Holmbeck, 2013; Tuminello et al., 2012), and both were based on the current data set. As previously noted, these studies were limited by a cross-sectional study design and lack of inclusion of attention as another potentially salient cognitive factor.

Moreover, much of the research on cognitive functioning and parenting in youth with chronic medical conditions often relies on using a subjective measure (e.g., parent-completed questionnaire) and single reporter (e.g., mother) to assess these constructs, leaving interpretations of results vulnerable to common method variance (Holmbeck, Li, Schurman, Friedman, &
Coakley, 2002). Given that autonomy represents a developmental process, longitudinal analyses are critical when examining how the transfer of medical responsibilities unfolds over time. Further, the majority of studies that have examined medical responsibility among youth with SB have treated this construct as a singular outcome, effectively averaging the level of responsibility across all possible tasks that could be part of a child’s treatment regimen. However, there is no standardized medical treatment for all individuals with SB, and regimens may include different components based on the person’s unique SB-related needs and level of functioning. Indeed, youth with SB may acquire responsibility for their various medical tasks at different ages (Castillo et al., 2017; Davis et al., 2006; Psihogios et al., 2015). To address the heterogeneity of SB regimens, research that examines responsibility for health-related tasks separately is necessary. In conclusion, the preliminary evidence linking cognitive functioning, parenting factors, and self-management outcomes is promising, but there are several gaps in the literature that need to be explored, particularly among youth with neurodevelopmental disabilities and chronic health conditions.

**Overview of Current Studies**

Given the lack of literature on attention/executive functioning, parenting behaviors, and medical responsibility, the current set of studies aimed to elucidate these relations in youth and young adults with SB using a developmentally-informed, social-ecological framework (see Figure 1 for a visual model). The first study, “A Longitudinal Study of Depressive Symptoms, Neuropsychological Functioning, and Medical Responsibility in Youth with Spina Bifida: Direct and Mediating Pathways,” published in the *Journal of Pediatric Psychology*, focuses solely on individual factors impacting medical responsibility. This study examined two competing
pathways through which attention/executive functioning deficits and depressive symptoms were associated with delays in medical responsibility over time (Figure 2). To address methodological gaps in the literature, this study used three time points and a multi-informant, multimethod approach towards assessing attention/executive functioning, depressive symptoms, and medical responsibility. Specifically, neuropsychological functioning was assessed via performance-based and questionnaire-report methods, depressive symptoms were measured using reports from mothers, fathers, youth, and their teachers, and medical responsibility was measured via parent- and youth-report. Findings supported that deficits in attention and working memory were associated with lower future medical responsibility via increased depressive symptoms (Stern et al., 2018).

Given the robust associations between cognitive factors and medical responsibility, the second and third studies focused on better understanding relations between attention/executive functioning and medical responsibility and did not include measures of youth depressive symptoms. The second study, “Longitudinal Associations Between Neuropsychological Functioning and Medical Responsibility in Youth With Spina Bifida: The Moderational Role Of Parenting Behaviors,” published in Child Neuropsychology, built off of the findings of the first study by examining how parenting behaviors moderated associations between attention/executive functioning and medical responsibility (Figure 3). Utilizing a developmental and longitudinal perspective, this study expanded upon past literature (O’Hara & Holmbeck, 2013) by investigating age as an additional moderator, as parenting behaviors were expected to more likely moderate the relationship between neuropsychological skills and child outcomes at younger, versus older, ages. Similar to the first study, this study included a longitudinal design
and a multimethod, multi-informant approach, utilizing observational methods to assess parenting behaviors. An additional strength is that this study treated maternal and paternal parenting behaviors separately, to better understand parent-specific relationships within the family context.

Finally, the third study “Executive Functioning, Attention, Parenting Behaviors, and Growth In Medical Responsibility in Youth With Spina Bifida: A Task-Specific Approach” sought to extend the first two studies and address the heterogeneous nature of SB medical regimens by examining parenting and cognitive influences on medical responsibility outcomes from a task-specific perspective (i.e., healthcare appointments, communicating SB-related needs; catheterization, bowel program, skin care, and exercise; Figure 4). This study used growth analyses to model different trajectories of development across these various SB-related medical tasks, and investigated how parenting behaviors and cognitive factors predicted these trajectories. In line with the other studies, this study addressed gaps in the literature by testing these models with longitudinal, multimethod, and multi-informant data.
Figure 1. Model Examining the Impact of Executive Functioning, Attention, and Parenting Behaviors on Medical Responsibility among Youth with Spina Bifida

- Study 1: Executive Functioning, Attention, Youth Depressive Symptoms
- Study 2: Parenting (Acceptance, Behavioral Control, Psychological Control)
- Study 3 (task-specific): Medical Responsibility
  - Healthcare appointments
  - Communicating SB needs
  - Catheterization
  - Bowel Program
  - Skin Care
  - Exercise

*Note:
- Grey = Total medical responsibility
- Yellow = Medical responsibility tasks
Figure 2. Mediational Models of Alternate Pathways among Depressive Symptoms, Neuropsychological Functioning, and Medical Responsibility

Model 1: Cognitive Scar Hypothesis

More Depressive Symptoms Time 1 → Greater Neuropsychological Dysfunction Time 2 → Less Medical Responsibility Time 3

Model 2: Cognitive Vulnerability Hypothesis

Greater Neuropsychological Dysfunction Time 1 → More Depressive Symptoms Time 2 → Less Medical Responsibility Time 3
Figure 3. Moderating Role of Parenting on the Relationship between Attention & Executive Functioning and Medical Responsibility

Neuropsychological Risk Factors (Time 1):
- Attention
- Working Memory
- Cognitive Shifting
- Planning/Organizing

Parenting Behaviors (T1):
- Acceptance
- Behavioral Control
- Psychological Control

Medical Responsibility (Time 2)

Child Age
Figure 4. Impact of Executive Functioning/Attention and Parenting on Growth in Medical Responsibility

**Neuropsychological Factors (T1):**
- Attention
- Executive Functioning

**Parenting Behaviors (T1):**
- Acceptance
- Behavioral Control
- Psychological Control

**Medical Responsibility (T1-T5):**
- Healthcare Appointments Autonomy
- Communicating about SB Autonomy
- Catheterization Autonomy
- Bowel Program Autonomy
- Skincare Autonomy
- Exercise Autonomy
CHAPTER TWO

A LONGITUDINAL STUDY OF DEPRESSIVE SYMPTOMS, NEUROPSYCHOLOGICAL FUNCTIONING, AND MEDICAL RESPONSIBILITY IN YOUTH WITH SPINA BIFIDA: DIRECT AND MEDIATING PATHWAYS

Introduction

Spina bifida (SB) is a relatively common congenital birth defect that results from failure of the neural tube to close during embryonic development (Mahmood, Dicianno, & Bellin, 2011). SB is a heterogeneous condition, with the spinal lesion level affecting condition severity and individual functioning across several domains, including motor and orthopedic difficulties, bladder and bowel dysfunction, and neurological complications (e.g., Chiari II malformation, hydrocephalus, and epilepsy; Copp et al., 2015). In addition, individuals with SB are at risk for secondary health complications such as obesity, urinary tract infections, pressure sores, and shunt infections/malfunctions (Copp et al., 2015).

Youth with SB must adhere to a lifelong, daily medical regimen (Copp et al., 2015; O’Hara & Holmbeck, 2013), and the transition of responsibility for managing this medical regimen from parents to youth has become a critical component of development (Beacham & Deatrick, 2013). Although many youth with SB have an interest in becoming autonomous with respect to their medical responsibilities (e.g., bladder and bowel programs, skin checks; Holmbeck & Devine, 2010), individuals with SB often exhibit developmental delays in self-help skills, resulting in lower levels or a delay in the acquisition of independent functioning (Andren
Additionally, independence in managing medical responsibilities relies on physical (e.g., strength, dexterity), cognitive (e.g., executive functioning), and psychosocial (e.g., emotional maturity; Beacham & Deatrick, 2013; Modi et al., 2012) abilities, all of which may pose significant challenges for youth with SB. Despite these challenges, longitudinal findings support a developmental trajectory where the majority of youth with SB gradually gain responsibility for medical tasks, such as catheterization and bowel program management, over time (Psihogios, Kolbuck, & Holmbeck, 2015; Stepansky, Roache, Holmbeck, & Schultz, 2010). Given that increased responsibility for one’s medical regimen allows youth with a chronic medical condition to advance developmentally (e.g., an increase in time spent with peers), it is important to understand processes that influence the attainment of medical responsibility in youth with SB.

Very few studies have been conducted to isolate modifiable risk factors that are associated with medical responsibility in youth with SB. One potentially important modifiable, individual factor is depressive symptomology. Depressive symptoms were found to be associated with decreased competency in completing self-management activities in adults with SB (Bellin et al., 2010). Although research has shown that youth with SB, especially adolescents, are at a significantly greater risk for developing depressive symptoms compared to healthy peers (Appleton et al. 1997; Holmbeck et al., 2003), the relationship between depressive symptoms and attainment of medical responsibility has yet to be studied in youth with SB. It is possible that depressive symptoms compromise medical responsibility by decreasing youth’s decision-making abilities and attention, which are required to complete health-care related tasks on a daily basis (Modi et al., 2012). In other illness populations (e.g., type 1 diabetes), youth depressive
symptoms have been associated with a decrease in motivation to complete medical tasks (Guo et al., 2013) as well as increased parent responsibility for medical tasks (Helgeson, Reynolds, Siminerio, Escobar, & Becker, 2008). Therefore, the processes through which depression may influence responsibility for medical care for youth with SB should be examined.

One possible mechanism is that depressive symptoms may disrupt neuropsychological functioning, leading to persistent cognitive deficits (the cognitive scarring model; Allott, Fisher, Amminger, Goodall, & Hetrick, 2016). Studies with otherwise healthy adolescents have found associations between the experience of acute depressive symptoms and executive functioning, memory, and attentional impairments (Wilkinson & Goodyer, 2006). Youth with SB are susceptible to neuropsychological impairments due to neurological factors (e.g., presence of hydrocephalus, Chiari II malformation, and shunt complication; Copp et al., 2015). Specifically, they experience difficulties with attention and executive functioning (e.g., problem-solving, initiation, working memory, planning, organization, and self-monitoring), and these pre-existing deficits may be exacerbated by depressive symptoms. Such deficits could affect the higher order cognitive skills needed to attain autonomy in completing medical tasks.

An alternate hypothesis is that the difficulties with executive functioning and attention experienced by youth with SB are primarily responsible for decreased medical responsibility. Executive dysfunction has been predictive of lower levels of medical responsibility for youth with SB (Psihogios et al., 2016). However, it is possible that neuropsychological difficulties reduce these youth’s ability to cope and problem solve when confronted with stressors and, as a consequence, make them more susceptible to depressive symptoms (the cognitive vulnerability model; Lee, Hermens, Porter, & Redoblado-Hodge, 2012). In other words, depressive symptoms
may mediate the relationship between neuropsychological deficits and decreased attainment of medical autonomy for youth with SB. In fact, deficits in executive functioning and attention have been found to put individuals with SB at risk for the development of future depressive symptoms (Lennon, Klages, Amaro, Murray, & Holmbeck, 2015). Thus, it is also possible that neuropsychological impairment hinders the development of medical autonomy in youth with SB via increased depressive symptoms.

Despite our knowledge that both depressive symptoms and neuropsychological functioning are related to the development of medical autonomy (and each other), few studies to date have examined the interrelationships of these variables in youth with SB (Donlau et al., 2011; O’Hara & Holmbeck, 2013; Psihogios et al., 2016). Therefore, the current study examined relations between depressive symptoms, attention/executive functioning, and medical autonomy in youth with SB. Specifically, this study explored two potential pathways to delays in medical autonomy: 1) depressive symptoms as predictors of medical autonomy as mediated by attention/executive functioning (the cognitive scarring model; Figure 5, Model 1), and 2) attention/executive functioning as predictors of medical autonomy as mediated by depressive symptoms (the cognitive vulnerability model; Figure 5, Model 2). It was hypothesized that greater depressive symptoms would be associated with worse neuropsychological functioning which, in turn, would predict lower levels of medical autonomy. With respect to the alternate pathway, it was hypothesized that poorer neuropsychological functioning would be associated with greater depressive symptoms which, in turn, would predict lower levels of medical autonomy. Additionally, the current study sought to address gaps in the literature by testing these models with longitudinal, multimethod, and multi-informant data.
Figure 5. Mediational Models of Alternate Pathways among Depressive Symptoms, Neuropsychological Functioning, and Medical Responsibility

**Model 1: Cognitive Scar Hypothesis**

- More Depressive Symptoms
- Greater Neuropsychological Dysfunction Time 2
- Less Medical Responsibility Time 3

**Model 2: Cognitive Vulnerability Hypothesis**

- Greater Neuropsychological Dysfunction Time 1
- More Depressive Symptoms Time 2
- Less Medical Responsibility Time 3
Methods

Participants

Participants were recruited for an ongoing, larger longitudinal study examining family, neuropsychological, and psychological functioning among children and adolescents with SB (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). Families of youth with SB 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, and 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-based data collections).

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 SB (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 did not include participants who functioned intellectually at two or more standard
deviations below the population mean (i.e. 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. Therefore, the final sample used in the analyses included 114 children and adolescents with spina bifida (52.63% female; M_{age} = 10.96 (SD = 2.43); 51.75% Caucasian, 11.40% African American, 17.54% Hispanic, 5.26% Other; Table 1).

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 either Time 2 or Time 3 (n = 38, 33%) did not differ significantly from youth who participated at all three data collection waves with respect to gender, socioeconomic status, type of SB, lesion level, shunt status, or IQ. However, youth who did not participate at either Times 2 or 3 were significantly older at Time 1 \([M = 11.74\text{ compared to } 10.61; t(106) = -2.28, p = .03\).}
Table 1. Youth Demographic and Spina Bifida Information at Time 1

<table>
<thead>
<tr>
<th></th>
<th>Youth (N=114)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>M (SD) or N (%)</td>
</tr>
<tr>
<td>Gender: female</td>
<td>60 (52.63%)</td>
</tr>
<tr>
<td>Age</td>
<td>10.96 (2.43)</td>
</tr>
<tr>
<td>Race</td>
<td></td>
</tr>
<tr>
<td>Caucasian</td>
<td>59 (51.75%)</td>
</tr>
<tr>
<td>African-American/Black</td>
<td>13 (11.40%)</td>
</tr>
<tr>
<td>Hispanic/Latino</td>
<td>20 (17.54%)</td>
</tr>
<tr>
<td>Other</td>
<td>6 (5.26%)</td>
</tr>
<tr>
<td>Family SES</td>
<td>42.32 (14.99)</td>
</tr>
<tr>
<td>IQ</td>
<td>92.41 (15.67)</td>
</tr>
<tr>
<td>Spina bifida type</td>
<td></td>
</tr>
<tr>
<td>Myelomeningocele</td>
<td>85 (74.56%)</td>
</tr>
<tr>
<td>Lipomeningocele</td>
<td>9 (7.89%)</td>
</tr>
<tr>
<td>Not Sure/Not reported</td>
<td>13 (4.40%)</td>
</tr>
<tr>
<td>Lesion level</td>
<td></td>
</tr>
<tr>
<td>Thoracic</td>
<td>11 (9.65%)</td>
</tr>
<tr>
<td>Lumbar</td>
<td>74 (64.9%)</td>
</tr>
<tr>
<td>Sacral</td>
<td>23 (20.18%)</td>
</tr>
<tr>
<td>Unknown/not reported</td>
<td>6 (5.26%)</td>
</tr>
<tr>
<td>Shunt: present</td>
<td>73 (64.04%)</td>
</tr>
</tbody>
</table>
Procedure

This study was approved by university and hospital Institutional Review Boards. Trained undergraduate and graduate student research assistants collected data from families during two separate three-hour home visits at Time 1, and one three-hour home visit at both Time 2 and Time 3. Informed consent from parents and assent from youth were obtained prior to the start of the first visit. Parents also filled out releases of information to permit data collection from medical charts, health professionals, and teachers. During data collection, youth and their parents completed questionnaires independently. The questionnaires were offered in both English and Spanish; questionnaires that were only available in English were adapted for Spanish speakers by a translation team using back translation procedures. Additionally, research assistants completed a brief neuropsychological battery with the child. Families received monetary compensation of $150 and small gifts (e.g., logo t-shirts, pens, water bottles) for participating.

Measures

Demographics. Parents reported on youth and family demographic information through questionnaires at Time 1, including age, gender, race, and ethnicity. 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).

Youth IQ. At Time 1, youth were administered the Vocabulary and Matrix Reasoning subtests of the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999) which were used to estimate a Full Scale IQ score. These subtests have demonstrated high levels of internal consistency for youth 6-16 years old (α = .89 for Vocabulary, α = .92 for Matrix Reasoning; Wechsler, 1999).
**Depressive Symptoms.** Depressive symptoms were measured via child-, parent-, and teacher-report. Children completed the Child Depression 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, which demonstrated acceptable levels of internal consistency at both Time 1 and Time 2 (α=.82; α=.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 assess behavioral and emotional problems over the past six and two months, respectively. For this study, a subscale of depressive symptoms was derived based on 15 items from in the Anxious/Depressed and Withdrawn/Depressed subscales to form a CBCL-Depression Scale (CBCL-D; Clarke, Lewinsohn, Hops, & Seeley, 1992). As this adapted scale has not been normed, raw mean total scores were calculated in lieu of T-scores, which demonstrated adequate internal consistency at Time 1 and Time 2 for mother (α=.74; α=.64), father (α=.69; α=.71), and teacher report (α=.78; α=.84).

**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. The following areas of neuropsychological functioning were examined: 1) attention, 2) working memory, 3) planning and organizational skills.

**Attention.** Parents and teachers completed the Swanson, Nolan, and Pelham Teacher and Parent Rating Scale (SNAP-IV; Swanson, 1992). The SNAP-IV is comprised of 18 items derived from criteria for Attention-Deficit/ Hyperactivity Disorder from the DSM-IV (American Psychiatric Association, 1994). Mean subscale scores were calculated for the inattention subscale, which demonstrated high internal consistency at Time 1 and Time 2 for mother (α=.93;
Parents and teachers also completed the Attention Problems subscale of the CBCL and TRF, respectively (Achenbach & Rescorla, 2001). This subscale demonstrated adequate levels of internal consistency in the current study ($\alpha=.73-.82$). At Time 1 and Time 2, youth were administered a performance-based measure of attention, the Number Detection (ND) Subtest of the Cognitive Assessment System (CAS; Naglieri & Das, 1997). Internal consistency reliability ($\alpha = .77$) and test-retest reliability ($r = .77$) for the ND subtest are high across age groups (Naglieri & Das, 1997).

**Working Memory.** Parents and teachers completed the Working Memory subscale of the Behavior Rating Inventory of Executive Functioning (BRIEF, Gioia et al., 2000). This is a valid measure of multiple domains of executive functioning, including working memory, over the past six months (Gioia et al., 2000a, 2000b). For the BRIEF, higher scores indicate greater impairment. The Working Memory subscale demonstrated high internal consistency at Time 1 and Time 2 for mother ($\alpha=.90; \alpha=.91$), father ($\alpha=.90; \alpha=.89$), and teacher report ($\alpha=.91; \alpha=.92$). Youth were administered the Digit Span subtest of the Wechsler Intelligence Scale for Children (WISC-IV; Wechsler, 2003) as a performance-based measure of working memory ability. The Digit Span subtest has good internal consistency ($r = .87$) and test-retest reliability ($r = .83$; Williams, Weiss, & Rolfhus, 2003).

**Planning and Organizational Skills.** 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 the ability to organize one’s thoughts and to plan one’s actions to achieve present and future goals. This subscale demonstrated high internal consistency at Time 1 and Time 2 for mother ($\alpha=.92; \alpha=.92$), father ($\alpha=.90; \alpha=.87$), and teacher report ($\alpha=.91$;
α=.92). The Organization of Materials subscales measures the child’s tendency to keep his or her spaces neat and orderly. High internal consistency was found at Time 1 and Time 2 for mother (α=.88; α=.86), father (α=.88; α=.84), and teacher report (α=.79; α=.78). For a performance-based measure of planning skills, youth were administered the Planned Connections (PCn) subtest of the Cognitive Assessment System (CAS). The PCn subtest has high internal consistency (α = .77) and test-retest reliability (r = .73) (Naglieri & Das, 1997).

**Medical Responsibility.** 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 SB responsibilities and health-related tasks within the family (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 task item, a score of “1” indicates the parent is primarily responsible, “2” indicates responsibility is shared equally between the parent and child, and “3” indicates the child was primarily responsible. Mean scores were calculated for the total responsibility scale. Items that participants rated as “not applicable” were excluded from the total scale score. Previous studies have not included 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 (e.g., Psihogios, Kolbuck, & Holmbeck, 2015).

The mean scores of the CDI, CBCL and SNAP-IV at Time 1 and Time 2 fell within the average range relative to the normative data samples. Across participant gender and age, BRIEF Working Memory subscale mean t-scores fell between 54-61 for parent-report and 47-60 for
teacher-report, Plan/Organize subscale mean t-scores fell between 53-61 for parent-report and 59-73 for teacher-report, and Organization subscale mean t-scores fell between 51-55 for parent-report and 47-59 for teacher-report. It should be noted that these mean scores may be an overestimation of the overall study sample's executive functioning abilities, as lower functioning individuals were excluded from analyses. At Times 1 and 2, respectively, twenty-four percent and eighteen percent of participants were reported to have borderline or clinically significant attention problems via the CBCL. Mean performance on both CAS subtests fell in the low average range at Time 1 and Time 2 (Mean Scaled Scores = 6-7) relative to the normative data. Finally, mean performance on the Digit Span subtest was average at both Time 1 (M = 8.06; SD = 2.88) and Time 2 M = 8.64, SD = 2.96).

**Statistical Treatment**

All analyses included the following covariates: child lesion level, age, SES, and target variables at previous waves of data collection. Given the concerns about statistical overcorrection and the contention that IQ should not be controlled for in examinations of specific cognitive processes in neurodevelopmental disorders, IQ was not included as a covariate in this study (Dennis et al., 2009). To decrease the number of analyses and reduce the possibility of shared method variance, composite scores were created when possible that included multiple reporters and/or measures (Holmbeck et al., 2002). Composite scores were created if they met the following criteria: Pearson correlation coefficients were run to assess for adequate associations ($r \geq 0.40$) between two reporters and/or measures and Cronbach alphas were computed to assess for adequate internal consistency ($\alpha > 0.60$) among three or more reporters and/or measures.
Two meditational models were tested using Preacher and Hayes’ (2008) bootstrapping methods. The cognitive scarring model examined the impact of youth depressive symptoms at Time 1 on SB medical autonomy at Time 3, as mediated by neuropsychological functioning (i.e., attention, working memory, and planning/organizing ability) at Time 2. The cognitive vulnerability model examined the impact of neuropsychological deficits at Time 1 on SB medical autonomy at Time 3, as mediated by depressive symptoms at Time 2. Bootstrapping has been validated in the literature and is preferred over other methods, as bootstrapping is less conservative and reduces the possibility of Type II errors (Preacher & Hayes, 2008).

Results

Preliminary Analyses

Means, standard deviations, and bivariate correlations among study variables are displayed in Table 2. Mother-report, father-report, teacher-report, and performance-based assessment of youth attention, working memory, and planning/organizing abilities were aggregated to form global composite variables. Medical responsibility data were also aggregated across parent and youth reports. While mother- and father-report of youth depressive symptoms could be combined across reporters, they were not adequately correlated with self- or teacher-report of youth depressive symptoms. Thus, self-, parent-, and teacher-report of youth depressive symptoms were examined separately in the analyses.

Listwise deletion was used to handle missing data. Sample sizes for models with child-, parent-, and teacher-reported depressive symptoms at Time 1 were 67, 70, and 65, and at Time 2 were 67, 68, and 56, respectively. Missing data were due to attrition across time points. Further, while composites for parent-reported variables could accommodate missing data across time
points from either mothers or fathers, fewer teachers participated at each time point. Assuming a power of .80, and an alpha of .05, a sample size of 78 is required to detect medium effect sizes and a sample size of 36 is required to detect large effect sizes (Fritz & MacKinnon, 2007). Thus, the current study had enough power to detect effects between medium and large.
Table 2. Correlations among Depressive Symptoms, Neuropsychological Variables, Medical Responsibility Variables, and Covariates

<table>
<thead>
<tr>
<th>Variable</th>
<th>1.</th>
<th>2.</th>
<th>3.</th>
<th>4.</th>
<th>5.</th>
<th>6.</th>
<th>7.</th>
<th>8.</th>
<th>9.</th>
<th>10.</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. CDI</td>
<td>–</td>
<td>.12</td>
<td>.14</td>
<td>.17</td>
<td>.31**</td>
<td>.27*</td>
<td>.15</td>
<td>-0.3</td>
<td>-18</td>
<td>-16</td>
</tr>
<tr>
<td>2. CBCL-P – Depression(^a)</td>
<td>.24*</td>
<td>–</td>
<td>-0.03</td>
<td>.30**</td>
<td>.12</td>
<td>.23*</td>
<td>-0.09</td>
<td>.03</td>
<td>.15</td>
<td>-0.02</td>
</tr>
<tr>
<td>3. TRF – Depression(^a)</td>
<td>.09</td>
<td>.30*</td>
<td>–</td>
<td>.51**</td>
<td>.50**</td>
<td>.42**</td>
<td>-0.40**</td>
<td>-16</td>
<td>-0.09</td>
<td>.01</td>
</tr>
<tr>
<td>4. Attention(^a)</td>
<td>-0.05</td>
<td>.28**</td>
<td>.52**</td>
<td>–</td>
<td>.80**</td>
<td>.82**</td>
<td>-0.41**</td>
<td>-17</td>
<td>.08</td>
<td>.03</td>
</tr>
<tr>
<td>5. Working Memory(^a)</td>
<td>.08</td>
<td>.21*</td>
<td>.48**</td>
<td>.84**</td>
<td>–</td>
<td>.79**</td>
<td>-0.38**</td>
<td>-0.28**</td>
<td>-11</td>
<td>-13</td>
</tr>
<tr>
<td>6. Plan/Organizing(^a)</td>
<td>.01</td>
<td>.30**</td>
<td>.44**</td>
<td>.78**</td>
<td>.76**</td>
<td>–</td>
<td>-0.24**</td>
<td>-0.28**</td>
<td>.02</td>
<td>-0.09</td>
</tr>
<tr>
<td>7. Med. Responsibility</td>
<td>.04</td>
<td>-0.15</td>
<td>-0.28*</td>
<td>-0.33**</td>
<td>-0.26*</td>
<td>-0.19</td>
<td>–</td>
<td>.54**</td>
<td>-0.03</td>
<td>-0.15</td>
</tr>
<tr>
<td>8. Age(^b)</td>
<td>.17</td>
<td>-0.02</td>
<td>-0.02</td>
<td>-0.09</td>
<td>-0.12</td>
<td>-0.06</td>
<td>.54**</td>
<td>–</td>
<td>.05</td>
<td>.01</td>
</tr>
<tr>
<td>9. SES(^b)</td>
<td>-0.12</td>
<td>.21</td>
<td>.02</td>
<td>-0.07</td>
<td>-0.08</td>
<td>.04</td>
<td>-0.03</td>
<td>.05</td>
<td>–</td>
<td>.03</td>
</tr>
<tr>
<td>10. Lesion Level(^b)</td>
<td>-0.01</td>
<td>.03</td>
<td>.16</td>
<td>-0.07</td>
<td>-0.09</td>
<td>-0.07</td>
<td>-0.15</td>
<td>.01</td>
<td>.03</td>
<td>–</td>
</tr>
<tr>
<td>M (SD) T1 Dep., T2 Neuro.</td>
<td>1.1(.1)</td>
<td>-0.03(.1)</td>
<td>-0.08(.9)</td>
<td>-0.01(.8)</td>
<td>-0.01(.7)</td>
<td>.01(.7)</td>
<td>2.2(4)</td>
<td>10.9(2.4)</td>
<td>42.3(15)</td>
<td>--</td>
</tr>
<tr>
<td>M (SD) T2 Dep., T1 Neuro.</td>
<td>1.2(.2)</td>
<td>.00(.9)</td>
<td>.00(1.0)</td>
<td>-0.01(.8)</td>
<td>.02(.7)</td>
<td>-0.02(.7)</td>
<td>--</td>
<td>--</td>
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</tr>
</tbody>
</table>

Notes. Values above the diagonal reflect variables from the first model (i.e., Time 1 depressive symptoms, Time 2 neuropsychological factors). Values below the diagonal reflect variables from the alternate model (i.e., Time 1 neuropsychological factors, Time 2 depressive symptoms). 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. All cognitive variables were scored such that higher scores represent greater neuropsychological deficits in attention, working memory, and planning/organizing abilities; *These variables are based on standardized Z scores. \(^a\)These variables are covariates. Descriptive statistics for lesion level are presented in Table 1. *p < .05, **p < .01.
**Mediation Analyses**

Mediation analyses were conducted to examine the indirect effects of neuropsychological deficits and depressive symptoms on medical responsibility. Time 1 mediators and Time 2 medical responsibility scores were also entered as covariates. To maximize sample size and investigate differential relationships among depressive symptoms and individual cognitive deficits, each model was tested separately with the three neuropsychological factors (i.e., attention, working memory, and planning/organizing) and self-, parent-, and teacher-report of child depressive symptoms, for a total of nine models.

**Model 1 (Cognitive Scar Hypothesis).** The first objective of this study was to examine if neuropsychological functioning mediated the impact of child depressive symptoms on medical responsibility in youth with SB longitudinally. Results indicated no significant indirect effects (all $p$’s $>$ .05). When attention was examined as a mediator, there was a significant direct, positive effect of parent-reported child depressive symptoms at Time 1 on child medical responsibility 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 likely indicates statistical suppression; as a result, this finding will be regarded as a statistical artifact and will not be interpreted further (Pandey & Elliott, 2010). In the model using self-reported child 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\(^1\).

\(^1\) Consistent with reported results, mediation models using maximum likelihood estimation in MPlus identified a direct effect of parent-reported child depressive symptoms on medical responsibility in the model with attention as a mediator ($B=.06$, $SE=.03$, 95% LLCI to ULCI = .004 to .11). An additional negative direct effect of teacher-reported
Model 2 (Cognitive Vulnerability Hypothesis). The second model examined if child depressive symptoms mediated the longitudinal impact of neuropsychological functioning on medical responsibility in youth with SB. The results are presented in Figures 6-8. 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). The indirect effect of planning/organizing abilities on medical responsibility 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/organizing skills when adjusting for depressive symptoms was greater than the total effect, results likely indicated statistical suppression (MacKinnon, Krull & Lockwood, 2000). Therefore, this finding will be regarded as a statistical artifact².

² Consistent with reported results, mediation models using maximum likelihood estimation in MPlus identified a significant indirect effect of teacher-reported child depressive symptoms on the relations between attention and medical responsibility (B=.04, SE=.02, 95% LLCI to ULCI = .09 to .01) and working memory and medical responsibility (B=.03, SE=.02, 95% LLCI to ULCI = .09 to .004). Also consistent with reported results, there was a significant indirect effect of teacher-reported child depressive symptoms on the relation between planning and organizing skills and medical responsibility (B=.05, SE=.03, 95% LLCI to ULCI = .13 to .01), which was best explained by statistical suppression since the direct effect (B=.05, SE=.05) was greater than the total effect (B=.00, SE=.05). Significant total and direct effects emerged for the relations between parent-reported child depressive symptoms and both attention (B=.08, SE=.03, total effect 95% LLCI to ULCI = .14 to .01; B=.08, SE=.03, direct effect 95% LLCI to ULCI = .15 to .02) and working memory (B=.07, SE=.03, total effect 95% LLCI to ULCI = .13 to .002; B=.07, SE=.03, direct effect 95% LLCI to ULCI = .14 to .003).
Figures 6-8. Mediation Models of Child Neuropsychological Functioning at Time 1, Depressive Symptoms at Time 2, and Medical Responsibility at Time 3.

**Figure 6.** Notes. a) Direct effect of attention on medical responsibility in model controlling for parent-reported depressive symptoms as a mediator; b) Indirect effect of attention on medical responsibility through teacher-reported depressive symptoms. Neither the total effect nor the direct effect was significant for the model controlling for teacher-reported depressive symptoms as a mediator. *p<.05; **p<.01.

**Figure 7.** Notes. a) Indirect effect of working memory on medical responsibility through teacher-reported depressive symptoms. Neither the total effect nor the direct effect was significant for the model with a significant indirect effect. *p<.05.

**Figure 8.** Notes. a) Indirect effect of planning/organizing on medical responsibility through teacher-reported depressive symptoms. Neither the total effect nor the direct effect was significant for the model with a significant indirect effect. *p<.05.

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3 For Figures 6-8, analyses were tested separately for each of the three mediators and three independent variables. In all models, attention, working memory, and planning/organizing represent global, composite factors.
Discussion

The current study examined depressive symptoms and neurocognitive deficits in relation to medical responsibility over time in youth with (SB). To clarify the ambiguous relationship between depressive symptoms and neurocognitive deficits, two mediation pathways were tested. In the first pathway (i.e., the cognitive scarring model), neurocognitive deficits were expected to mediate the relationship between depressive symptoms and medical responsibility, such that more severe depressive symptoms would predict greater deficits in attention and executive functioning, and reduced cognitive abilities would predict lower medical responsibility. In the second pathway (i.e., the cognitive vulnerability model), depressive symptoms were expected to mediate the relationship between neurocognitive deficits and medical responsibility, such that more profound cognitive deficits would predict greater depressive symptoms, which would in turn predict lower levels of medical responsibility. This study found support for the latter model, in that deficits in attention and working memory were associated with medical responsibility via increased depressive symptoms. As medical autonomy constitutes a key developmental goal for many youth with SB (Holmbeck & Devine, 2010), it is important to understand how relevant cognitive and psychological factors, together, play a role in this gradually unfolding process.

Results indicated that the hypotheses were partially supported, and clarify the directional relationships among these individual factors in youth with SB. By demonstrating that greater deficits in attention and working memory were associated with less medical responsibility, these findings align with previous research and provide further support for the bio-neuropsychosocial model of medical autonomy and adherence in youth with SB (Holmbeck & Devine, 2010; Psihogios et al., 2016; Tuminello, Holmbeck, Olson, 2012). Although poor psychological
adjustment in adolescents with other chronic illnesses has been found to complicate the transition of health care responsibilities (Reed-Knight, Blount, & Gilleland, 2014), depressive symptoms have not been examined as a predictor of medical responsibility in youth with SB. Thus, the finding from the current study that youth with more severe depressive symptoms struggled to develop independence in their medical care represents a unique contribution to the literature.

Further, mediation results suggest that one way in which certain neurocognitive deficits may hinder the development of medical responsibility in youth with SB prospectively is through an increased risk for experiencing depressive symptoms. From a clinical perspective, it is possible that youth with poor attention and working memory have difficulty following instructions, completing multi-step tasks, and planning for long-term goals (Kelly et al., 2012). This difficulty 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 achieving higher levels of medical responsibility. Thus, when conceptualizing the development of medical responsibility in SB, it is important to consider not only the neurocognitive impairments associated with SB, but also how these deficits may lead to increased depressive symptoms.

As neuropsychological functioning did not mediate the relationship between depressive symptoms and medical responsibility, the exact mechanism through which depressive symptoms may influence future medical responsibility remains unclear. Given the developmental stage of the participants in this study, it is also plausible that family or peer factors may better explain the relationship between depressive symptoms and medical responsibility. Indeed, peer conflict in adolescents has recently been identified as a barrier to medical responsibility in youth with SB.
(Psihogios et al., 2016). Other individual factors, such as lowered intrinsic motivation or self-efficacy in managing one’s medical condition, may explain this relationship as well. Moreover, executive functioning and attentional skills are not fully developed until the mid-twenties, and youth with SB continue to experience delays in the growth of these abilities through adolescence and emerging adulthood (Tarazi, Zabel, & Mahone, 2008). Given these differing developmental trajectories, it is possible that relations between cognitive deficits and depressive symptoms in SB change over time.

Interestingly, main effects in the mediation model were only found for teacher-report of youth depressive symptoms, but not self- or parent-report. Lennon, Klages, Amaro, Murray, & Holmbeck (2015) similarly found that neuropsychological functioning predicted teacher-report, but not self- or parent-report of youth internalizing symptoms. It is speculated that teachers may be more objective reporters of depressive symptoms in youth with SB than parents, as they are more readily able to compare a child with SB to other typically developing, same-aged peers (Lennon et al., 2015). On the other hand, it is possible that teachers who are unfamiliar with SB may misinterpret certain cognitive and behavioral features of SB (e.g., poor initiation, amotivation) as symptoms of depression. However, teachers may also be at a unique advantage as they are more likely to observe depressive symptoms that have emerged due to cognitive challenges because they observe the child daily in a school setting. Additionally, the cognitive deficits present in youth with SB may impair their ability to accurately report on their own depressive symptoms (Wasserman, Holmbeck, Lennon, & Amaro, 2012). Future research should explore the different perceptions of depressive symptoms in SB based on reporter and environment.
**Strengths and Limitations.** This study had several strengths, including the utilization of multiple methods and reporters, performance- and questionnaire-based assessments of executive and attentional functioning, and a longitudinal, mediational design. However, there are several limitations that should be addressed in future work. As cognitive deficits are a direct consequence of SB itself, the unique relationships among neuropsychological factors, depressive symptoms, and medical responsibility may not generalize to youth with chronic illnesses that do not congenitally impact the central nervous system. While a strength of this study was its multi-method assessment of cognitive variables, only three domains of executive functioning were assessed with both performance and questionnaire measures. Future research should examine how other executive functions (e.g., inhibition, cognitive flexibility) relate to depressive symptoms and medical responsibility in SB, as these skills have been implicated in both the broader depression and self-management literatures (Bagner, Williams, Geffken, Silverstein, & Storch, 2007). Other limitations to consider include a small sample size that limited the potential to identify small mediation effects, and a relatively wide age range. Additionally, some individual subscales had relatively low internal consistency scores (e.g., the CBCL-D scale).

Finally, while this study aimed to investigate two pathways in depth, it did not examine other potentially important factors related to the medical responsibility process, 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 responsibility in youth with SB (Psighogios et al., 2016). To date, no studies have examined the influence of community or macro-level (e.g., health care system) factors on SB 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 medical responsibility over time in SB.

**Conclusion.** The results of the current study have important implications for promoting medical responsibility in youth with SB. 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 SB, it appears that depressive symptoms, attention, and executive functioning are intertwined and have a unique impact on medical responsibility in this population. Second, depressive symptoms appear to be one pathway through which attention and executive impairment may hinder medical responsibility. This key finding paves the way for further research on other pathways that may mediate the impact of neuropsychological functioning on medical responsibility in SB. Further, given the increased prevalence of depressive symptoms in youth with SB (Holmbeck et al., 2003), this study serves as a guide for research on other factors that may explain the relationship between depressive symptoms and medical responsibility (e.g., intrinsic motivation, self-efficacy).

**Clinical Implications.** 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. 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 adolescents with SB. As families begin the transfer process, providers may also choose to incorporate specialized cognitive training programs
(Stubberud, Langenbahn, Levine, Stanghelle, & Schanke, 2014) or assistive technologies for executive weaknesses (e.g., visual schedules) to support an adolescent with SB who is struggling in these areas.
CHAPTER THREE
LONGITUDINAL ASSOCIATIONS BETWEEN NEUROPSYCHOLOGICAL FUNCTIONING AND MEDICAL RESPONSIBILITY IN YOUTH WITH SPINA BIFIDA: THE MODERATIONAL ROLE OF PARENTING BEHAVIORS

Introduction

Spina bifida (SB) is a congenital birth defect that results when the neural tube fails to fully close in the first trimester of pregnancy. SB is a heterogeneous neurodevelopmental disorder which, depending on disease severity (e.g., lesion level, gross motor functioning, shunt status), can result in a constellation of various orthopedic, urinary, bowel, and neurological difficulties. Given the complex cognitive, physical, and medical needs, the demands of SB require the family to follow a multi-step medical regimen, which may include medication management, clean intermittent catheterization, maintaining a bowel program, monitoring for shunt infections or obstructions, checking for pressure injuries, and coordinating medical appointments.

Among pediatric populations, self-management of one’s medical regimen is an essential component of autonomy development. Self-management is conceptualized as a multifaceted, overarching construct that encompasses medical adherence, medical responsibility, self-care skills, and condition knowledge. Understanding how medical responsibilities are transferred from parents to youth is critical, as the successful transfer of these tasks is considered a developmental milestone and is associated with other salient developmental goals including
greater functional independence, effective transition to adult health care, gaining employment, obtaining higher education, and maintaining romantic relationships (Friedman, Holmbeck, DeLucia, Jandasek, & Zebracki, 2009; Warschausky, Kaufman, Evitts, Schutt, & Hurvitz, 2017). However, due to their physical and cognitive challenges, youth with SB are at risk for reduced autonomy across multiple functional domains as compared to typically developing youth (Davis, Shurtleff, Walker, Seidel, & Duguay, 2006).

Existing models of adjustment and self-management among youth with SB and other chronic health conditions utilize bioneuropsychosocial and social-ecological approaches, and point to individual, family (e.g., family conflict), and systemic factors (e.g., access to health care resources) as related to self-management outcomes (Modi et al., 2012; Psihogios, Murray, Zebracki, Acevedo, & Holmbeck, 2016; Reed-Knight, Blount, Gilleland, 2014). Neuropsychological functioning, particularly more highly developed executive functioning and attentional skills, has demonstrated robust, positive associations with self-management outcomes, including medical responsibility (Heffelfinger et al., 2008; O’Hara & Holmbeck, 2013; Psihogios et al., 2016; Stern, Driscoll, Ohanian, & Holmbeck, 2018; Warschausky et al., 2017). Within the context of a chronic health condition, executive functioning/attention skills are used by the individual to self-regulate emotions and behaviors, plan, organize, problem-solve, and coordinate in order to accomplish goals, including the tasks required to manage their illness (Berg et al., 2017). These factors are particularly important to investigate in SB because youth with SB often demonstrate executive functioning and attentional deficits (Rose & Holmbeck, 2007). In fact, prior research has found strong direct effects of attention and executive
functioning skills on medical responsibility above and beyond other salient individual factors, such as depressive symptoms (Stern et al., 2018).

Parenting behaviors have also been linked to self-management outcomes in chronic health populations (Lindsay, Kingsnorth, & Hamdani, 2011). Research has identified dimensions of parenting behaviors that are salient for child adjustment and autonomy outcomes, such as acceptance, behavioral control, and psychological control, which will be examined in this study (Zimmer-Gembeck & Collins, 2008). Parental acceptance (i.e., nurturance, warmth, affection, emotional support) and behavioral control (i.e., high parental demandingness of appropriate behavior, enforcement of behavioral compliance), are seen as adaptive parenting behaviors and are associated with more positive child adjustment outcomes (Steinberg & Silk, 2002). In contrast, parental psychological control, which is characterized by the use of manipulative or intrusive practices to control a child’s behavior, is viewed as maladaptive and is thought to have adverse consequences for youth psychosocial adjustment, including undermining a child’s growing autonomy (Steinberg & Silk, 2002).

More generally, supportive parenting behaviors (characterized by sensitivity, acceptance, appropriate parental monitoring, and adequate limit setting) can facilitate the transfer of medical responsibilities to youth, while maladaptive parenting (i.e., harsh, intrusive, and unsupportive) may impede developing responsibility (Holmbeck, Johnson, et al., 2002; Tuminello, Holmbeck, Olson, 2012). Youth with SB depend more on adults for completing self-management tasks than do typically developing peers (Friedman et al., 2009), which underscores parenting as an important modifiable factor to investigate in relation to the development of medical responsibility (Holmbeck et al., 2003). Additionally, prior literature has pointed to the
importance of examining maternal and paternal parenting behaviors separately, as mothering and fathering behaviors not only differ from each other, but can have unique effects on youth adjustment outcomes (Lansford, Laird, Pettit, Bates, & Dodge, 2014).

Although there is limited research regarding relations among parenting, executive functioning, and self-management in youth with SB, evidence suggests that parenting behaviors may moderate the impact of attention and executive functioning on adjustment outcomes in youth more broadly (Kawabata et al., 2012; Potter et al., 2011). Prior literature demonstrates the role of parenting practices in shaping self-regulatory abilities throughout childhood and across adolescence for both typically developing youth and those with neurodevelopmental disabilities (Hutchinson, Feder, Abar, & Winsler, 2016; Sosic-Vasic et al., 2017). Adaptive parenting has been associated with the development of stronger child self-control and executive functioning skills, while negative parenting practices have been associated with weaker executive functioning skills (Sosic-Vasic et al., 2017). Further, the moderating role of parenting on the relation between higher order cognitive skills and several child outcomes (e.g., autonomy, behavioral difficulties, social adjustment, and academic achievement) has been documented across pediatric neurodevelopmental populations, including youth with ADHD, traumatic brain injury, and epilepsy (Fastenau et al., 2004; Kawabata et al., 2012; Potter et al., 2011).

Despite the importance of medical responsibility, O’Hara & Holmbeck (2013) is the only study to our knowledge that has investigated associations among parenting, executive functioning, and health behaviors in youth with SB using a moderation model. These relationships were explored cross-sectionally, revealing that adaptive parenting behaviors indeed buffered against the expected negative effects of executive functioning deficits on self-
management outcomes (O’Hara & Holmbeck, 2013). Specifically, maternal behavioral control and paternal psychological control moderated the association between executive functioning and adherence, but not medical responsibility. In summary, the role of parenting in modifying associations between neurocognitive functioning and medical responsibility has not been adequately investigated in youth with SB.

Based on existing literature, supportive parents may help buffer against the negative effects of executive dysfunction on medical responsibility among youth with SB. Adaptive parenting behaviors may be particularly important for youth with lower executive functioning and attentional skills, as they require increased support to promote autonomy development and complete health-care related tasks. In contrast, parenting may be less salient for youth with higher executive functioning skills, as these children may be able to adequately manage healthcare responsibilities independently with less parental support. Further, this study aims to understand the different contributions from maternal and paternal parenting to child medical responsibility by including both mothers and fathers of youth with SB.

The current study extends the findings of O’Hara & Holmbeck (2013) in important ways. First, O’Hara & Holmbeck (2013) only examined executive functioning as a singular construct instead of examining domains of executive functioning, and did not include a measure of attentional skills, which restricted the scope of the analyses involving neuropsychological functioning. Second, we aimed to use multiple time points, as these associations have not yet been examined longitudinally, thus reducing the ability to isolate temporal relationships.

We also expanded upon the O’Hara and Holmbeck (2013) study by focusing on the moderational role of age. A developmental perspective highlights the importance of child age in
influencing the moderational role that parenting has on the relationship between executive functioning and medical responsibility (Berg et al., 2017; Lindsay et al., 2011; Reed-Knight et al., 2014). As the emphasis of illness management shifts from family-based management to self-management across adolescence, youth with SB tend to assume more responsibility for health-related tasks with increasing age (Yun & Kim, 2017). Developmentally, it could be expected that the influence of parenting on child outcomes will diminish over time as parents become less involved in adolescents’ daily functioning and individual or external influences become stronger determinants of behavior (Zimmer-Gembeck & Collins, 2008). However, families of youth with SB do not show normative increases in family conflict and youth individuation during the transition to adolescence, suggesting that parenting influences may not be as responsive to developmental changes in SB as they are in families of typically developing youth (Coakley, Holmbeck, Friedman, Greenley, & Thill, 2002). Given these past findings, age was included as an additional moderator in our study to elucidate the relationships among individual and family factors and medical responsibility.

**Current Study.** Building off existing social-ecological models, an examination of systems-level factors, in addition to individual factors, is needed to better understand determinants of medical responsibility in youth with SB. We hypothesized that 1) lower executive functioning/attentional skills would be associated with lower levels of medical responsibility, 2) greater parental acceptance, greater parental behavioral control, and less parental psychological control would be associated with higher levels of medical responsibility, 3) greater parental acceptance and behavioral control would buffer against the negative effects of neuropsychological dysfunction on medical responsibility, 4) greater parental psychological
control would exacerbate the negative impact of neuropsychological deficits on medical responsibility, and 5) parenting would be more likely to moderate the relationship between executive functioning/attentional skills and medical responsibility for younger, versus older, participants (see Figure 9).
Figure 9. Moderating Roles of Parenting and Child Age on the Relationship between Attention & Executive Functioning and Medical Responsibility

Neuropsychological Risk Factors (Time 1):
- Attention
- Working Memory
- Cognitive Shifting
- Planning/Organizing

Parenting Behaviors (T1):
- Acceptance
- Behavioral Control
- Psychological Control

Medical Responsibility (Time 2)

Child Age (T1)
Participants were recruited as part of an ongoing, larger longitudinal study investigating psychosocial, neuropsychological, and familial functioning among children and adolescents with SB (e.g., Devine et al., 2012). The current study examined data collected at Time 1 (ages 8-15 year) and two years later at Time 2. Children with SB and their parents were recruited from four hospitals in the Midwest and a statewide SB association. Inclusionary criteria for participation included: (a) a diagnosis of spina bifida (types included myelomeningocele, lipomeningocele, and 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 our laboratory to facilitate home-based data collections.

Two-hundred and forty-six families were approached during recruitment. Out of the 163 families who agreed to participate initially, 21 families could not be contacted or later declined, and 2 families did not meet all of the inclusion criteria. For the purposes of this study, because higher cognitive capacity is required for managing medical tasks, 30/140 (~20%) participants who functioned intellectually at two or more standard deviations below the population mean (i.e. an estimated intelligence quotient (IQ) score below 70; [American Psychiatric Association, 2013]) were not included. Of the 110 participants that were included at Time 1, 89 (81%) participated at Time 2. Therefore, the final sample used in the analyses included 89 children and adolescents with spina bifida (55.1% female; M_{age} = 11.10 [SD = 2.44]; 59.6% Caucasian, 13.5% African American, 20.2% Hispanic, 6.7% Other), 86 mothers, and 79 fathers (see Table 3).
Table 3. Demographic and Medical Information for Youth who Participated at Time 1 and Time 2

<table>
<thead>
<tr>
<th></th>
<th>Youth (N=89)</th>
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<tbody>
<tr>
<td></td>
<td>M (SD) or N (%)</td>
</tr>
<tr>
<td>Gender: female</td>
<td>49 (55.1%)</td>
</tr>
<tr>
<td>Age</td>
<td>11.10 (2.44)</td>
</tr>
<tr>
<td>Race</td>
<td></td>
</tr>
<tr>
<td>Caucasian</td>
<td>53 (59.6%)</td>
</tr>
<tr>
<td>African-American/Black</td>
<td>12 (13.5%)</td>
</tr>
<tr>
<td>Hispanic/Latino</td>
<td>18 (20.2%)</td>
</tr>
<tr>
<td>Other</td>
<td>6 (6.7%)</td>
</tr>
<tr>
<td>Family Hollingshead SES</td>
<td>43.08 (14.54)</td>
</tr>
<tr>
<td>IQ</td>
<td>92.90 (15.50)</td>
</tr>
<tr>
<td>Spina bifida type</td>
<td></td>
</tr>
<tr>
<td>Myelomeningocele</td>
<td>73 (82.0%)</td>
</tr>
<tr>
<td>Lipomeningocele</td>
<td>12 (13.5%)</td>
</tr>
<tr>
<td>Not Sure/Not reported</td>
<td>4 (4.4%)</td>
</tr>
<tr>
<td>Lesion level</td>
<td></td>
</tr>
<tr>
<td>Thoracic</td>
<td>7 (7.9%)</td>
</tr>
<tr>
<td>Lumbar</td>
<td>46 (51.7%)</td>
</tr>
<tr>
<td>Sacral</td>
<td>31 (34.8%)</td>
</tr>
<tr>
<td>Unknown/not reported</td>
<td>5 (5.6%)</td>
</tr>
<tr>
<td>Shunt: present</td>
<td>62 (69.7%)</td>
</tr>
<tr>
<td>Mothers</td>
<td></td>
</tr>
<tr>
<td>Married or living with significant other</td>
<td>72 (80.9%)</td>
</tr>
<tr>
<td>Single, divorced, or widowed</td>
<td>13 (14.6%)</td>
</tr>
<tr>
<td>Declined to report</td>
<td>1 (1.1%)</td>
</tr>
<tr>
<td>Fathers</td>
<td></td>
</tr>
<tr>
<td>Married or living with significant other</td>
<td>69 (77.5%)</td>
</tr>
<tr>
<td>Single, divorced, or widowed</td>
<td>3 (3.4%)</td>
</tr>
<tr>
<td>Declined to report</td>
<td>7 (7.9%)</td>
</tr>
</tbody>
</table>
**Attrition.** Youth who did not participate at Time 2 did not significantly differ from youth who participated at both time points with respect to type of SB, lesion level, shunt status, gender, race, IQ, Time 1 medical responsibility, neuropsychological functioning, or parenting behaviors. Youth who only participated at Time 1 and not Time 2 were significantly older than those who participated at both time points \([M = 12.14 \text{ versus } 10.80; t(108) = 2.28, p = .03]\).

**Procedure**

Trained undergraduate and graduate student research assistants collected data from families during two separate three-hour home visits at Time 1, and one three-hour home visit at Time 2. Informed consent from caregivers and assent from youth were obtained prior to data collection. Caregivers also signed information release forms to allow for data collection from medical charts, health professionals, and teachers. Caregivers completed several questionnaires separately. Questionnaires were available in both English and Spanish; questionnaires that were only available in English were adapted for Spanish speakers by a translation team using back translation procedures. Youth completed neuropsychological testing with research assistants at Time 1. Additionally, parents and children participated in a series of videotaped interaction tasks. Family tasks included a warm-up game, a discussion of two age-appropriate vignettes, a discussion of transferring disease-specific responsibilities to the child, and a discussion of conflict issues that were previously identified in self-report questionnaires. Families received monetary compensation of $150 and small gifts (e.g., logo t-shirts, pens, water bottles) at each time point for participating.

**Measures**

**Demographics and Medical Data.** Parents reported on youth and family demographic
information via questionnaires at Time 1, including age, gender, race, and ethnicity. 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). Child medical data, including SB type and lesion level, were collected via medical chart review. When medical chart data were not available, parent-report was used.

**Youth IQ.** At Time 1, youth were administered the Vocabulary and Matrix Reasoning subtests of the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999) which were used to estimate a Full Scale IQ score. These subtests have demonstrated high levels of internal consistency for youth 6-16 years old (α = .89 for Vocabulary, α = .92 for Matrix Reasoning; Wechsler, 1999).

**Neuropsychological Functions.** Child attention skills and executive functions were assessed via performance-based measures as well as parent-report, at Time 1. The following areas of neuropsychological functioning were examined: 1) attention, 2) working memory, 3) cognitive flexibility, and 4) planning and organizational skills. Across all domains of neuropsychological functioning, questionnaire measures were reverse scored to be in the same direction as performance-based measures, such that higher scores on all scales and subtests indicated stronger attention or executive functioning skills.

**Attention.** Parents completed the Swanson, Nolan, and Pelham Teacher and Parent Rating Scale (SNAP-IV; Swanson, 1992). The SNAP-IV is comprised of 18 items derived from the criteria for Attention-Deficit/ Hyperactivity Disorder from the DSM-IV (American Psychiatric Association, 1994). Mean subscale scores were calculated for the inattention subscale. The SNAP-IV inattention subscale demonstrated high internal consistency across
mother (α=.92) and father report (α=.92). Youth were also administered a performance-based measure of attention, the Number Detection (ND) Subtest of the Cognitive Assessment System (CAS; Naglieri & Das, 1997). Internal consistency reliability (α = .77) and test-rest reliability (r = .77) for the ND subtest are high across age groups (Naglieri & Das, 1997). The Sky Search, Sky Search DT, Score!, and Score DT subtests from the Test of Everyday Attention for Children were used as additional performance-based measures of different attentional skills, including selective attention, sustained attention, and sustained-divided attention (TEA-CH; Manly, Robertson, Anderson, & Nimmo-Smith, 1999). Adequate test-retest reliability has been reported across subtests (Manley et al., 1999).

**Working Memory.** Parents completed the Working Memory subscale of the Behavior Rating Inventory of Executive Functioning (BRIEF, Gioia et al., 2000a). The BRIEF measures behaviors related to multiple domains of executive functioning over the past six months (Gioia et al., 2000a, 2000b). In this study, the Working Memory subscale demonstrated high internal consistency across mother (α=.90) and father report (α=.91). The Digit Span subtest of the Wechsler Intelligence Scale for Children (WISC-IV; Wechsler, 2003) was used as a performance-based measure of working memory ability. The Digit Span subtest has adequate internal consistency (r = .87) and test-retest reliability (r = .83; Williams, Weiss, & Rolfhus, 2003).

**Cognitive Flexibility.** Parents completed the Shift subscale of the BRIEF, which measures one’s ability to solve problems flexibly and consider various aspects of a problem or situation (Gioia et al., 2000a, 2000b). The Shift subscale demonstrated adequate to high internal consistency across mother (α=.82) and father report (α=.70). Youth were administered the Verbal
Fluency Test of the Delis Kaplan Executive Function System (D-KEFS, Delis, Kaplan, & Kramer, 2001). The Category Switching condition of the Verbal Fluency Test was used as a performance-based measure of cognitive flexibility. This subtest has adequate test-retest reliability ($r = .53-.65$; Delis, Kramer, Kaplan, & Holdnack, 2004).

**Planning and Organizational Skills.** Parents completed the Plan/Organize and the Organization of Materials subscales of the BRIEF (Gioia et al., 2000a, 2000b). The Plan/Organize subscale measures the ability to develop and carry out a set of tasks related to a specific goal. The Organization of Materials subscales measures the child’s tendency to keep their environment neat and orderly. High internal consistency was found across both subscales for mothers ($\alpha = .88-.92$) and fathers ($\alpha = .88-.90$). The Planned Connections (PCn) subtest of the Cognitive Assessment System (CAS) was used as a performance-based measure of planning skills. The PCn subtest has high internal consistency ($\alpha = .77$) and test-retest reliability ($r = .73$) (Naglieri & Das, 1997).

**Parenting Behaviors.** Parenting behaviors were assessed at Time 1 via observational methods. Observational data were coded using the Family Interaction Macro Coding System (FIMS) developed by Holmbeck, Zebracki, Johnson, Belvedere, and Hommeyer (2007) that was adapted from a methodology established by Smetana et al. (1991; see Holmbeck et al., 2002, and Kaugers et al., 2011 for detailed description). Undergraduate and graduate research assistants were trained by discussing individual item codes and reviewing previously coded family interactions with an expert coder. To complete training, research assistants achieved a 90% agreement rate with the expert coder’s ratings on previously coded videos. Coders then independently viewed four family interaction tasks on videotape and provided 5-point Likert
scale ratings on dimensions of parenting behaviors including parental acceptance, behavioral control, and psychological control. The following items were included in each parenting behavior scale: acceptance (listens to others, humor and laughter, warmth, anger [reverse-scored], and supportiveness); behavioral control (confidence in stating opinions, parental structuring of the task, and parental dominance); and psychological control (pressures others to agree, tolerate differences and disagreements [reverse-scored], receptive to statements made by others [reverse-scored], and promotes autonomy in the child [reverse-scored]). For example, “Warmth,” one component of parental acceptance, captures positive connections in a dyadic relationship (e.g. mother-child, father-child) as shown through verbal or nonverbal behaviors (1 = “very cold” and 5 = “very warm”). Higher scores indicated higher observed levels of each parenting behavior. For each task, behaviors were rated by two coders; item level means of the raters for each task were averaged across the tasks and raters to produce a single score for each dimension for each parent. The FIMS parenting behaviors scales demonstrated acceptable scale reliability scores (α=.68-.88) and interrater reliability coefficients (ICCs=.76-.88).

Medical Responsibility. Caregivers 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 SB responsibilities and health-related tasks within the family (e.g., conducting daily skin checks). Parents rated who was primarily responsible for each task (e.g., parent, child, shared, or not applicable). For each task item, a score of “1” indicates the child is primarily responsible, “2” indicates responsibility is shared equally between the parent and child, and “3” indicates the parent was primarily responsible. Items were reverse-coded so that higher scores
reflected greater child responsibility. Mean item scores were used to represent the total responsibility scale. Items that participants rated as “not applicable” were excluded from the mean item scores. Previous studies have not computed internal consistency values for the total scale score of this measure, as reliability software uses listwise deletion when computing alpha coefficients, thus eliminating any participant who provided one or more “not applicable” responses (e.g., Psihogios, Kolbuck, & Holmbeck, 2015; Stern et al., 2018).

**Statistical Treatment**

Data were analyzed using IBM SPSS software, Version 26. For preliminary analyses, data reduction techniques were utilized to reduce the number of analyses and minimize the possibility of type 1 errors. Composite scores were created based on means across multiple reporters and measures. Scores were aggregated across reporters and methods if they met the following criteria: when Pearson correlation coefficients were ≥ .40 between two reporters, and when Cronbach alphas were > .60 among three or more reporters/measures. The following variables were made into composites using standardized values: Attention (SNAP-IV, Tea-Ch [all subtests], CAS-ND), Working Memory (BRIEF-Working Memory, WISC-IV Digit Span), Cognitive Shifting (BRIEF-Shift, D-KEFS Category Switching), Planning/Organizing (BRIEF-Plan/Organize, BRIEF-Organization of Materials, CAS-PCn), and Medical Responsibility (mother- and father-report).

For preliminary analyses, descriptive statistics were used to calculate psychometric properties (i.e., means, standard deviations, scale ranges) of all measures and characterize levels of neuropsychological functioning, parenting behaviors, and child medical responsibility. Outlier (as defined by a z-score > 3.00 that was not part of the normal distribution) and skewness
analyses were conducted at the composite level using guidelines established by Tabachnick & Fidell (2007) and West et al. (1996). Variables with a skewness value > 2.1 were considered a substantial departure from normality and were transformed to create approximately normal distributions. Conservative alpha levels (.001) were used to evaluate the significance of skewness.

Child lesion level, family socioeconomic status, and child medical responsibility at Time 1 were included as covariates in all moderation analyses. Child IQ was not controlled in the analyses, given the contention that intellectual functioning should not be controlled when examining higher order cognitive processes in individuals with neurodevelopmental disorders and because of concerns about statistical overcorrection (Dennis et al., 2009). Pearson correlation coefficients were used to determine relations among executive functioning/attention and parenting behaviors at Time 1 and child medical responsibility at Time 2 (Hypotheses 1-2). An alpha of .05 was used to determine significant effects.

Three-way interactions among attention/executive functioning, parenting behaviors, and child age at Time 1 in predicting medical responsibility at Time 2 (Hypotheses 3-6) were examined using the Hayes PROCESS Macro (Model 3, 2018). A total of 24 models were run (4 neuropsychological factors as predictors x 2 parents x 3 parenting behaviors). As the PROCESS Macro examines all two-way and three-way interactions simultaneously, separate models testing two-way interactions between neuropsychological factors and parenting behaviors were run using the PROCESS Macro. Parenting behavior was identified as the primary moderator (M; Hypotheses 3-4) and child age operated as the secondary moderator (W; Hypothesis 5). When a significant two-way or three-way moderating effect was detected, post hoc analyses utilized
simple slopes at \( \pm 1 \) standard deviation of the parenting behavior and/or child age to examine the impact of executive functioning/attention on medical responsibility at high and low levels of parenting behaviors and/or child age.

Results

Preliminary Analyses

Descriptive properties of variables are shown in Table 4. Composite scores were created for attention \((\alpha = .62)\), working memory \((\alpha = .58)\), cognitive shifting \((\alpha = .63)\), planning/organizing \((\alpha = .67)\), and child medical responsibility \((r = .73)\). Data for observed maternal and paternal parenting behaviors were not significantly correlated and were analyzed separately. Eight cases were detected as outliers among the variables, which were transformed by adding or subtracting one unit from the nearest value in the distribution (Cohen et al., 2003). All variables were within the acceptable range for skewness and did not require further transformation. Bivariate correlations showed that Time 1 attention \((r = .31)\), cognitive shifting \((r = .35)\), maternal acceptance \((r = .28)\), and socioeconomic status \((r = .27)\) were positively related to Time 2 medical responsibility, and Time 1 paternal psychological control \((r = -.25)\) was negatively related to Time 2 child medical responsibility (all \(p’s < .05\)). Additionally, Time 1 maternal acceptance was positively related to Time 1 working memory \((r = .28)\) and cognitive shifting \((r = .16)\), and Time 1 maternal psychological control was negatively related to Time 1 planning/organizing \((r = .28)\), and Time 1 maternal psychological control was negatively related to Time 1 planning/organizing \((r = -.18)\).

Power analyses were conducted to determine if the sample size was appropriate for the proposed statistical analyses (Cohen, 1992). Given the number of variables in each moderation
model (1 predictor, 2 moderators, one 3-way interaction, three 2-way interactions, 3 covariates) and assuming a power of .80 and an alpha of .05, a sample size of 54 is needed to detect large effects, a sample size of 117 is needed to detect medium effects, and a sample size of 819 is needed to detect small effects. Thus, the current study had the ability to detect medium-large to large effect sizes.
Table 4. Correlations among Neuropsychological Variables, Parenting Behaviors, Medical Responsibility, and Covariates

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<td>1. T1 Med. Resp. (\text{a,b})</td>
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<td>.10</td>
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<td>2. T2 Med. Resp. (\text{a})</td>
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<td>3. Attention (\text{c})</td>
<td>.34**</td>
<td>.31**</td>
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<td>4. Working Memory (\text{c})</td>
<td>.19</td>
<td>.21</td>
<td>.71**</td>
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<td>5. Cognitive Shifting (\text{c})</td>
<td>.37**</td>
<td>.35**</td>
<td>.52**</td>
<td>.52**</td>
<td>–</td>
<td></td>
<td>.16*</td>
<td>.04</td>
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<td>6. Plan/Organizing (\text{c})</td>
<td>.15</td>
<td>.16</td>
<td>.56**</td>
<td>.70**</td>
<td>.43**</td>
<td>–</td>
<td>.12</td>
<td>.03</td>
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<td>7. Acceptance</td>
<td>.03</td>
<td>.17</td>
<td>.04</td>
<td>.03</td>
<td>.09</td>
<td>.01</td>
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<td>.34**</td>
<td>.64**</td>
<td>.045</td>
<td>.39**</td>
<td>.09</td>
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<td>8. Behavioral Control</td>
<td>-.20</td>
<td>-.12</td>
<td>-.24</td>
<td>-.20</td>
<td>-.11</td>
<td>-.10</td>
<td>.54**</td>
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<td>.07</td>
<td>-.08</td>
<td>.32**</td>
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<td>9. Psych. Control</td>
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<td>-.25*</td>
<td>-.12</td>
<td>-.12</td>
<td>-.11</td>
<td>-.15</td>
<td>-.79**</td>
<td>-.27*</td>
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<td>-.14</td>
<td>-.24*</td>
<td>-.03</td>
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<td>10. Age</td>
<td>.57**</td>
<td>.53**</td>
<td>.21*</td>
<td>.11</td>
<td>.18</td>
<td>.10</td>
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<td>-.20</td>
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<td>11. SES (\text{b})</td>
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<td>.27*</td>
<td>.08</td>
<td>.01</td>
<td>-.02</td>
<td>-.21</td>
<td>.27*</td>
<td>.05</td>
<td>-.29*</td>
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<td>12. Lesion Level (\text{b})</td>
<td>-.01</td>
<td>-.08</td>
<td>.07</td>
<td>.20</td>
<td>.02</td>
<td>.06</td>
<td>-.08</td>
<td>-.22</td>
<td>.02</td>
<td>.02</td>
<td>-.07</td>
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<tr>
<td>(M (SD)) Maternal Par. (\text{d})</td>
<td>1.75(.36)</td>
<td>1.96(.40)</td>
<td>.04(.60)</td>
<td>.06(.77)</td>
<td>.07(.64)</td>
<td>.05(.69)</td>
<td>3.51(.34)</td>
<td>3.75(.41)</td>
<td>2.22(.36)</td>
<td>11.10(2.34)</td>
<td>43.1(14.54)</td>
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<tr>
<td>(M (SD)) Paternal Par. (\text{e})</td>
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Notes. Values above the diagonal reflect correlations with maternal parenting behaviors. Values below the diagonal reflect correlations with paternal parenting behaviors. SES = socioeconomic status measured by Hollingshead Four Factor Index. All cognitive variables were scored such that higher scores represent stronger skills in attention, working memory, cognitive shifting, and planning/organizing abilities; *Child medical responsibility. These variables are covariates. These variables are based on standardized Z scores. Descriptives for maternal parenting behaviors are provided in this row. Descriptives for paternal parenting behaviors are provided in this row. Descriptive statistics for lesion level are presented in Table 1. *\(p < .05\), ** \(p < .01\).
Moderation Analyses

Two-way interactions. Hierarchical multiple regression analyses were conducted with the PROCESS Macro to test the interactive associations between youth attention/executive functioning and parenting behaviors at Time 1 in relation to child medical responsibility at Time 2 (Hayes, 2018), controlling for medical responsibility at Time 1, child age, child lesion level, and family socioeconomic status. A significant two-way interaction was found between Time 1 planning/organizing abilities and paternal acceptance (F(1,53)=7.33, ΔR²=.04, b =.33, SE = .12, p=.01), with the model accounting for 70% of the variance in Time 2 medical responsibility [F(7,53)=18.05, p<.01], primarily due to the entry of the Time 1 planning/organizing variable. Post-hoc analyses showed that planning/organizing skills were significantly, positively related to future medical responsibility at high, but not moderate or low levels of paternal acceptance (b = .23, SE = .08, p = .01, 95% CI =.07, .39; see Figure 10).

The two-way interaction between Time 1 planning/organizing abilities and paternal psychological control was also significant (F(1,53)=6.87, ΔR²=.04, b =-.38, SE = .14, p=.01), with the model similarly accounting for 70% of the variance in Time 2 medical responsibility [F(7,53)=17.66, p<.01]. Post-hoc analyses revealed that planning/organizing skills were significantly, positively related to future medical responsibility at low (b = .27, SE = .09, p = .01, 95% CI =.08, .46) and moderate (b = .12, SE = .06, p = .03, 95% CI =.01, .24), but not high levels of paternal psychological control (see Figure 11). There were no significant two-way interactions between neuropsychological functioning and the maternal parenting behaviors.
Figure 10. Paternal Acceptance Moderating the Relation between Planning/Organizing and Medical Responsibility
Figure 11. Paternal Psychological Control Moderating the Relation between Planning/Organizing and Medical Responsibility
**Three-way interactions.** Three-way interactions among youth attention/executive functioning, parenting behaviors, and child age at Time 1 in relation to child medical responsibility at Time 2 were then tested, controlling for medical responsibility at Time 1, child lesion level, and family socioeconomic status. Child age was specified as the second moderator to test whether age impacted the strength of these associations (i.e., “moderated moderation”).

A significant three-way interaction was observed among youth cognitive shifting, maternal acceptance, and child age (F(1,65)=5.59, ΔR²=.03, b = -.19, SE = .08, p=.02). The results for this three-way interaction are graphically depicted in Figure 12. This model accounted for a significant portion of variance in Time 2 medical responsibility [F(10,65)=13.38, p<.01, R²=.67]. The direction of the moderation between cognitive shifting and acceptance was probed by examining the effects of cognitive shifting at high (1 SD above the mean), moderate (mean), and low (1 SD below the mean) levels of maternal acceptance. The interaction between cognitive shifting and acceptance was significant among adolescents [i.e., age=13.53 years; b = -.56, F(1,65)=5.25, p=.03] but not pre-adolescents [b = -.09, F(1,65)=0.42, p=.52] or younger children [b = 0.38, F(1,65)=2.59, p=.11].

Among adolescents and in the presence of low maternal acceptance, cognitive shifting abilities were associated with more medical responsibility [b = .24, SE = .11, t= 2.22, p=.02, 95% CI =.03, .49]. However, there was no significant effect of cognitive shifting on medical responsibility in the presence of moderate to high levels of acceptance. Put another way, and as can be seen in Figure 4, adolescents with less developed cognitive shifting skills and low levels of maternal acceptance demonstrated less medical responsibility, while adolescents with low levels of cognitive shifting skills and high levels of maternal acceptance demonstrated more
medical responsibility. Among youth with more highly developed cognitive shifting skills, medical responsibility scores were similar at low, moderate, and high levels of acceptance. While there was not a significant interaction for younger children (age 8.57 years), greater cognitive shifting abilities were significantly associated with more medical responsibility at higher levels of acceptance among these children \[b = .26, \text{se} = .12, t= 2.22, p = .02, 95\% \text{ CI} = .03, .49\]. However, the association between cognitive shifting and medical responsibility was nonsignificant at both lower and moderate levels of acceptance. No other three-way interactions among neuropsychological functioning, parenting behaviors, or age were significant \((p’s > .05)\).
Figure 12. Maternal Acceptance and Child Age Moderating the Relation between Cognitive Shifting and Medical Responsibility
Discussion

The current study sought to examine how parenting behaviors and child age moderated the association between attention/executive functioning and later medical responsibility among a sample of youth with SB. We hypothesized that positive parenting would serve a protective role in buffering against the known negative impact of cognitive functioning on chronic health self-management behaviors. Taking development into account, we also hypothesized that these relations would be stronger for younger children than for adolescents, as parenting may be more influential during this period compared to other (e.g., peer) social contexts. Since youth with SB tend to benefit from families that encourage independence (Loomis et al., 1997), but are at increased risk for delays in autonomy (Friedman et al., 2009), it is important to understand factors that facilitate or hinder this developmental process.

Stronger attention and executive functioning skills, as well as higher levels of parental acceptance and behavioral control and lower levels of psychological control were expected to be related to greater youth responsibility for SB-related tasks. In partial support of these hypotheses, correlational analyses demonstrated that greater attention and cognitive shifting skills, higher levels of maternal acceptance, and lower levels of paternal psychological control were related to more youth medical responsibility two years later. These findings are notable in that inattention, distractibility, and shifting deficits are relatively common among youth with SB (Heffelfinger et al., 2008). When considering SB, our results highlight that stronger attention skills may allow a child to direct and sustain their attention towards salient medical tasks, or use divided attention to complete complex, multi-step tasks, such as clean intermittent catheterization. Further, cognitive shifting skills may be key components of executive functioning that allow the child or adolescent
to flexibly shift between competing demands and manage their daily medical responsibilities across different settings (e.g., home, school).

Within the context of medical responsibility, high acceptance may represent warm, caring, and sensitive parenting that promotes involvement of the child in their medical tasks in a developmentally appropriate way (Lerch & Thrane, 2019). In turn, high psychological control may represent negative, intrusive, and overly harsh parenting behaviors that discourage the child from participating in their own medical care. For youth with SB, and based on the findings of this study, the lack of detrimental, psychological controlling behaviors from fathers may be as important to autonomy development as the presence of accepting behaviors from both mothers and fathers. While parenting that is low in psychological control is generally viewed as more favorable for child psychosocial functioning across mothers and fathers (Steinberg & Silk, 2002), fathers of youth with SB who demonstrate low levels of psychological control, and thus are less intrusive and critical, may respond to their children in a supportive manner that promotes the development of autonomy for medical tasks more so than mothers. Prior literature has shown strong connections between maternal parenting and pediatric health management, but results have been inconsistent as to whether increased involvement of fathers is related to positive self-management and health outcomes among pediatric chronic illness populations (Taylor, Fredericks, Janisse, & Cousino, 2019). This study adds to the literature by showing that supportive paternal parenting plays a role in the transfer of medical responsibility in a sample of youth with SB.

Our results showed that paternal acceptance and paternal psychological control moderated or enhanced the relation between planning/organizing skills and medical
responsibility among youth with SB. These interactions remained significant even after controlling for child age and medical responsibility at the previous time point. Consistent with our hypotheses, greater planning/organizing skills were associated with medical responsibility two years later in the context of high acceptance and low psychological control, respectively. We also found support for a more complex, three-way interaction among cognitive shifting skills, maternal acceptance, and child age. In contrast to expectations, acceptance appeared to moderate the association between cognitive shifting and medical responsibility for adolescents with SB (M_{age}=13.53 years), but not for younger children. Further, the relation between cognitive shifting and medical responsibility was only significant at low levels of acceptance. However, consistent with hypotheses, among adolescents with lower cognitive shifting skills, those with mothers who demonstrated higher levels of acceptance had greater medical responsibility two years later than those with mothers who demonstrated lower levels of acceptance. On the other hand, among those with higher levels of cognitive shifting skills, medical responsibility did not vary as a function of levels of maternal acceptance.

While the findings were statistically nonsignificant for younger children (M_{age}=8.57 years), when examining Figure 4 there is a positive trend (p < .10) between cognitive shifting and medical responsibility within the context of high maternal acceptance for young children. This pattern seemed to parallel the two-way interaction observed among paternal acceptance and planning/organizing skills, except that it was significant for the entire sample (M_{age} of 11.10 years). One possible interpretation of these findings is that accepting parents are sensitive to their child’s age and cognitive capabilities, and regulate the amount of medical responsibility transferred to their child accordingly. Thus, accepting parents may be more in tune with their
child’s readiness to take on SB-related tasks, such that they grant more medical responsibility to adolescents and less responsibility to younger children with relatively less developed executive functioning skills. Further, youth with SB who possess strong executive functioning skills, particularly in the domain of cognitive shifting, may not be as dependent on parental support for acquiring responsibility for their medical care.

Interestingly, and in contrast with our findings, a previous study of SB based on the same data set did not find evidence for an interaction between executive functioning and parenting behaviors in relation to youth medical responsibility (O’Hara & Holmbeck, 2013). As this earlier study utilized cross-sectional data, one explanation for this discrepancy is that parental acceptance and psychological control moderate the association between executive functioning and medical responsibility over time. Additionally, O’Hara and Holmbeck used a global executive functioning composite which aggregated across multiple executive functions, while this study found significant moderation effects for specific attention/executive functioning skills (i.e., cognitive shifting, planning/organizing) and not others (e.g., working memory). Prior research investigating factors related to medical responsibility among youth with SB have similarly noted nuanced associations between executive functions and self-management outcomes (Stern et al., 2018). Indeed, subtle relative weaknesses in executive functions have been identified among youth with SB (Rose & Holmbeck, 2007). As such, our current findings point to the importance of treating such higher order cognitive abilities as discrete skills when investigating executive functioning in SB.

While the current study has multiple areas of strength, such as its longitudinal design, use of multimethod, multi-informant data to reduce the possibility of common method variance, and
investigation of complex moderation models, several limitations should be noted. The small sample size (N’s=60-80) reduced the power of our study and precluded our ability to detect smaller interaction effects. Our significant findings should also be interpreted with caution given the large number of moderation models that were nonsignificant. Additionally, this study included parenting behaviors of both mother and fathers, but did not examine how discrepancies in parenting style or how much time each parent spent with their child impacted the transfer of medical responsibilities. Accounting for dyad-level differences or similarities between parents in relation to cognitive skills and medical responsibility among youth with chronic medical conditions is an important target for future work.

Further, this study has conceptualized the transfer of medical responsibilities as part of overall adjustment for an individual with SB, and thus viewed gains in responsibility as a positive adaptation outcome. Indeed, a smooth, gradual shift in the allocation of SB-specific responsibilities from parents to the adolescent or emerging adult is a normative part of development (Reed-Knight, Blount, & Gilleland, 2014). On the other hand, it should be noted that increased medical responsibility may not necessarily be beneficial for every child with SB or their family. Other relevant factors, including physical health, cognitive capabilities, self-efficacy, family beliefs surrounding illness management, and cultural values about individuality versus interdependence must be carefully considered when making decisions about self-management for youth with a chronic health condition (Modi et al., 2012). Complete responsibility or autonomy for one’s health care may not be an appropriate goal for some youth with SB who have more severe cognitive or physical impairments (Psihogios et al., 2015). Similarly, those who are raised in environments where a more collectivistic care style is
normative may benefit from shared family responsibility of SB-related tasks throughout adolescence and into adulthood (Ohanian et al., 2018).

Relatively, we emphasize that obtaining responsibility for medical care is simply one component of self-management, and should be examined within the context of other self-management behaviors, including treatment adherence and condition knowledge. For instance, the transfer of medical responsibilities may be related to positive psychosocial and health outcomes for an adolescent who adheres to their providers’ treatment recommendations, but could lead to medical complications for an adolescent who is nonadherent. Due to the risk for nonadherence during adolescence, a combination of developmentally appropriate increases in youth medical responsibility with continued parental monitoring, support, or collaboration tends to be associated with the most favorable outcomes for youth with a chronic illness, including those with SB (Lerch & Thrane, 2019; King et al., 2012). Future research adopting a bioneuropsychosocial or social-ecological approach should take these individual, family, and cultural factors into account when investigating the transfer of medical responsibilities in relation to other adjustment outcomes among youth with SB.

In addition to the inclusion of other relevant factors within the broader social-ecological framework, future work should examine growth in medical responsibility among older adolescents and young adults with SB. The transfer of healthcare responsibilities is a developmental process which continues to unfold into emerging adulthood (Reed-Knight, Blount, & Gilleland, 2014). To better understand this process, growth analyses would be able to model trajectories of medical responsibility across adolescence and young adulthood. As executive functions similarly develop across adolescence while parent-child relationships
simultaneously reorganize during this period (Berg et al., 2017; Lerch & Thrane, 2019), future research may examine how changes in cognitive skills and parenting over time are concurrently associated with growth in medical responsibility among youth with SB. Further, while this study investigated parenting as a moderator of executive functioning, caregiving behaviors are reciprocally influenced by child factors and may be moderated by child executive functioning (Gueron-Sola, Bedford, Wagner, & Propper, 2018). Thus, future research may examine ways in which executive functioning moderates relations between different parenting behaviors and self-management outcomes for youth with SB.

Finally, and in line with prior literature (Sosic-Vasic et al., 2017), significant and cross-sectional relations emerged between adaptive maternal parenting behaviors and stronger executive functioning skills. A longitudinal perspective is needed to determine if greater maternal acceptance and behavioral control promote executive functioning skills among youth with SB, or if youth with stronger executive functioning skills evoke positive parenting behaviors among mothers. The link between socioeconomic status and medical responsibilities in families of youth with SB also warrants further investigation.

In conclusion, medical responsibility among youth with SB is a function of multiple individual and parent-level factors. This study extends the literature by examining attention/executive functioning skills and parenting in association with medical responsibilities among youth with neurodevelopmental conditions. For youth with SB, mothers and fathers can help them obtain appropriate responsibility for their health care by providing an autonomy-supportive and structured environment in which they can develop their own executive functioning skills. Interventions designed to promote self-management among older children and
adolescents with SB would benefit from helping youth strengthen their executive functioning skills while providing parents with autonomy-supportive strategies to support their child’s cognitive competencies and medical responsibility (Malheiro, Gaspar, & Barros, 2017). Additionally, our findings underscore the unique influence of both mothers and fathers on the development of children with a chronic illness, and lend further support for a growing movement in pediatric research for the inclusion of fathers in future research and clinical considerations (Taylor et al., 2019).
CHAPTER FOUR
EXECUTIVE FUNCTIONING, ATTENTION, PARENTING BEHAVIORS, AND GROWTH
IN MEDICAL RESPONSIBILITY IN YOUTH WITH SPINA BIFIDA:
A TASK-SPECIFIC APPROACH

Introduction

For youth with a chronic health condition, developing health care autonomy is a key task for adolescence, and is considered part of the foundation for achieving independence and transitioning into young adulthood (Beacham & Deatrick, 2013). The transfer of responsibility for health-related issues from family- to self-management is a complex process which unfolds over time and depends on both youth and parent components. Acquiring responsibility for one’s health care may be particularly important to study among youth with spina bifida (SB), a congenital neural tube defect that impacts multiple systems and can lead to significant cognitive and physical impairments (Copp et al., 2015). Depending on the lesion level and degree of illness severity, SB is associated with varying degrees of sensory loss and paralysis, neurological complications (e.g., hydrocephalus), urinary/bowel difficulties, deficits in executive functioning and inattention, seizures, and learning disabilities.

Youth with SB tend to lag behind their peers with regard to developing autonomy across different domains (Devine, Wasserman, & Gershenson, 2011; Friedman, Holmbeck, DeLucia, Jandasek, & Zebracki, 2009; Holmbeck et al., 2003). Further, many individuals with SB display
lower levels of intrinsic motivation, greater passivity, and increased dependence on caregivers, which complicates their development of autonomy more generally (Holmbeck, Westhoven, et al., 2003; O’Hara & Holmbeck, 2013; Rose & Holmbeck, 2007; Psihogios, Murray, Zebracki, Acevedo, & Holmbeck, 2016). When considering the psychosocial needs of adolescents with SB, the Spina Bifida Association care guidelines explicitly recommend a gradual and appropriate development of healthcare responsibility (Spina Bifida Association, 2018). Despite the need to prioritize studies of autonomy among youth with SB, research exploring medical responsibility in this population remain sparse.

As described in prior literature, certain individual- and family-level factors, including executive functioning/attentional skills and parenting behaviors, are associated with autonomy-related outcomes in youth with SB, such as the transfer of responsibility for medical tasks and independent living (Holmbeck et al., 2002; Psihogios et al., 2016; Ries et al., 2003; Sawin, Brei, & Adams, 2003; Tuminello, Holmbeck, & Olson, 2012). Such research has found that stronger executive functioning/attention and adaptive parenting behaviors (e.g., acceptance, appropriate limit-setting) are related to higher levels of medical responsibility, while weaker cognitive skills and maladaptive parenting behaviors (e.g., overprotectiveness, excessive intrusiveness, psychological manipulation) are related to lower levels of responsibility. Indeed, it has been hypothesized that multiple characteristics, including physical capability, knowledge of SB, skill mastery, cognitive ability, and familial support may be associated with self-management in SB (Jacobson et al., 2013). This study aims to build on past literature by evaluating higher order neuropsychological functioning and parenting behaviors as predictors of change over time in
medical responsibility as reflected in the development of self-management across multiple, specific SB-related regimen tasks.

Due to the heterogeneous nature of SB, individuals with SB often must adhere to medical regimens that vary as a function of the level of condition severity and an individual’s unique needs. Moreover, the pervasive impact of SB makes self-management routines complex and multi-faceted. Common tasks that an individual with SB may need to engage in include medication management, clean intermittent catheterization, bowel management programs, monitoring for shunt malfunction, conducting skin checks for pressure wounds, caring for assistive devices, and maintaining a specialized diet and exercise habits. In addition to completing several multi-step medical tasks throughout the day, individuals must also remember the specific times at which to initiate them (i.e., remembering to remember to do tasks; Jacobson et al., 2013). Finally, there are a number of tasks that need to be completed on a non-daily basis, such as ordering medications and medical supplies, and scheduling doctors’ appointments. Because the specific medical regimen will differ from person to person, using a total medical responsibility score as a singular outcome in research may not adequately capture the heterogeneity of SB management requirements. Instead, examining medical responsibility through a task-specific perspective may be more appropriate. Thus, in the current study, change across time in one’s responsibility for SB-specific medical tasks is examined for each task separately, so as to provide greater insight into the complexities and nuances of the condition management demands.

Few studies have examined medical responsibility for specific SB-related medical tasks. Prior research shows that adolescents with SB acquire autonomy for health-related and adaptive
skills, including knowledge of secondary medical complications, toileting, intermittent self-catheterization, bowel program, skin checks, exercise, and making appointments, at different ages, suggesting that these tasks should be examined separately (Castillo et al., 2017; Davis, Shurtleff, Walker, Seidel, & Duguay, 2006; Psihogios, Kolbuck, & Holmbeck, 2015). Similarly, Stepansky et al. (2009) noted that responsibility for catheterization is transferred from the parents to the child at an earlier age than bowel program management. Psihogios et al. (2016) examined responsibility for catheterization and bowel program as discrete tasks, and found that neuropsychological functioning, family stress, and peer conflict were related to bowel management, but not catheterization. However, the Psihogios et al. (2016) study was limited in detecting statistically significant findings as it relied on a categorical approach to medical responsibility (i.e., participants were considered ‘autonomous’ or ‘not autonomous’ for their healthcare responsibilities based on a cutoff score). Further, there have been no studies that have examined cognitive and parenting factors in relation to the transfer of medical responsibilities for multiple SB medical regimen tasks.

Some have speculated that youth may assume responsibility for tasks related to urinary and bowel incontinence earlier because of the personal and private nature of such procedures (Stepansky et al., 2009). If this were the case, individual factors, such as cognitive functioning, would be a more relevant determinant of self-management for these domains. On the other hand, parents may be less likely to transfer responsibility for non-regular tasks such as ordering supplies and making doctors’ appointments, as such tasks require additional planning, intrinsic motivation, and problem-solving skills. Given that individuals with SB struggle with these types of higher order cognitive skills, they may rely more heavily on parents to keep track of when
supplies are running low, reach out to medical supply companies to place orders, schedule medical appointments, and coordinate information among necessary providers. A task-specific approach allows us to determine if cognitive functioning versus parenting behaviors differentially predict medical responsibility outcomes for different medical regimen skills.

As the development of medical responsibility is a process, the literature supports using a longitudinal approach to examine the transfer of responsibilities over time (Psihogios et al., 2015; Psihogios et al., 2016; Stepansky et al., 2009). Recently, Kayle et al. (under review) found evidence for two trajectory groups of medical responsibility in a sample of youth with SB; one group acquired responsibility for medical tasks more quickly over time than a slower growth group. Further, these groups differed significantly by child IQ, gender and familial stress, such that male youth with lower IQs and greater familial stress were more likely to be in the group which acquired medical responsibility at a slower rate. Previous studies have investigated associations between executive functioning, parenting behaviors, and medical responsibility among youth with SB (O’Hara & Holmbeck, 2013; Psihogios et al., 2016). However, none of these past efforts have examined how these specific cognitive or parenting factors promote or impede the growth of independence with medical responsibilities over time within this population. Growth analyses enable the exploration of developmental trajectories in medical responsibility (DeLucia & Pitts, 2005). Further, growth analyses clarify temporal relationships between key predictors (i.e., executive functioning/attention and parent behaviors) and subsequent change in medical autonomy. In support of this method, Friedman et al., (2009) used growth curve modeling to show that youth with SB demonstrate different rates of growth in emotional and behavioral autonomy over time as compared with their typically developing peers.
**Current study:** The aims of the present study were: 1) to describe the patterns of growth in medical responsibility for separate healthcare tasks among youth with SB, and 2) to determine whether executive functioning/attention and parenting behaviors differentially contribute to the development of medical responsibility across various healthcare tasks in youth with SB. Consistent with the literature on medical responsibility (Psihogios et al., 2015), it was hypothesized that youth with SB would assume more responsibility for their healthcare tasks over time. It was also hypothesized that individual cognitive abilities (executive functioning/attention) would be more likely to influence the growth of medical responsibility for tasks that are considered more personal in nature (e.g., bladder and bowel management) and that parenting behaviors would be more likely to influence the development of medical responsibility for tasks which are non-daily and require advanced planning and coordination (e.g. making doctors’ appointments).

**Method**

**Participants**

Participants were recruited as part of a larger, ongoing longitudinal study exploring neuropsychological functioning, psychosocial adjustment, and family relationships among youth with SB (e.g., Devine et al., 2012). Data were collected across five time points (ages 8-15 years at Time 1; 16-23 years at Time 5) and occurred approximately every two years. Families of children with SB were recruited from four hospitals in the Midwest and a statewide SB association. Participants were included if they met the following criteria: (a) spina bifida diagnosis (types included myelomeningocele, lipomeningocele, and myelocystocele); (b) age 8–15 years; (c) ability to speak and read English or Spanish; (d) involvement of at least one
primary caregiver; and (e) residence within 300 miles of our laboratory to allow for home-based data collections.

During recruitment, two-hundred and forty-six families were approached. Of the 163 families who agreed to participate, 2 families did not meet all of the inclusion criteria, and 21 families could not be contacted or later declined. The final sample of participants included 140 families of children with SB (53.6% female; $M_{age} = 11.43$ [SD = 2.46]; 52.9% Caucasian; Table 5). While previous studies examining medical responsibility excluded participants with an estimated intellectual functioning below 70 (Stern et al., 2018; Stern et al., in press), all youth were included in this study regardless of their IQ. The purpose of including all participants was to maximize sample size for growth analyses and to capture the broad cognitive capabilities of spina bifida, given recent findings regarding associations between cognitive functioning and SB medical responsibility (Kayle et al., under review). Youth who declined to participate did not differ significantly from those who agreed to participate with regard 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 $\chi^2 (1) = 1.08, p > .05$. 
<table>
<thead>
<tr>
<th></th>
<th>Youth (N=140)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>M (SD) or N (%)</td>
</tr>
<tr>
<td>Participants</td>
<td>140 (100%)</td>
</tr>
<tr>
<td>Age</td>
<td>11.43 (2.46)</td>
</tr>
<tr>
<td>Gender: female</td>
<td>75 (53.6%)</td>
</tr>
<tr>
<td>Race</td>
<td></td>
</tr>
<tr>
<td>Caucasian</td>
<td>74 (52.9%)</td>
</tr>
<tr>
<td>African-American/Black</td>
<td>19 (13.6%)</td>
</tr>
<tr>
<td>Hispanic/Latino</td>
<td>39 (27.9%)</td>
</tr>
<tr>
<td>Asian</td>
<td>2 (1.4%)</td>
</tr>
<tr>
<td>Other</td>
<td>6 (4.3%)</td>
</tr>
<tr>
<td>Spina bifida type</td>
<td></td>
</tr>
<tr>
<td>Myelomeningocele</td>
<td>122 (87.1%)</td>
</tr>
<tr>
<td>Non-myelomeningocele</td>
<td>17 (12.1%)</td>
</tr>
<tr>
<td>Unknown/not reported</td>
<td>1 (0.7%)</td>
</tr>
<tr>
<td>Lesion level</td>
<td></td>
</tr>
<tr>
<td>Thoracic</td>
<td>23 (16.4%)</td>
</tr>
<tr>
<td>Lumbar</td>
<td>69 (49.3%)</td>
</tr>
<tr>
<td>Sacral</td>
<td>41 (29.3%)</td>
</tr>
<tr>
<td>Unknown/not reported</td>
<td>7 (5.0%)</td>
</tr>
<tr>
<td>Shunt: present</td>
<td>109 (77.9%)</td>
</tr>
<tr>
<td>IQ</td>
<td>85.75 (19.54)</td>
</tr>
<tr>
<td>Family SES</td>
<td>39.12 (16.09)</td>
</tr>
</tbody>
</table>
Procedure

At Time 1, trained graduate student and undergraduate research assistants collected data over two separate three-hour home visits; one three-hour home visit was required at subsequent time points, which were spaced approximately two years apart (Times 2-5). Prior to data collection, caregivers provided informed consent and youth provided assent. Families received $150 and small gifts (e.g., t-shirts) for compensation. Caregivers also signed release of information forms to allow for data collection from medical charts, health professionals, and teachers. Caregivers and youth completed questionnaires independently. Questionnaires were available in both English and Spanish; questionnaires that were only available in English were adapted for Spanish speakers by a translation team using back translation procedures. Participants also completed a neuropsychological test battery at Time 1. Families participated in a series of audio and videotaped interaction tasks to provide observational data. These tasks included a warm-up game, a discussion of two age-appropriate vignettes, a discussion of transferring disease-specific responsibilities to the child, and a discussion of conflict issues that were previously identified in paper questionnaires. The task order was randomly assigned. Youth also participated in observational tasks with their best friend, but these data are not presented in this report.

Measures

Demographics and Medical Data. At Time 1, caregivers reported on youth and family demographic information via questionnaires, including age, gender, race, and ethnicity. 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,
Parent report and/or medical chart review were used to collect child medical data (i.e., SB type and lesion level).

**Youth IQ.** The Vocabulary and Matrix Reasoning subtests of the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999) were administered at Time 1, which were used to estimate a Full Scale IQ score. These subtests have demonstrated high levels of internal consistency for youth 6-16 years old (α = .89 for Vocabulary, α = .92 for Matrix Reasoning; Wechsler, 1999).

**Neuropsychological Functions.** At Time 1, child attentional functioning and executive functioning skills were assessed via performance-based measures as well as parent- and teacher-report. As described below, the following areas of neuropsychological functioning were examined: 1) attention, 2) working memory, 3) cognitive flexibility, and 4) planning and organizational skills.

**Attention.** The Swanson, Nolan, and Pelham Teacher and Parent Rating Scale (SNAP-IV; Swanson, 1992) was completed by parents and teachers. The SNAP-IV includes 18 items derived from the criteria for Attention-Deficit/Hyperactivity Disorder from the DSM-IV (American Psychiatric Association, 1994). Mean subscale scores were calculated for the inattention subscale, and subscale totals were reverse scored so that higher scores reflected stronger attentional ability. The SNAP-IV inattention subscale has demonstrated high internal consistency (α = .90) across race and age groups (Bussing et al., 2008). Youth also completed the Number Detection (ND) Subtest of the Cognitive Assessment System as a performance-based measure of attention (CAS; Naglieri & Das, 1997). Internal consistency reliability (α = .77) and test-rest reliability (r = .77) for the ND subtest are high across age groups (Naglieri & Das,
To provide additional measures of various attention skills (i.e., selective attention, sustained attention, and sustained-divided attention), the Sky Search, Sky Search DT, Score!, and Score DT subtests from the Test of Everyday Attention for Children were used (TEA-CH; Manly, Robertson, Anderson, & Nimmo-Smith, 1999). Adequate test-retest reliability has been reported across subtests (Manley et al., 1999). For the ND subtest of the CAS and subsequently described performance-based measures of executive functioning, higher scores reflect stronger cognitive skills.

**Working Memory.** The *Working Memory* subscale of the Behavior Rating Inventory of Executive Functioning was completed by caregivers and teachers (BRIEF, Gioia et al., 2000a). The BRIEF taps into different domains of executive functioning by measuring aspects of the child's behavior in a natural environment over the past six months (Gioia et al., 2000a, 2000b). As higher scores on the BRIEF indicate greater impairment in executive functioning, total subscale scores were reverse coded so that higher scores reflected stronger executive skills. The Working Memory subscale has demonstrated high internal consistency (α=.89) and test-retest reliability (r=.85) for parent report. To provide a performance-based assessment of working memory, the Digit Span subtest of the Wechsler Intelligence Scale for Children (WISC-IV; Wechsler, 2003) were used. The Digit Span subtest has good internal consistency (r = .87) and test-retest reliability (r = .83; Williams, Weiss, & Rolfhus, 2003).

**Cognitive Flexibility.** The *Shift* subscale of the BRIEF was completed by parents and teachers, which assesses the ability to make transitions and solve problems with flexibility (Gioia et al., 2000a, 2000b). The Shift subscale has demonstrated high internal consistency (α=.81) and test-retest reliability (r = .78) for parent report. Youth completed the Verbal Fluency Test of the
Delis Kaplan Executive Function System as a performance-based assessment of cognitive flexibility (D-KEFS, Delis, Kaplan, & Kramer, 2001). Specifically, the Category Switching condition of the Verbal Fluency Test was used. This subtest has adequate test-retest reliability ($r = .53-.65$; Delis, Kaplan, & Kramer, 2004).

**Planning and Organizational Skills.** The Plan/Organize and the Organization of Materials subscales of the BRIEF were completed by parents and teachers (Gioia et al., 2000a, 2000b). The Plan/Organize subscale captures the ability to set goals, and develop steps and organize information in order to meet future goals. The Organization of Materials subscales measures the ability to organize and keep track of one’s belongings. These subscales have demonstrated high internal consistency ($\alpha=.87-.90$) and test-retest reliability scores ($r = .82-.85$) for parent report. The Planned Connections (PCn) subtest of the Cognitive Assessment System (CAS) was used as a performance-based measure of planning skills. The PCn subtest has high internal consistency ($\alpha = .77$) and test-retest reliability ($r = .73$) (Naglieri & Das, 1997).

**Parenting Behaviors:** Parenting behaviors were assessed at Time 1 via observational methods. Four family interaction tasks were coded using the Family Interaction Macro Coding System (FIMS) developed by Holmbeck, Zebracki, Johnson, Belvedere, and Hommeyer (2007) that is based on a methodology devised by Smetana et al. (1991). Coders separately viewed the interaction tasks on videotape and rated items related to parenting behaviors on a 5-point Likert scale. Specific macro-level scales used in this study were parental acceptance, behavioral control, and psychological control. Acceptance included the following items: “listens to others”, “humor and laughter”, “warmth”, “anger” (reverse-scored), and “supportiveness”. Behavioral control was comprised of the following items: “confidence in stating opinions”, “parental
structuring of the task”, and “parental dominance”. Psychological control was derived from the following items: “pressures others to agree”, “tolerates differences and disagreements” (reverse-scored), “receptive to statements made by others” (reverse-scored), and “parent promotes autonomy in the child” (reverse-scored). Higher scores indicated higher observed levels of each parenting behavior. For analyses, two coders rated behaviors within each task. Item level means for each task were averaged across the tasks and both raters to create a single score for each dimension for mothers and father separately. The FIMS parenting behaviors demonstrated acceptable to strong scale reliability scores ($\alpha=.68-.88$) and acceptable interrater reliability coefficients ($ICCs=.74-.88$).

**Medical Responsibility.** 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), was completed by youth across five timepoints spanning eight years to assess the division of medical responsibilities within the family. The SOSBMR includes items related to SB responsibilities and health-related tasks. This study included the following subscales based on clinical relevance and sufficient variability across all five timepoints in our study sample: healthcare appointments, communicating SB-related needs to teachers, friends, and relatives, catheterization, bowel management, skin care/checking for pressure wounds, and exercise. Specific items included in each subscale are provided in Table 6. For each task item, a score of “1” indicates the child is primarily responsible, “2” indicates responsibility is shared equally between the parent and child, and “3” indicates the parent was primarily responsible. Youth also reported when certain tasks were not applicable to their medical regimen. Items were reverse-coded so that higher scores reflect
greater child responsibility. Mean item scores were used to represent each responsibility subscale. Items that participants rated as “not applicable” were excluded from the mean item scores. Internal consistency values for scales were not computed, as reliability software uses listwise deletion when computing alpha coefficients, which eliminates all participants who provided one or more “not applicable” responses (Driscoll et al., in press; Stern et al., 2018; Psihogios, Kolbuck, & Holmbeck, 2015).
Table 6. Spina Bifida Medical Responsibility Items by Task

<table>
<thead>
<tr>
<th>Task</th>
<th>Items</th>
</tr>
</thead>
<tbody>
<tr>
<td>Appointments</td>
<td>• Remembering day of clinic appointment</td>
</tr>
<tr>
<td></td>
<td>• Making appointments with doctors</td>
</tr>
<tr>
<td></td>
<td>• Talking with doctors about medical questions and requests</td>
</tr>
<tr>
<td>Communicating about SB</td>
<td>• Explaining absences from school to teachers</td>
</tr>
<tr>
<td></td>
<td>• Telling teachers about SB</td>
</tr>
<tr>
<td></td>
<td>• Telling relatives about SB</td>
</tr>
<tr>
<td></td>
<td>• Telling friends about SB</td>
</tr>
<tr>
<td>Catheterization</td>
<td>• Remembering to catheterize regularly</td>
</tr>
<tr>
<td></td>
<td>• Washing hands and genital areas before catheterizing</td>
</tr>
<tr>
<td></td>
<td>• Gathering appropriate catheterization equipment</td>
</tr>
<tr>
<td></td>
<td>• Lubricating catheter</td>
</tr>
<tr>
<td></td>
<td>• Properly inserting catheter</td>
</tr>
<tr>
<td></td>
<td>• Draining bladder completely and removing catheter</td>
</tr>
<tr>
<td></td>
<td>• Cleaning, storing, and discarding catheterization equipment properly</td>
</tr>
<tr>
<td>Bowel Management</td>
<td>• Taking suppositories, enemas, stool softeners, or laxatives as needed</td>
</tr>
<tr>
<td></td>
<td>• Maintaining a regular bowel toileting time</td>
</tr>
<tr>
<td></td>
<td>• Cleaning up after self if an accident occurs</td>
</tr>
<tr>
<td></td>
<td>• Monitoring bowel functioning by keeping a log</td>
</tr>
<tr>
<td></td>
<td>• Remembering to eat foods with lots of fiber</td>
</tr>
<tr>
<td></td>
<td>• Remembering to drink lots of fluid</td>
</tr>
<tr>
<td>Skincare</td>
<td>• Avoiding products that may contain latex, if allergic</td>
</tr>
<tr>
<td></td>
<td>• Protecting skin from temperature, textures, and injury</td>
</tr>
<tr>
<td></td>
<td>• Conducting daily skin checks for pressure wounds</td>
</tr>
<tr>
<td>Exercise</td>
<td>• Following a regular physical exercise routine</td>
</tr>
</tbody>
</table>
**Analysis Plan.** Descriptive statistics were calculated with IBM SPSS software, Version 26. Outliers (as defined by a z-score > 3.30) and skewness analyses were conducted using guidelines established by Tabachnick & Fidell (2007) and West et al. (1996). Variables were considered skewed if their skewness value was greater than 2.0. Seven cases were detected as outliers among the variables (including paternal acceptance, maternal and paternal behavioral control, maternal and paternal psychological control), which were transformed by adding or subtracting one unit from the nearest value in the distribution (Cohen et al., 2003). None of the variables met criteria for skewness and did not require transformation. To reduce the potential number of analyses, measures were aggregated across reporter and/or method using standardized values if they met the following criteria: when Pearson correlation coefficients were ≥ .40 between two reporters, and when Cronbach alphas were > .60 among three or more reporters/measures. Based on these criteria, an attention composite (α=.67) was created from parent and teacher-report on the SNAP-IV and TEA-Ch subtests. A working memory composite (α=.67) was formed from the BRIEF subscale (parent-and teacher-report) and WISC-IV. A cognitive flexibility composite (α=.68) was formed from the BRIEF subscale (parent- and teacher-report) and D-KEFS. A planning/organizing composite (α=.72) was created from the BRIEF subscales (parent- and teacher-report) and CAS. To further reduce the number of analyses, a global executive functioning (EF) composite (α=.83) was formed from the working memory, cognitive flexibility, and planning/organization domains.

**Growth Analyses.** Growth curves using linear mixed effects models were estimated with SAS Proc Mixed statistical software (SAS Proc Mixed; SAS Institute, 1996; Laird & Ware, 1982) to investigate patterns of change in medical responsibility over time, as defined by
changes across participant age. “Time” was specified by chronologic age instead of arbitrary study time points for data collection, to examine how growth parameters vary as a function of age and to maintain a developmental perspective (see Stiles-Shields et al., 2019, for further explanation). Age was centered at the mean at Time 1 (11.5 years). Data from five time points (spanning from ages 8-15 at Time 1 to ages 16-23 at Time 5) were utilized in the analyses.

A series of models were estimated, beginning with unconditional growth models, to describe changes in medical responsibility across each healthcare task. The unconditional growth curve models examined participant age as a predictor of medical responsibility. Mixed effects modeling simultaneously estimated average intercepts and slopes (i.e., fixed effects) and variability in intercepts and slopes. The significance of variances of these growth parameters were first tested to determine heterogeneity across participants; which was used to indicate if there was adequate variability to proceed with the growth analyses (Ghisletta, Renaud, Jacot, & Courvoisier, 2015).

Then, conditional growth curve models were estimated to examine the effects of baseline attention/EF skills and parenting behaviors as predictors of growth in medical responsibility. The following time-invariant covariates were entered as Time 1 predictors of growth parameters: cognitive functioning (attention and EF), and parenting behaviors (maternal and paternal acceptance, behavioral control, and psychological control). Child lesion level and family socioeconomic status at Time 1 were included as covariates in all analyses. Due to recent findings that intelligence (IQ) was related to growth in medical responsibility in youth with SB (Kayle et al., under review) analyses were run with and without child IQ entered as a covariate, to determine the effects of attention/EF and parenting above and beyond the influence of IQ.
Separate mixed-models were estimated for growth in responsibility for each health-related task. Missing data were addressed using full maximum likelihood estimation (Mehta & West, 2000).

**Results**

**Trajectories of Healthcare Responsibilities.** Results of unconditional growth models, including estimates of fixed effects for intercepts and slopes as well as variability in intercepts and slopes, are presented in Table 7 (see Figure 13 for a visual representation). Significant, positive linear slopes were found for all SB-related tasks, indicating that youth medical responsibility increased as participants became older ($p$’s > .05). Significant variability in intercepts was found across all scales except for healthcare appointments, indicating that youth differed in their average level of medical responsibility for those tasks at age 11.5 years. Significant variability in slopes was found for all scales except for communicating about SB and skincare, signifying that youth differed in how rapidly they acquired medical responsibility over time for those tasks. Due to the lack of variance around the intercept for healthcare appointments and slope for SB communication and skincare, variance for these respective parameters were constrained to 0 in subsequent models.
Table 7. Longitudinal Trajectories of Spina Bifida Medical Responsibility Tasks: Results of Unconditional Growth Models

<table>
<thead>
<tr>
<th>Variable</th>
<th>Fixed Effects</th>
<th>Variance Components</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>b</td>
<td>SE</td>
<td>p</td>
</tr>
<tr>
<td>Appointments</td>
<td>Intercept</td>
<td>1.223</td>
<td>.022</td>
</tr>
<tr>
<td></td>
<td>Slope</td>
<td>0.069</td>
<td>.007</td>
</tr>
<tr>
<td>Communicating about SB</td>
<td>Intercept</td>
<td>2.040</td>
<td>.042</td>
</tr>
<tr>
<td></td>
<td>Slope</td>
<td>0.050</td>
<td>.007</td>
</tr>
<tr>
<td>Catheterization</td>
<td>Intercept</td>
<td>2.194</td>
<td>.057</td>
</tr>
<tr>
<td></td>
<td>Slope</td>
<td>0.077</td>
<td>.009</td>
</tr>
<tr>
<td>Bowel Program</td>
<td>Intercept</td>
<td>2.083</td>
<td>.049</td>
</tr>
<tr>
<td></td>
<td>Slope</td>
<td>0.064</td>
<td>.009</td>
</tr>
<tr>
<td>Skincare/Pressure Wounds</td>
<td>Intercept</td>
<td>1.931</td>
<td>.049</td>
</tr>
<tr>
<td></td>
<td>Slope</td>
<td>0.083</td>
<td>.008</td>
</tr>
<tr>
<td>Exercise</td>
<td>Intercept</td>
<td>2.256</td>
<td>.056</td>
</tr>
<tr>
<td></td>
<td>Slope</td>
<td>0.056</td>
<td>.010</td>
</tr>
</tbody>
</table>

*Age, centered at 11.5 years, was used to define time (slope).

The final model for this task did not include a standard error or p-value for the variance component for intercept, indicating variability for the intercept was close to 0.
Figure 13. Unconditional Growth Models of Child Responsibility for SB Medical Tasks
Predictors of Healthcare Responsibilities. Attention, executive functioning (EF), and maternal and paternal acceptance, behavioral control, and psychological control were then entered separately as predictors of growth for each aspect of SB medical responsibility (Table 8). Without controlling for IQ, baseline child attention was positively related to the intercept for communicating about SB ($b = 0.123$, $SE = 0.034$, $p < .01$), catheterization ($b = 0.348$, $SE = 0.094$, $p < .01$), bowel program ($b = 0.275$, $SE = 0.081$, $p < .01$), and skincare/pressure wound management ($b = 0.258$, $SE = 0.983$, $p < .01$), suggesting that youth with greater attention skills had more responsibility for these tasks at 11.5 years. Executive functioning was positively associated with the intercept for catheterization ($b = 0.289$, $SE = 0.098$, $p < .01$) and skincare/pressure wounds ($b = 0.183$, $SE = 0.872$, $p = .04$), such that youth with stronger EF skills had more responsibility for these tasks at age 11.5 years. Attention ($b = 0.038$, $SE = 0.011$, $p < .01$) and EF ($b = 0.024$, $SE = 0.011$, $p = .03$) were significant positive predictors of individual differences in the slope for appointment making, indicating that youth with stronger attention/EF skills experienced a faster rate of gains in medical responsibility for managing their healthcare appointments, compared with youth with less developed baseline levels of attention and EF.

Maternal behavioral control was negatively related to the intercept for communicating about SB ($b = -0.327$, $SE = 0.103$, $p < .01$), suggesting that youth with mothers who exhibited greater behavioral control had less responsibility for communicating about SB with their friends, teachers, and relatives than those with mothers who demonstrated less behavioral control. Paternal psychological control significantly negatively predicted individual differences in slope for appointment making ($b = -0.045$, $SE = 0.022$, $p = .04$), such that youth with fathers who
demonstrated more psychological control acquired responsibility for this task more slowly than youth with fathers who were less psychologically controlling.
Table 8. Predictors of Growth Models of Medical Responsibility Outcomes across Age, Without Controlling for IQ

<table>
<thead>
<tr>
<th>Medical Responsibility Task Outcome</th>
<th>Predictors of Trajectories</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Attention</td>
</tr>
<tr>
<td>Healthcare Appointments</td>
<td>Intercept</td>
</tr>
<tr>
<td></td>
<td>-0.042</td>
</tr>
<tr>
<td>Communication about SB</td>
<td>0.123*</td>
</tr>
<tr>
<td>Catheterization</td>
<td>0.348***</td>
</tr>
<tr>
<td>Bowel Program</td>
<td>0.275***</td>
</tr>
<tr>
<td>Skincare</td>
<td>0.258**</td>
</tr>
<tr>
<td>Exercise</td>
<td>0.089</td>
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</tbody>
</table>

*p <.05; **p<.01; ***p<.001.
Baseline child IQ, when added as a covariate, was significantly related to the intercept for catheterization ($b = 0.012, SE = 0.003, p < .01$), bowel program ($b = 0.008, SE = 0.003, p < .01$), skincare/pressure wounds ($b = 0.010, SE = 0.003, p < .01$), and exercise ($b = 0.006, SE = 0.003, p < .01$), such that youth with a higher IQ had more responsibility for these tasks at 11.5 years.

Child IQ positively predicted individual differences in slope for healthcare appointments ($b = 0.001, SE = 0.0003, p = .01$), bowel program ($b = 0.001, SE = 0.0004, p = .02$), and exercise ($b = 0.001, SE = 0.0005, p = .04$), indicating that youth with a higher IQ acquired responsibility for these tasks at a faster rate than those with a lower IQ.

When IQ was included in models as a covariate (Table 9), attention remained a significant positive predictor of individual differences in intercept for communicating about SB ($b = 0.045, SE = 0.010, p = .01$) and slopes for healthcare appointments ($b = 0.043, SE = 0.011, p < .01$). Executive functioning remained a significant positive predictor of individual differences in slopes for healthcare appointments ($b = 0.026, SE = 0.011, p = .02$). Executive functioning also emerged as a significant positive predictor of individual differences in intercept for communicating about SB ($b = 0.023, SE = 0.010, p = .02$). Maternal behavioral control remained a significant negative predictor of intercept for communicating about SB ($b = -0.321, SE = 0.104, p < .01$) and paternal psychological control remained a significant negative predictor of individual differences in slope for healthcare appointments ($b = -0.045, SE = 0.022, p = .04$).
Table 9. Predictors of Growth Models of Medical Responsibility Outcomes across Age, Controlling for IQ

<table>
<thead>
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<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Healthcare Appointments</td>
<td></td>
<td>-0.039</td>
<td>0.043**</td>
<td>-0.059</td>
<td>0.026**</td>
<td>-0.035</td>
<td>-0.008</td>
<td>-0.033</td>
<td>-0.028</td>
<td>-0.031</td>
<td>-0.035</td>
<td>0.0002</td>
<td>0.005</td>
<td>-0.010</td>
<td>-0.014</td>
<td>-0.033</td>
<td>-0.045*</td>
</tr>
<tr>
<td>Communication about SB</td>
<td></td>
<td>0.045*</td>
<td>0.026</td>
<td>0.023**</td>
<td>0.013</td>
<td>-0.133</td>
<td>0.005</td>
<td>-0.321**</td>
<td>0.011</td>
<td>-0.074</td>
<td>-0.017</td>
<td>0.161</td>
<td>0.012</td>
<td>0.106</td>
<td>-0.004</td>
<td>-0.226</td>
<td>-0.008</td>
</tr>
<tr>
<td>Catheterization</td>
<td></td>
<td>0.134</td>
<td>-0.002</td>
<td>0.083</td>
<td>0.001</td>
<td>-0.133</td>
<td>-0.005</td>
<td>0.009</td>
<td>-0.013</td>
<td>0.012</td>
<td>0.007</td>
<td>-0.035</td>
<td>0.003</td>
<td>-0.013</td>
<td>0.005</td>
<td>-0.057</td>
<td>0.019</td>
</tr>
<tr>
<td>Bowel Program</td>
<td></td>
<td>0.130</td>
<td>0.008</td>
<td>0.018</td>
<td>0.019</td>
<td>-0.163</td>
<td>0.002</td>
<td>0.121</td>
<td>-0.018</td>
<td>0.085</td>
<td>0.014</td>
<td>-0.279</td>
<td>0.011</td>
<td>-0.150</td>
<td>-0.002</td>
<td>0.206</td>
<td>-0.002</td>
</tr>
<tr>
<td>Skincare</td>
<td></td>
<td>0.078</td>
<td>0.006</td>
<td>-0.002</td>
<td>0.012</td>
<td>-0.022</td>
<td>-0.008</td>
<td>-0.186</td>
<td>-0.007</td>
<td>-0.088</td>
<td>-0.004</td>
<td>&lt;.001</td>
<td>0.017</td>
<td>0.136</td>
<td>-0.018</td>
<td>0.077</td>
<td>-0.028</td>
</tr>
<tr>
<td>Exercise</td>
<td></td>
<td>-0.025</td>
<td>0.018</td>
<td>0.014</td>
<td>0.012</td>
<td>-0.216</td>
<td>0.026</td>
<td>0.089</td>
<td>-0.012</td>
<td>0.247</td>
<td>-0.027</td>
<td>-0.192</td>
<td>0.010</td>
<td>-0.030</td>
<td>-0.018</td>
<td>0.181</td>
<td>-0.016</td>
</tr>
</tbody>
</table>

*p <.05; **p<.01; ***p<.001.
Discussion

This study explored trajectories of growth in child responsibility for different spina bifida (SB)-related healthcare tasks from ages 8 to 23 years, and examined the role of cognitive and parenting factors as predictors of these trajectories. Although an increase in youth responsibility for medical tasks is a well-documented process that occurs across adolescence in other chronic illness populations (e.g., type 1 diabetes; Silva & Miller, 2019), this phenomenon has not been studied extensively in youth with SB. We found that, overall, youth with SB gained responsibility gradually and linearly for their medical tasks across adolescent and young adulthood. In our sample, scores showed that early adolescents (or preteenagers, i.e., 11.5 years old) shared responsibility for communicating about their medical condition with others, catheterization, bowel program management, skincare, and exercise with their parents, and they gradually progressed towards being predominantly, but not solely, responsible for tasks in young adulthood. Parents were primarily responsible for managing healthcare appointments well into young adulthood, with young adults assuming shared responsibility with their parents for their medical appointments at age 22.

The gradual transfer of responsibilities across development represents a time when adolescents and young adults can practice managing their medical regimen and develop competencies within the safe and supportive context of their family environment before they transition to self-management and greater independence. Given that youth with SB tend to demonstrate delays in behavioral and emotional autonomy more broadly compared to typically
developing peers (Friedman et al., 2009), it is promising that individuals continued to gain responsibility for their medical tasks across adolescence and young adulthood.

Findings also suggest that, for the majority of tasks, there is variation in how much responsibility they have as a preteenager as well as the rate at which they assume autonomy for different tasks. Further, certain variables demonstrated utility in predicting these trajectories across adolescence and young adulthood. Within the cognitive domain, baseline attention was positively associated with individual variability in overall child responsibility for communicating about SB, catheterization, bowel program, and pressure wounds, indicating that youth with greater attention skills displayed more responsibility for these tasks at preadolescence (age 11.5 years). Further, attention was positively associated with change in responsibility for managing healthcare appointments, indicating that youth with greater attention acquired responsibility for this task at a faster rate than those with lower attentional skills. EF skills were positively associated with the child responsibility for catheterization and skincare at preadolescence. Higher EF was also predictive of faster growth in child acquisition of responsibility for managing healthcare appointments. Full-scale intellectual ability (IQ) was a salient predictor of overall child medical responsibility and growth in acquisition for responsibility across multiple tasks. Moreover, some of the initial effects of attention and EF remained significant even after including IQ in the models.

Although, within the parenting domain, findings were less robust, maternal behavioral control was negatively associated with the child responsibility for communicating about their condition at preadolescence, such that youth of mothers with more behavioral control displayed less responsibility at 11.5 years. Paternal psychological control was negatively associated with
the growth in responsibility for appointment keeping, indicating that youth of fathers with more psychological control acquired responsibility for this task more slowly over time. Both of these parenting behaviors retained their significance after child IQ was entered into the models as a covariate.

Taking these results together, cognitive functioning, including greater general intellectual ability, attention, and executive functioning, behavioral control, and psychological control may impact the acquisition of responsibility for SB-related medical tasks. Our finding that child IQ was robustly related to child medical responsibility across tasks is in line with existing evidence (Kayle et al., under review; Psihogios et al., 2016; Friedman et al., 2009). Intellectual ability may reflect capability of completing medical tasks or condition severity, and has been linked to parental overprotection and lower autonomy granting among parents of youth with SB (Holmbeck, Johnson, et al., 2002).

Our hypotheses, that individual cognitive factors would predict growth in responsibility for tasks that were more private in nature while parenting behaviors would predict growth in responsibility for tasks that are nondaily and require interfacing with others, were partially supported, in that attention/EF and specific parenting behaviors (i.e., behavioral and psychological control) were predictive of growth in certain tasks, but not others. Why would cognitive and parenting factors be related to child acquisition of responsibility for healthcare appointments and communicating about SB with others, but only cognitive, and not parenting behaviors, be related to child responsibility for catheterization, bowel program, and skincare tasks? Perhaps, for individuals with neurological conditions, cognitive functioning profoundly impacts their ability to execute such complicated, multi-step daily self-management tasks (e.g.,
catheterization, bowel programs) more so than the influence of parental behaviors. Indeed, neuropsychological functioning has emerged as a strong predictor of self-management in past research (Psihogios et al., 2016).

Regarding SB communication and medical appointments, individuals with SB have a condition that impacts multiple systems, which requires them to plan for, remember, and attend medical appointments with multiple providers (e.g., primary care provider, neurosurgeon, urologist, orthopedic surgeon, physical medicine and rehabilitation, gastroenterologist; Copp et al., 2015). Attention and EF may reflect stronger self-regulatory abilities and are tied to other aspects of self-management, including medical adherence (Psihogios et al., 2016). Due to increased self-regulation and social communication abilities, individuals with stronger cognitive skills may feel more confident and ready to assume responsibility for engaging with healthcare providers, teachers, and friends sooner than those with less developed cognitive functioning.

Further, while it is not expected that preteenagers be responsible for making clinic appointments, maternal behavioral control may reflect either parental overprotectiveness or lack of child readiness to engage with doctors during medical visits. Moreover, paternal psychological control may reflect parenting that is not autonomy-supportive and potentially hinders adolescents from increasing engagement with their physicians during medical appointments. Parents of youth with SB tend to exhibit higher levels of psychological control than families of typically developing children, and this behavior is associated with poorer outcomes for youth with SB (Holmbeck, Shapera et al., 2002; Murray et al., 2015). Self-advocacy skills are essential to the safety and well-being of individuals with disabilities. Thus, it is important that clinicians and families find
ways to empower and support youth in communicating their SB-related needs to the medical team or others (e.g., friends, teachers) as necessary.

Although we expected that consistent, high levels of parental warmth and acceptance would promote growth in medical responsibility, acceptance showed little utility in predicting child autonomy in this study. This is surprising given that high acceptance has been tied to positive youth adjustment outcomes in SB (Greenley, Holmbeck, & Rose, 2006). However, our measure of parenting behaviors did not differentiate between medical versus non-medical situations. It is possible that a measure of parental acceptance more finely tuned towards medical care would yield different results. Additionally, it is possible that other family, parent, or peer-related factors (e.g., family stress; Kayle et al., under review; parental distress; Driscoll et al., in press; peer conflict; Psihogios et al., 2016 are more relevant to trajectories of medical responsibilities than parental acceptance.

Though the current study was innovative in terms of the study methodology, such as the inclusion of multiple informants, use of performance-based and observational measures, and a longitudinal design, there were several limitations. Attention/EF and parenting behaviors were treated as time-invariant predictors. While beyond the scope of this study, it is possible that change in attention/EF and parenting, rather than baseline functioning across these domains, is more strongly associated with growth in SB tasks. That is, future research should explore how growth in attention/EF skills covaries with growth in medical responsibility. As parenting is a dynamic, developmental process that shares a transactional relationship with child behavior (Roskam & Meunier, 2012), future work should also examine if changes in autonomy-supportive parenting behaviors are more closely associated with medical responsibility, rather than
parenting at baseline. Additional contextual factors, including the effects of co-parenting, cultural beliefs, and expectations about child autonomy for medical care were not considered in relation to SB medical responsibility and serve as areas for further investigation. Additionally, bidirectional or transactional relationships between our predictors and medical responsibility were not examined. Specifically, youth who exhibit difficulties with assuming responsibility for their SB regimen may evoke certain parenting behaviors, such as increased control, which may, in turn, affect their development of autonomy.

Lastly, we only focused on predicting trajectories of medical responsibility as opposed to other self-management variables. Ultimately, to achieve a richer, holistic picture of what constitutes adaptive development of self-management for each child, more research is needed to understand how medical responsibility changes over time in tandem with other self-management processes. These may include medical adherence, knowledge and mastery of SB tasks, and self-efficacy perceptions related to self-management. It will also be important to consider potential barriers that may underlie a gap in an individual’s capacity to complete their own SB-related tasks and their actual performance of medical tasks in their environment (e.g., financial strain impacting family’s ability to obtain medical supplies; World Health Organization, 2007).

The significant variation in growth demonstrated across many of the SB tasks in this study highlight how each child is on their own trajectory towards self-management. Future research should aim to answer the following questions: What constitutes the “right” time for families to begin transferring responsibility for medical management to the child, and how can clinicians and families best support this developmental process? It is likely that these timelines differ depending on the complexity of the task, the child’s cognitive and psychosocial readiness,
and the broader environmental or contextual factors that may impact the task at hand. Due to the heterogenous nature of SB, it may not be helpful or clinically relevant to compare the transfer of medical responsibilities of a child with a high IQ and strong attention and executive functioning skills to a child with relative deficits in these areas. In fact, it may be more adaptive for parents of individuals with a significant intellectual or cognitive disability to retain responsibility for complex tasks and for such individuals to follow a modified trajectory. Ideally, clinicians and families will tailor support for transferring medical responsibilities to the unique needs and preferences of the individual.

Clinically, providers should know where families fall on the continuum of self-management (from parent to child responsibility) for their various SB-related tasks so as to assist them with the transfer of responsibilities across adolescence. Clinicians working in a rehabilitation or multidisciplinary setting can administer measures of medical responsibility to youth and engage families in conversations about the transfer of these skills early on to prepare them for greater independence. Given that the full-scale IQ test used in this study was comprised of two relatively short subtests, *Vocabulary* and *Matrix Reasoning*, this measure could be administered within a clinic setting to give providers and families a better understanding of the child’s potential trajectories for assuming responsibility for their medical care. Pediatric psychologists can screen youth for problems with attention or EF during clinical interviews, or assist families with obtaining referrals to neuropsychologists who can further assess these abilities.

Finally, psychologists can also consider how the burden of attention/EF deficits and parental overcontrol impacts independence with medical care for youth with SB beyond the
influence of IQ alone. Targeting deficits in these areas to assist adolescents with increasing responsibility for their SB regimen may require making modifications to the environment, such as increased structure, or modifications to the medical regimen itself (e.g., working with the physician to simplify the bowel management routine; Tarazi, Mahone, & Zabel, 2007). Family-based interventions that target maladaptive parenting behaviors and provide psychoeducation about cognitive deficits as they relate to self-management issues may be beneficial as well.
CHAPTER FIVE

DISCUSSION

Review of Study Purposes and Results

Medical responsibility, a domain of self-management, is an important part of autonomy development for youth with SB. Given the challenges that youth with SB face with autonomy more broadly, it is critical that researchers and clinicians understand how the transfer of medical responsibilities from parents to children unfolds over time and what factors impact this process. This collection of research utilized a multi-informant, multi-method design and sought to understand different ways in which individual factors and parenting behaviors related to medical responsibility in youth with SB. These studies are grounded within existing social-ecological and biopsychosocial frameworks of self-management for youth with chronic medical conditions (Modi et al., 2012; Psihogios et al., 2016).

The first study focused on individual factors, and examined two mediational pathways through which child depressive symptoms and attention/executive functioning deficits were associated with medical responsibility over time among youth with SB. Study 1 found that depressive symptoms mediated the relations between attention/working memory and child medical responsibility. Specifically, greater deficits in attention and working memory at Time 1 were associated with more severe depressive symptoms at Time 2, which in turn were associated with lower child medical responsibility at Time 3. Further, although this study found some evidence that youth depressive symptoms were significantly associated with level of
medical responsibility (teacher-report of child depressive symptoms was associated with medical responsibility but not parent-report or self-report), attention and executive functioning skills emerged as salient factors related to child SB medical responsibility.

Building off of the findings from the first study, the second study aimed to further examine associations between higher order cognitive functioning and child medical responsibility while incorporating the role of contextual factors, specifically parenting behaviors, as moderators. This study also aimed to take a developmental perspective by examining child age as a second moderator (i.e., “moderated moderation”). Significant two-way interactions between child neuropsychological functioning and parenting behaviors were found between youth planning/organizing abilities and paternal acceptance, as well as youth planning/organizing abilities and paternal psychological control. Greater youth planning/organizing skills at Time 1 were related to more medical responsibility at Time 2 within the context of high paternal acceptance, and low paternal psychological control. A significant three-way interaction was found among youth cognitive shifting abilities, maternal acceptance, and child age. Maternal acceptance moderated the relation between cognitive shifting skills at Time 1 and medical responsibility at Time 2 for adolescents, but not younger children. Among adolescents with less developed cognitive shifting skills, those with mothers who were rated as higher in acceptance showed greater medical responsibility than those with mothers who were rated as lower in acceptance. Among adolescents with stronger cognitive shifting abilities, medical responsibility did not vary as a function of maternal acceptance.

The third study used growth analyses to examine trajectories of child medical responsibility across adolescence and into young adulthood. To acknowledge the wide variety of
tasks that could be included in a medical regimen for individuals with SB, this study took a task-specific approach to medical responsibility, rather than examining it as a singular construct (as had been the case in the previous studies). An additional objective was to explore attention/executive functioning and parenting behaviors as predictors of these growth trajectories. Findings demonstrated that youth with SB gradually acquired responsibility over time for all medical tasks, including managing healthcare appointments, communicating with others about SB, catheterization, bowel program, skin care, and exercise. Further, stronger attention and executive functioning skills emerged as significant, positive predictors of growth in medical responsibility, while excessive maternal behavioral control and paternal psychological control emerged as negative predictors of medical responsibility.

Several conclusions can be drawn from these findings. Similar to other pediatric chronic medical conditions (Wiebe et al., 2014), responsibility for SB-related medical tasks is best conceptualized as a developmental process that is gradually transferred from parents to youth. Further, youth with SB may differ in their baseline level of medical responsibility for certain tasks, as well as the rate at which they acquire responsibility for these tasks, suggesting that a “one-size-fits-all” approach to understanding medical responsibility within this population is not appropriate (Kayle et al., under review). While child medical responsibility has been examined cross-sectionally among youth with SB (O’Hara & Holmbeck, 2013), this research is among the first to examine longitudinal trajectories of child medical responsibility for SB across the key developmental periods of adolescence and young adulthood.

How and when youth with SB gain responsibility for their medical care likely depends on a complex interaction between the demands of the individual medical task, youth cognitive and
psychosocial functioning, child age, and parenting factors. Stronger intellectual ability, attention, and executive functioning, as well as higher levels of parental acceptance and lower levels of behavioral and psychological control, may serve as facilitators of medical responsibility, while lower cognitive abilities, greater depressive symptoms, low levels of parental acceptance, and high levels of behavioral and psychological control are risk factors for less medical responsibility in SB. These findings are noteworthy in that 1) youth with SB tend to exhibit deficits across multiple attention and executive functioning domains (Rose & Holmbeck, 2007), and 2) parents of youth with SB tend to exhibit more overcontrolling behaviors compared to typically developing children (Holmbeck, Shapera, et al., 2002). In particular, the significant attention and executive functioning deficits that individuals with SB face, even with average intelligence, often become more salient in adolescence and can impede upon their ability to accomplish medical tasks independently.

Due to the longitudinal study design used throughout this collection of research, it can be inferred that individual child factors, particularly attention/executive functioning, and parenting behaviors are associated with medical responsibility over time. Moreover, attention/executive functioning skills, depressive symptoms, and parenting behaviors are related to one another and medical responsibility in nuanced ways. Our findings lend further support to the bio-neuropsychosocial and social-ecological models of pediatric self-management (Modi et al., 2012; Psihogios et al., 2016), demonstrating that researchers and clinicians must consider how these pieces fit together to impact medical responsibility, rather than in individual silos. The findings that attention and executive functioning skills were each significantly related to youth depressive symptoms and parenting behaviors also underscores the significance of conceptualizing
interrelationships among these variables. For example, executive functioning and parental psychological control were directly related to child medical responsibility for SB-related tasks, but psychological control also served as a moderator in the association between executive functioning and medical responsibility.

The first two studies demonstrate the importance of examining the different domains of attention and executive functioning abilities separately, as they may share varying relationships with youth depressive symptoms, parenting behaviors, and medical responsibility. The third study highlights the benefits of treating SB-related medical tasks individually, as certain tasks (e.g., catheterization) may be transferred to adolescents before others (e.g., healthcare appointments). The second and third studies emphasize the significance of including both mothers and fathers in research of families and youth with SB, as maternal and paternal acceptance, behavioral control, and psychological differed in their relations with youth cognitive functioning and medical responsibility. While clear distinctions in the effects of mothering versus fathering on SB medical responsibility did not emerge for acceptance and behavioral control, paternal psychological control was associated with medical responsibility across both studies, while maternal psychological control was not. Although mothers typically take on the primary role in managing their child’s healthcare and fathers are traditionally seen as more removed from family medical management issues (Waizenhofer, Buchanan, & Jackson-Newsom, 2004), this research underscores the unique contributions of the father-child relationship to SB self-management processes (Taylor, Fredericks, Janisse, & Cousino, 2019). Fathers who demonstrate low psychological control may be an important component of an autonomy-supportive environment when considering SB medical responsibility.
The direction of parenting effects was mostly consistent with hypotheses, in that higher levels of parental acceptance and lower levels of psychological control were related to more child medical responsibility. However, while it was hypothesized that higher behavioral control would be related to greater autonomy, higher levels of maternal behavioral control were related to less child medical responsibility in the third study. It is possible that behavioral control may reflect parental overcontrol, rather than firm limit-setting, in this context. Overall, these findings underscore the unique influence of both parents on the development of children with a chronic illness, and lend further support for the inclusion of fathers in future research and clinical considerations (Bogossian et al., 2019). It is clear that additional research is needed to disentangle the effects of mothers and fathers on medical responsibility in youth with SB.

The present studies did not find evidence that participant gender was significantly related to medical responsibility outcomes. The current literature on gender and SB autonomy is mixed. Kayle et al. (under review) and Friedman et al. (2009) demonstrated that boys with SB are at greater risk for delayed medical and nonmedical autonomy compared to girls with SB. However, gender failed to significantly predict individual decision making, intrinsic motivation, or independent behavior in a sample of youth with SB (Friedman, 2005). It is important to note that the Kayle et al. study detected gender differences at the level of rate of growth in medical responsibility, not overall levels of medical responsibility. Further, Kayle et al. examined total SB medical responsibility rather than responsibility for specific SB tasks. Thus, male gender may predict certain aspects of medical responsibility (e.g., total growth) as opposed to overall levels or growth in specific tasks related to medical responsibility.
Strengths, Limitations, and Future Research

The present studies have key methodological strengths, including their longitudinal design, use of multiple informants and methods, and examination of an important yet under-researched aspect of self-management in youth with a chronic medical condition. Several limitations should be noted when interpreting findings that provide considerations for future research. Statistically, all three studies were only powered to detect medium to large effects. Longitudinal studies with pediatric populations traditionally are often underpowered due to low base rates in the population (Holmbeck, Bruno, & Jandasek, 2005). As seen in the second study, some findings were statistically trending, but nonsignificant. These studies should ideally be replicated with larger sample sizes that could ultimately detect smaller effects. Additionally, the questionnaire-based measure of medical responsibility only took into account child or parental perceptions of the division of SB-related responsibilities in the household. Future research should include an objective means of assessing SB medical responsibility that is less susceptible to social desirability effects (e.g., 24-hour daily diary interviews; Quittner et al., 2008).

These studies treated youth medical responsibility as outcome and did not consider bi-directional or transactional effects; that is, how changes in a child’s responsibility for their medical care may impact individual or family functioning. Adolescents who do not make gains in medical responsibility over time may develop poor self-esteem or depressive symptoms as they compare themselves to peers, which in turn could negatively impact self-management (Lindsay, Kingsnorth, & Hamdani, 2011; Reed-Knight, Blount, & Gilleland, 2014). It is possible that youth with SB who lack the initiation skills to take an increasingly active role in their medical care may elicit greater control from parents. Indeed, children who struggle with intrinsic
motivation can evoke autonomy-controlling parenting behaviors (Grolnick, 2009). While parenting behaviors were treated as static and time-invariant here, parent-child relationships and family dynamics reorganize across adolescence (Berg et al., 2017; Roskam & Meunier, 2012). Simultaneously, attention and executive functioning skills continue to develop across adolescence and into emerging adulthood (Berg et al., 2017; Taylor, Barker, Heavey, & McHale; 2015). Examining these predictors as longitudinal rather than static may better reflect the development of autonomy. Thus, future studies should explore how changes in parenting behaviors, attention, and executive functioning skills are concurrently associated with changes in child medical responsibility.

Moreover, while this research focused on exploring associations among key cognitive skills, parenting variables, and SB medical responsibility in greater depth, social-ecological models of pediatric self-management point to other individual, family, and community factors that should be evaluated in relation to medical responsibility (Modi et al., 2012; Psihogios et al., 2016). Research concerning youth with SB and other neurodevelopmental conditions (e.g., traumatic brain injury, autism spectrum disorder) has highlighted scaffolding as a parenting variable that could have potential implications for medical responsibility in youth with SB (Gerrard-Morris et al., 2010; Will, 2012; Winning et al., under review). Future work may also examine how caregiving behaviors interact with each other (i.e., co-parenting), in addition to the role that siblings play in the completion of SB medical tasks. Despite the salience of peer relationships during adolescence, few studies have examined peer factors in relation to medical responsibility (Psihogios et al., 2016).
Regarding distal factors, family socioeconomic status (SES) was associated with SB medical responsibility as well as maternal and paternal parenting behaviors in the current studies. In fact, SES has been shown to be significantly related to parenting behaviors as well as executive functioning skills in typically developing children and individuals with neurodevelopmental conditions (Muller, 2013; Potter et al., 2011). Financially, a child may have difficulty gaining responsibility for their SB care if the family is unable to obtain the medical supplies needed to carry out SB-related tasks, or afford transportation to attend medical appointments. Thus, access to resources should be included as a predictor in future studies of SB medical responsibility.

While these studies viewed increased medical responsibility as an adaptive youth outcome, there are caveats to this assumption. Developing responsibility for one’s healthcare depends not only on the child’s developmental stage, but also their physical, cognitive, and psychosocial ability to perform medical tasks (Beachman & Deatrick, 2013). For adolescents with SB who have severe intellectual deficits or limited mobility, complete medical autonomy may not be feasible. Conversely, it may be beneficial to the child’s physical health and well-being for caregivers to retain shared or total responsibility for complex medical tasks.

Furthermore, the unfolding of youth autonomy occurs within a cultural context and is shaped by familial and societal norms (Wray-Lake, Crouter, & McHale, 2010). From a Western perspective, striving for increased independence across development is idealized. However, in collectivistic cultures that value developmental goals of relatedness and interdependence, total child autonomy for their medical care is not necessarily an adaptive or appropriate goal (McCabe, 1996; Tamis-Lemonda et al., 2008). In one case study, Ohanian et al. (2018) described
a Palestinian-American young adult with SB who deferred medical decision-making to her family members, and whose parents managed her SB-related care. Parent management of her SB was adaptive for this family as it aligned best with Islamic principles (Rathor, Azarisman, & Hasmoni, 2016). Similarly, in Latino culture, it may be normative for an individual’s extended family to remain involved in treatment decision-making and planning due to *familismo* (i.e., significance of the family). Indeed, pediatric health care providers who show respect for *familismo* can help facilitate adherence among Latino youth with chronic health conditions (Antshel, 2002). Examining the impact that cultural beliefs and values have on the unfolding of medical responsibility in youth with SB is an important area for future work.

Finally, although medical responsibility is a valuable aspect of SB self-management to study in its own right, it is critical to understand how medical responsibility interacts with other self-management processes, such as medical adherence, knowledge of SB tasks, and perceptions of illness. To ensure a smooth transition from family- to self-management, responsibility for medical tasks should increase as youth develop self-efficacy for SB management and learn more about SB, their unique health history, and their individualized medical regimen (Lerch & Thrane, 2019). In other pediatric chronic health conditions, increases in adolescent medical responsibility have been linked to decreases in adherence, but this relationship depends on the youth’s level of cognitive and psychosocial maturity (e.g., type 1 diabetes; Silva et al., 2019; Wiebe et al., 2014).

Further, evidence suggests that continued parental monitoring, as developmentally-appropriate, can protect against the deterioration of care during adolescence (Wiebe et al., 2014). Increases in child responsibility that correspond with decreases in medical adherence could result in life-threatening secondary complications for individuals with SB, such as urinary tract
infections, pressure wounds, or limb amputation (Copp et al., 2015). In addition to traditional indicators of youth adjustment, such as independent living or employment/higher education, future research should examine the presence of secondary medical complications as an outcome related to SB medical responsibility. Exploring how medical responsibility changes over time in concordance with other self-management processes will allow researchers and clinicians to better understand how to best meet the needs of youth with SB across the developmental spectrum.

**Clinical Implications and Conclusions**

Clinically, providers should thoroughly evaluate and monitor how families are dividing medical responsibility across various tasks throughout adolescence and young adulthood, as excessive parental involvement and poor self-management can be barriers to successfully navigating the transition to adult healthcare services (Zhou, Roberts, Daliwal, Della, 2016). Further, a child’s cognitive and psychosocial functioning must be taken into account when considering how responsible they can be with their SB medical care. Within the context of a multidisciplinary SB clinic, psychologists can advocate for increased screening for attention and executive functioning challenges and access to formal neuropsychological testing for youth who demonstrate difficulties in these domains. Indeed, clinical guidelines recommend that youth with SB undergo neuropsychological testing due to the increased risk for attention and executive functioning problems that often become apparent in early adolescence (Spina Bifida Association [SBA], 2018).

Neuropsychological testing can provide a comprehensive understanding of a child’s cognitive strengths and weaknesses, as well as recommendations that will help clinicians and families strengthen the development of skills which underlie increased medical responsibility,
such as decision-making, goal-setting, communication, and self-regulation (SBA, 2018). For example, youth with cognitive deficits may benefit from modifying complicated, multi-step medical regimens or using technology to support increased responsibility (e.g., phone alarms; Dicianno et al., 2016). For those with communication difficulties, health professionals may take steps to increase the structure of medical visits by encouraging patients to write down questions they have for the physician before the appointment. The family context constitutes another area for assessment and intervention, as caregiving behaviors can impact the unfolding of medical responsibility over time. Clinicians can encourage parents to provide an autonomy-supportive environment in which youth with SB can safely learn and practice their medical tasks. Family-based interventions that help the family recognize a child’s cognitive strengths or weaknesses and promote autonomy-supportive behaviors as they relate to the child’s SB-related care may be beneficial.

This research has showed that cognitive, psychosocial, and parenting factors relate to medical responsibility in youth with SB. Many of the variables included in social-ecological frameworks of pediatric self-management remain untested, particularly broader environmental or community contexts. Thus, there are several avenues for further research. Ultimately, as the evidence base on self-management in youth with SB continues to evolve, it is hoped that observational research will help inform interventions that can support youth with SB and their families as they navigate the transition towards self-management and increased independence.
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)
- Swanson, Nolan, and Pelham – Fourth Edition (SNAP-IV)
- Teacher Report Form (TRF)

**Direct Assessment Measures:**

- Cognitive Assessment System (CAS)
- Delis Kaplan Executive Function System (D-KEFS)
- Family Interaction Macro Coding System (FIMS)
- Test of Everyday Attention for Children (TEA-Ch)
- Wechsler Abbreviated Scale of Intelligence (WASI)
- Wechsler Intelligence Scale for Children-Fourth Edition (WISC-IV)
REFERENCE LIST

Chapter One


spina bifida: A longitudinal investigation of medical adherence, responsibility-sharing, and independence skills. *Journal of Pediatric Psychology, 40*(8), 790-803.


depressive symptoms, neuropsychological functioning, and medical responsibility in youth with spina bifida: Examining direct and mediating pathways. *Journal of Pediatric Psychology, 43*(8), 895-905.


Chapter Two


Chapter Three


Chapter Four


behaviors are associated with condition-related responsibility in youth with spina bifida. *Journal of Pediatric Psychology."


Chapter Five


VITA

Dr. Stern graduated from the Clinical Psychology PhD program at Loyola University Chicago, with specialties in child, adolescent, and family issues and neuropsychology. She received her B.A. in Psychology from the University of Pennsylvania in 2013, graduating summa cum laude. Her interest in pediatric psychology developed while she was an undergraduate at the University of Pennsylvania, where she conducted research on posttraumatic stress in ill and injured children and coping in pediatric cancer at the Children’s Hospital of Philadelphia. At Loyola, Dr. Stern has been a member of Dr. Grayson Holmbeck’s research lab, studying families of youth with spina bifida. Dr. Stern received her M.A. in Clinical Psychology from Loyola in 2017. Her thesis examined the relationships among neuropsychological functioning, depressive symptoms, and self-management in youth with spina bifida. As part of Dr. Holmbeck’s lab, she has worked on multiple projects related to pediatric psychology, which has resulted in presentations at conferences, peer-reviewed journal articles, and book chapters. Her work has received multiple awards and honors from regional and national organizations including the Society for Pediatric Psychology, Illinois Spina Bifida Association, and Loyola University Chicago. She also been elected to serve on multiple leadership boards within the Society of Pediatric Psychology. She completed her clinical doctoral internship in Child, Adolescent, and Pediatric Psychology at Rush University Medical Center in Chicago, IL.