



2022

Growth of Condition-Related Knowledge Among Youth with Spina Bifida: Associations with Neurocognitive Functioning and Self-Management Skills

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LOYOLA UNIVERSITY CHICAGO

GROWTH OF CONDITION-RELATED KNOWLEDGE AMONG YOUTH
WITH SPINA BIFIDA: ASSOCIATIONS WITH NEUROCOGNITIVE
FUNCTIONING AND SELF-MANAGEMENT SKILLS

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

PROGRAM IN CLINICAL PSYCHOLOGY

BY
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CHICAGO, IL
MAY 2022

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ACKNOWLEDGEMENTS

I would like to thank my mentor, Dr. Grayson Holmbeck, for his support, encouragement, and advice throughout the thesis process. I would also like to thank Dr. Amy Bohnert for her thoughtful advice and guidance. Dr. Zoe Smith was also instrumental to this project, and I am so thankful for her support and guidance. Lastly, I am exceptionally grateful for the contributions of the youth and families who participated in the CHATS study, without whom this research would be impossible.

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ABSTRACT

Spina bifida (SB) is a congenital birth defect that may involve physical, medical, and neuropsychological complications due to central nervous system malformation (Copp et al., 2015). Ambulation problems, bladder and bowel dysfunction, and hydrocephalus require daily medical management tasks, including catheterization, bowel program management, and skin checks to avoid secondary complications. While self-management skills are typically gained during adolescence, executive dysfunction in SB may complicate gains (Dennis et al., 2006; Greenley, 2010). Indeed, evidence in other chronic conditions suggests that adolescent executive dysfunction is linked to poorer medical self-management. Condition-related knowledge has been identified as an important basis of medical self-management, as it involves condition etiology, awareness of potential complications, and a grasp of functional limitations (Greenley et al., 2006; Lansing & Berg, 2014). Little is known about condition-related knowledge growth in SB, and its associations with executive dysfunction and SB-related skills. Therefore, the current study examined (1) the average trajectory of growth in condition-related knowledge in youth with SB, (2) executive functioning variables as predictors of growth in condition-related knowledge, (3) growth in condition-related knowledge as a predictor of medical self-management skills in adolescence, and (4) the mediating role of SB condition-related knowledge on associations between executive dysfunction and medical self-management skills.

Participants ($n = 140$ youth with SB) were recruited as part of a larger longitudinal study (Holmbeck & Devine, 2010). The current study included youth report of condition-related

knowledge and medical self-management skills. Youth attention and executive functioning was assessed via parent- and teacher-report and performance-based assessment.

Youth condition-related knowledge increased in a linear manner over time. While performance did not predict growth in condition-related knowledge, better youth performance on working memory and attention tasks predicted a higher intercept for condition-related knowledge at T1. Teacher report of inattention was predictive of a lower intercept for condition-related knowledge at T1, but an increased slope of knowledge growth. Teacher report of executive dysfunction was predictive of a higher intercept for condition-related knowledge and a lower trajectory of growth. Parent report of inattention and executive dysfunction were not predictive of youth condition-related knowledge intercept or slope. Slope of condition-related knowledge was predictive of youth self-management skills. Condition-related knowledge partially mediated the association between youth attention, parent- and teacher-reported youth inattention, and parent-reported executive dysfunction, with variable directionality.

Results indicate that youth with SB gain condition-related knowledge over time. However, executive dysfunction may impede gains in condition-related knowledge and may benefit from developmentally appropriate intervention. Condition-related knowledge partially mediates the association between some aspects of executive dysfunction and medical self-management skills. Executive functioning supports and psychoeducation programs may be mechanisms for intervention, though further research assessing family and cultural factors is needed.

CHAPTER ONE

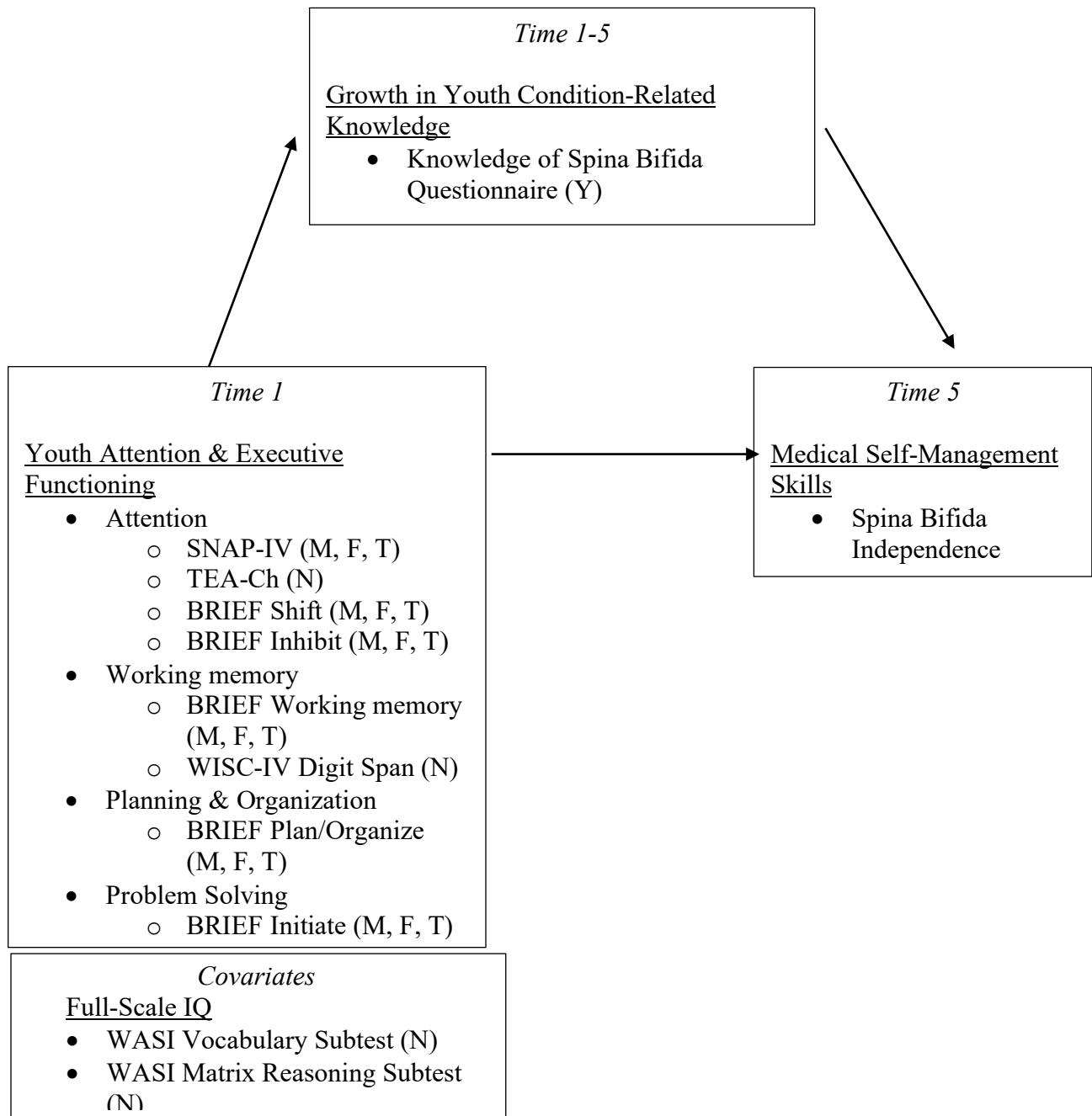
INTRODUCTION

Spina bifida (SB) is the most common birth defect affecting the central nervous system and is characterized by a variety of physical, medical, and neuropsychological complications (Copp et al., 2015). Individuals with SB may face ambulation problems, neurogenic bladder and bowel, hydrocephalus, skin injuries, and cognitive deficits, all of which require day-to-day management, including the following tasks: catheterization, a bowel program, and skin checks. Failure to master appropriate skills for the management of these tasks may contribute to secondary medical complications, which may be harmful and expensive to treat. While self-management skills are typically gained in adolescence as individuals become more independent, there is little known about the transition from childhood to adolescence in SB; moreover, gains in these skill areas may be complicated by cognitive deficits (Greenley et al., 2006; Rose & Holmbeck, 2007; Wasserman & Holmbeck, 2016).

Despite evidence in other pediatric populations (cystic fibrosis, rheumatoid arthritis, diabetes) that condition-related knowledge is an important component of independent medical management, this relationship has received little empirical attention in the context of SB (Greenley et al., 2006; Lansing & Berg, 2014). Additionally, little is known about factors that may influence gains in condition-related knowledge in this population. Therefore, the current study examined: (1) trajectories of growth in condition-related knowledge in youth with SB across the transition from childhood to adolescence, (2) cognitive and executive functioning as

predictors of trajectories of growth in condition-related knowledge, (3) growth in condition-related knowledge as a predictor of independent self-management skills, and (4) trajectories of growth in condition-related knowledge as a mediator of the relationship between cognitive/executive dysfunction and independent medical self-management skills in youth with spina bifida.

Existing literature fails to adequately characterize condition-related knowledge in youth with SB and empirical support for the possible role of condition-related knowledge in subsequent gains in independent medical management skills has not been forthcoming. Thus, the goal of the current study was to explore developmental trajectories of growth in condition-related knowledge. These trajectories will also be examined in relation to executive function and independent medical self-management in adolescence, aligned with research examining models of condition-related knowledge in other pediatric populations. This developmental period is particularly relevant because of increasing independence and the decreasing role of parents and/or caregivers across many domains. The current study will utilize a longitudinal research design, data from multiple informants, including parents, teachers, and youth, and performance-based neuropsychological measures. The following sections include a review of current research on medical self-management in youth with SB, an overview of condition-related knowledge in pediatric populations, and a summary of current literature regarding executive dysfunction in youth with SB. Additionally, a description of the current study, including objectives and specific hypotheses, is included.



Note. Reporters for each measure denoted by the following letters: M = mother, F = father, T = teacher, Y = youth self-report, N = neuropsychological assessment.

Figure 1. Mediating Role of Trajectories of Condition-Related Knowledge Growth on the Association Between Cognitive and Executive Functioning and Medical Self-Management Skills

CHAPTER TWO

REVIEW OF RELEVANT LITERATURE

Medical Self-Management Skills in Youth with Spina Bifida

As youth transition into adolescence and young adulthood, they gain independence across many domains of life. In typically developing individuals, this may include a gradual increase in responsibility, overall autonomy, and decision-making. Autonomy skills, including dressing, transportation, cooking, planning, and financial activities, have been shown to be acquired two to five years later for youth with SB as compared to typically developing peers (Davis et al., 2006). Differences in acquisition of autonomy skills can be attributed, at least in part, to differences in cognitive, rather than physical, ability. For individuals with a chronic medical condition, this developmental period may also include the transition from pediatric to adult medical providers, in addition to increases in self-management of medical tasks (Coyne et al., 2019). The importance of appropriate medical management is paramount; ineffective self-management may contribute to reduced adherence to prescribed treatment regimens which may, in turn, result in secondary complications, compromised health, and poor quality of life (Modi et al., 2012). Notably, evidence suggests that individual self-management declines as youth enter adolescence, additionally impacted by executive functioning and degree of condition-related knowledge (Modi et al., 2012; Psihogios et al., 2015).

In the context of SB, overall adherence to medical regimens in adolescence tends to be variable, due to normative developmental changes and the high number of complex medical

tasks to be completed (Psihogios et al., 2015). For youth with SB in particular, medical tasks on a day-to-day basis may include regular catheterization, a bowel program, monitoring skin for signs of pressure injury, and monitoring for shunt complications. Medical providers report that, for youth with moderate levels of condition severity, performing independent self-catheterization and independently carrying out skin monitoring tasks are attainable during the elementary school years, while independent bowel program management is attainable during middle school (Greenley, 2010). In the context of increased condition severity, however, independent self-catheterization was rated by providers as attainable during the middle school years, while completing daily skin checks and independent completion of bowel programs were rated as attainable after high school (Greenley, 2010).

That said, most youth with spina bifida are observed to gain at least some medical responsibility across late childhood and adolescence, though several factors contribute to the trajectory of responsibility acquisition (i.e., females with higher IQs, from families with low family stress tend to gain more responsibility more quickly; Kayle et al., 2020; Stiles-Shields et al., 2021). Other factors thought to contribute to effective self-management, improved adherence, and acquisition of medical responsibility include treatment knowledge, adaptive health beliefs, social skills, and coping skills (Modi et al., 2012; Stiles-Shields et al., 2021). It is also important to note that transfer of responsibility without corresponding skill acquisition may contribute to non-adherence to medical regimens and avoidable secondary complications (Psihogios et al., 2015). Cognitive ability, age, social skills, and appropriateness of increasing levels of responsibility should be considered when developing interventions to bolster independent medical management skills. Interventions of this type are few in number and randomized control

trials in the SB population are limited. Characterizing cognitive and knowledge-based factors that contribute to self-management skills may be beneficial in advancing this field.

Condition-Related Knowledge in Youth with Spina Bifida

Condition-related knowledge is a construct that encompasses an individual's understanding of their medical condition, including etiology, symptoms, complications, and management strategies (Johnson, 1984). Improved condition-related knowledge appears to bolster coping, self-efficacy, treatment adherence, and psychosocial adjustment in pediatric populations, but research is extremely limited in the context of SB (Greenley et al., 2006; Johnson, 1984; Strömfors et al., 2014). That said, research in other populations indicates that condition-related knowledge deficits in specific domains may be common in youth with chronic illness and may be linked to condition management (Greenley et al., 2006; Ievers et al., 1999). Additionally, gains in condition-related knowledge are expected in adolescence in other pediatric populations (i.e., diabetes), aligning with a period of increasing independence (Johnson, 1984). Developmental sequelae may occur later in spina bifida, however, due to potential cognitive deficits in this population. Indeed, an intervention to support independence in youth with spina bifida had limited success in facilitating increases in condition-related knowledge, with a lower level of cognitive functioning identified as a possible limitation (O'Mahar et al., 2010).

Importantly, deficits in one area of condition-related knowledge may not reflect deficits in other domains of knowledge. Because SB involves such an extensive range of medical complications, domain-specific knowledge may be a particularly important factor in understanding the role of condition-related knowledge in self-management skills and functioning (i.e., etiology versus management of complications). Domains of primary interest in SB include

knowledge of SB etiology, knowledge of functional status of individuals with SB (including potential complications), and knowledge of shunt functioning (specific to those with shunted hydrocephalus; Greenley et al., 2006). Each of these domains is captured in the Knowledge of Spina Bifida questionnaire, one of the only measures available for assessing condition-related knowledge in this population (Greenley et al., 2006; Wills et al., 1993).

In the few studies of condition-related knowledge in SB, youth tend to report a lack of accurate understanding across these domains (Greenley et al., 2006; Strömfors et al., 2014). Specifically, youth with SB report improved accuracy of knowledge pertaining to SB etiology across late childhood and early adolescence. A similar improvement was noted in certain areas of the functional status domain, including toileting and ambulation. Youth with shunts also demonstrated improvements in understanding of shunt functioning across this time period (Greenley et al., 2006). In another sample, youth with SB demonstrated satisfactory knowledge about bladder management, but knowledge about etiology and shunt functioning was lacking (Strömfors et al., 2014). This latter study indicated that youth were generally aware that they lacked knowledge pertaining to their condition but had little interest in learning more (Strömfors et al., 2014). This lack of motivation to acquire additional knowledge may be a useful focus for interventions that seek to improve condition-related knowledge in youth with SB.

Neuropsychological Function in Youth with Spina Bifida

Isolating predictors of growth trajectories in condition-related knowledge may be important in determining which factors are intervenable and may result in subsequent increases in future levels of knowledge. Neuropsychological function has been identified as an important component in understanding risk and resilience with regard to chronic illness self-management

during adolescence (Lansing & Berg, 2014). While self-management skills are often gained during adolescence for the typically developing population, cognitive deficits associated with SB may complicate the process of gaining independence due to challenges with self-regulation. Of note, adolescents managing chronic illnesses must regulate their thoughts and emotions about their condition and their medical management tasks, all of which may vary as a function of executive function and attention (Lansing & Berg, 2014). Like other domains of functioning, neuropsychological function in youth with SB is highly variable across individuals and is often associated with lesion level, hydrocephalus, and shunt status. As such, the neuropsychological profile in youth with SB is complex, with notable deficits across intelligence, memory, attention, and language domains (Dennis et al., 2006; Rose & Holmbeck, 2007). Deficits are thought to stem from central nervous system insults intrinsic to SB, and are thus persistent from birth through the lifespan, making cognitive challenges an important factor in understanding potential risk and resilience factors across development (Dennis et al., 2006).

Broadly, intelligence has been identified as a predictor of independence, academic achievement, daily functioning, and quality of life in spina bifida (Wasserman & Holmbeck, 2016). In other pediatric populations (i.e., juvenile rheumatoid arthritis, diabetes), cognitive status is recognized as a significant predictor of condition-related knowledge (Berry et al., 1993) and successful transition to self-care autonomy (Wysocki et al., 1996). While individual cognitive function varies considerably across youth with SB, extant literature suggests that three broad clusters (“average to low average,” “extremely low to borderline,” and “broadly average with verbal strength”) of cognitive function may exist, with a large portion of youth with SB falling in the “Average to Low Average” range of cognitive function (Wasserman & Holmbeck,

2016). These findings point to cognitive function as an important component of the neuropsychological profile that may have implications for growth in condition-related knowledge and independent medical self-management.

Executive functioning is an assembly of higher order cognitive abilities including, but not limited to, planning, inhibition, self-regulation, and behavior organization (Eslinger, 1996). Congenital hydrocephalus, as observed in many cases of SB, may disrupt neural circuitry in brain regions essential to these processes, contributing to executive dysfunction in individuals with SB and hydrocephalus (Rose & Holmbeck, 2007). Indeed, youth with SB and arrested or shunt-treated hydrocephalus were found to perform worse than youth with SB and no hydrocephalus and typically developing youth on tasks assessing verbal and nonverbal IQ, visuospatial functions, explicit memory, motor skills, reading and mathematics achievement, and executive functioning (Hampton et al., 2011). Further, shunt status has been identified as a significant predictor of performance on attention and executive functioning tasks, and youth with SB, most of whom had been shunted, performed worse on these tasks than their typically developing peers, even when controlling for intellectual functioning (Rose & Holmbeck, 2007).

Planning abilities are another aspect of executive functioning on which youth with SB have been documented to demonstrate more impairment than their typically developing peers, especially in the context of novel situations (Rose & Holmbeck, 2007). Planning involves determining and executing problem-solving strategies in order to progress from an initial state to a goal state, which may be important in self-regulation processes supporting independent self-management (Lansing & Berg, 2014). In addition, working memory may play an important role in volitional aspects of the self-regulatory processes involved in the self-management of chronic

illnesses; this aspect of executive function may support maintaining and accessing relevant information to carry out the necessary tasks of self-management (Lansing & Berg, 2014). Parent report indicates that youth with SB have more impairment with regard to working memory compared to typically developing peers, though teachers did not report a similar finding, which underscores the importance of employing multiple reporters when assessing this domain of functioning (Rose & Holmbeck, 2007). Further, there is some evidence that youth with SB have impaired explicit memory (memory of factual information, intentionally recalled), and memory impairments in adults with SB are well-documented (Dennis et al., 2006). Deficits in each of these processes may have detrimental effects on independent medical self-management skills and may also hinder acquisition of condition-related knowledge throughout childhood and adolescence. Prior research indicates that attention, planning and organization, and cognitive shifting may play distinct roles in the transfer of medical responsibility to youth with SB, as compared to general executive dysfunction (Stern et al., 2018; Stern et al., 2020). These specific domains of executive functioning may be particularly influential on youth behavior, including independent medical self-management tasks. Specifically, cognitive shifting involves the ability to adapt to environmental changes (i.e., need for medical task completion), and planning and organization involves goal-oriented decision making (i.e., generating and executing medical self-management skills).

Youth with SB are also at risk for specific attention-related dysfunction, much of which has been attributed to differences in midbrain structures and posterior cortex (Dennis et al., 2006). In particular, research has identified challenges with the posterior brain attention system, which is implicated in tasks that involve focusing and shifting attention (Brewer et al., 2001).

Notably, the process of disengagement is observed to be particularly impaired, and may reflect midbrain malformations due to hydrocephalus, which may be variable across cases (Brewer et al., 2001). Similarly, research indicates that attention-orienting may be a challenge for youth with SB. Individuals with spina bifida demonstrate challenges with bottom-up control of attention (i.e., slower orienting to, and disengagement from, stimuli that capture attention when compared to typically developing peers; Dennis et al., 2006). Indeed, this deficit may be particularly relevant to the development of independent self-management skills, and particularly in adolescence; paying attention to symptoms that require self-management skills is essential for successful self-management of chronic conditions (Compas & Boyer, 2001; Lansing & Berg, 2014; Stern et al., 2018). In a similar way, attention challenges may pose difficulties to gaining condition-related knowledge over childhood and adolescence, as learning requires attention-orienting and focus (Brewer et al., 2001; Dennis et al., 2006). Further, this specific dysfunction may hinder intervention efforts, particularly if they are short-term (O'Mahar et al., 2010).

Gaps in Current Literature

Existing literature highlights the importance of independent medical management skills, especially in adolescence. Additionally, attention and executive functioning deficits in youth with spina bifida have been well-characterized (Dennis et al., 2006; Rose & Holmbeck, 2007; Wasserman & Holmbeck, 2016). Despite this evidence, significant gaps in the literature remain as the transition from childhood to adolescence in the context of SB is not well understood. While a few studies have examined condition-related knowledge in SB (Greenley et al., 2006; Strömfors et al., 2014), these studies have had few longitudinal assessment points and small sample sizes, restricting conclusions and generalizability. Further, although condition-related

knowledge has been characterized as an important precursor to adjustment, functioning, and independence, no studies have examined growth in knowledge over the course of late childhood and adolescence, nor the implications of such growth for medical self-management. Especially important are considerations of transition to adult healthcare. As children enter adolescence, medical responsibility typically begins to shift from caregivers to the adolescent themselves, making condition-related knowledge and medical self-management particularly important in maintaining adherence to medical regimens during this period. To address these critical gaps, this study aimed to examine executive functioning in relation to growth in condition-related knowledge over a time span of eight years and determine associations between such growth in knowledge and medical self-management skills.

The Current Study

The current study aimed to expand existing literature by characterizing growth of condition-related knowledge in children with SB as they transition from childhood into adolescence and young adulthood. This study examined neurocognitive and executive functioning variables as potential predictors of growth in condition-related knowledge in youth with SB. Moreover, the mediating effects of condition-related knowledge was examined for the relationship between executive function and medical self-management skills. Understanding more fully factors that explain variability in condition-related knowledge and medical skill independence is essential to creating developmentally appropriate interventions for youth with SB to support independence and appropriate medical care during the transition to young adulthood.

Furthermore, this study included several methodological strengths; it utilized a multi-method (i.e., questionnaires, performance-based assessments), multi-informant (i.e., parents, teachers, adolescents) approach, spanning five time points (i.e., eight years) and offered a more comprehensive examination of condition-related knowledge and medical management skills in this population than previously available. While studies of condition-related knowledge in pediatric diabetes, cystic fibrosis, and asthma are relatively common (Holmbeck & Devine, 2010), there is a comparative dearth of literature pertaining to this subject in youth with spina bifida. Extending this research to the SB population, a chronic illness population with widely varied medical and cognitive complications, is important for improving medical management and appropriate independence for youth with SB.

Study Hypotheses

The current study had four objectives. The first objective of this study was to determine the average trajectory of condition-related knowledge accuracy across eight years in our sample of youth with SB. It was hypothesized that overall SB condition-related knowledge will increase for youth over time (Hypothesis 1).

Because of well-established links between attention and executive function and condition-related knowledge in other pediatric populations, better attention and executive functioning was hypothesized to be predictive of more growth in condition-related knowledge (Hypothesis 2).

The third objective was to determine the ability of condition-related knowledge growth to predict independent medical self-management skills. Greater growth in condition-related

knowledge was hypothesized to be predictive of more independent medical management skills (Hypothesis 3).

The fourth objective of this study was to examine the mediating role of SB condition-related knowledge growth on associations between attention and executive dysfunction and medical self-management skills in youth with SB. It was hypothesized that attention and executive dysfunction at T1 will limit participants' gains in condition-related knowledge, which will subsequently limit independent medical self-management at T5 in youth with spina bifida (Hypothesis 4).

CHAPTER THREE

METHODS

Participants

Participants were recruited as part of a larger, ongoing longitudinal study examining neuropsychological and psychosocial functioning and family adjustment among youth with spina bifida (SB; Holmbeck & Devine, 2010). Families with a child with SB were initially recruited from four hospitals and a statewide SB association in the Midwest. Families were sent recruitment letters and approached for recruitment during regularly scheduled clinic visits. Eligible youth with SB were 8-15 years of age at initial recruitment, were diagnosed with SB (including myelomeningocele, lipomeningocele, and myelocystocele), were able to speak and read English or Spanish, had at least one primary caregiver involved in the study, and lived within 300 miles of the laboratory (to facilitate data collection via home visits).

A total of 246 families were approached to participate in the study, of which 163 agreed. Twenty-one families later declined or were unable to be contacted, and two families did not meet inclusion criteria. The final sample of participants who completed baseline assessments consisted of 140 youth with SB and their families (see Table 1). Families who participated at baseline were contacted again for follow-up time points every two years. The current study includes 130 families who had condition-related knowledge data at least one time point.

Table 1. Youth Demographic and Condition-Related Characteristics at Time 1

	Total	M (SD) or N (%)
Participants		140 (100%)
Age		11.43 (2.46)
Gender: female		75 (53.6%)
Race		
Caucasian		74 (52.9%)
African-American/Black		19 (13.6%)
Hispanic/Latino		39 (27.9%)
Asian		2 (1.4%)
Bi-racial		6 (4.3%)
Spina bifida type		
Myelomeningocele		122 (87.1%)
Lipomeningocele		15 (10.7%)
Myelocystocele		2 (1.4%)
Unknown/not reported		1 (0.7%)
Lesion level		
Thoracic		23 (16.4%)
Lumbar		69 (49.3%)
Sacral		41 (29.3%)
Unknown/not reported		7 (5.0%)
Shunt present		109 (77.9%)
IQ		85.75 (19.54)
Family SES		39.12 (16.09)

Note: Demographic information is based on entire sample of 140 youth with spina bifida (SB) who participated in study tasks at T1.

Procedure

The current study was approved by university and hospital institutional review boards. Informed consent from parents and assent from children under 18 years old, or consent from youth over 18 years old, as well as release of information to allow data collection from healthcare providers, teachers, and medical records were obtained. Trained research assistants collected questionnaire and interview data via approximately three-hour-long home visits, of

which there were two at T1, and one at subsequent time points. Families and youth received \$150, a t-shirt, and a pen at each time point as compensation for their participation.

Participants completed questionnaires in English or Spanish, depending upon the language spoken by the family, in addition to performance-based neuropsychological evaluation and recorded interviews. When necessary, due to lack of availability of previously published Spanish versions, questionnaires were translated into Spanish by Spanish-speaking research assistants. Research assistants read questionnaires aloud to participants when needed (e.g., according to participant request, reading difficulties). The current study utilized parent-, teacher-, and youth-reported questionnaire data, as well as performance-based neuropsychological data.

Measures

Demographics

Parents reported on family demographics at T1, including information on child age, gender, and ethnicity, in addition to parent education, income, and occupation to assess socioeconomic status (SES) with the Hollingshed Index of Socioeconomic Status (Hollingshed, 1975). Higher scores on this measure indicate higher SES.

Youth Illness Characteristics

Parents reported on youth illness characteristics via the Medical History Questionnaire (MHQ; Holmbeck et al., 2003) and condition-related data were also collected via medical chart reviews at T1. Information gleaned from the MHQ and medical charts include type of SB, lesion level (i.e., sacral, lumbar, or thoracic), and ambulation ability. Gross motor function was evaluated on a five-point scale with the Gross Motor Function Classification System for Spina

Bifida (GMFCS; Palisano et al., 1997). Level I indicates minimal limitations in gross motor functioning; Level V indicates significant gross motor functioning limitations.

Youth Intelligence

At T1, youth were administered the Vocabulary and Matrix Reasoning subtests of the Wechsler Abbreviated Scale of Intelligence (WASI). Scores were used to estimate a Full-Scale IQ (FSIQ), which was used as an indicator of intellectual function in the current study (Wechsler, 1999). The WASI Vocabulary subtest includes 42 items assessing verbal knowledge and expressive vocabulary. The Matrix Reasoning subtest, on the other hand, includes 35 items evaluating nonverbal fluid reasoning and general intellectual ability. The WASI as a whole is a valid and reliable measure of intellectual function in children.

Youth Neurocognitive and Executive Function: Questionnaires

Neurocognitive and executive function in youth with SB was evaluated via parent- and teacher-report and performance-based neuropsychological assessment at T1. This study examined domains of executive functioning thought to be related to condition-related knowledge:

Executive Functioning. Mothers, fathers, and teachers reported on youth executive functioning at T1 using the Behavior Rating Inventory of Executive Functioning (BRIEF; Gioia et al., 2000). Parents and teachers are instructed to indicate whether the child never, sometimes, or often demonstrates a particular behavior over the past six months. The BRIEF includes eight subscales of executive function, including Inhibit, Shift, Emotional Control, Initiate, Working Memory, Plan/Organize, Organization of Materials, and Monitor. These eight domains are further organized within the Behavioral Regulation index, which includes Inhibit, Shift, and

Emotional Control, and the Metacognition Index, which includes Initiate, Working Memory, Plan/Organize, Organization of Materials, and Monitor. Taken together, all subscales can be combined to yield a Global Executive Composite Score. For the purposes of this study, the Inhibit, Shift, Working Memory, Plan/Organize, and Initiate subscales were used.

Attention. Parents and teachers completed the Swanson, Nolan, and Pelham Teacher and Parent Rating Scale – Version IV (SNAP-IV; Bussing et al., 2008; James M. Swanson, 1992), which was used to evaluate youth inattention, impulsivity, and hyperactivity. The SNAP-IV consists of 18 items assessing DSM-IV (American Psychiatric Association, 1994) criteria for Attention-Deficit/Hyperactivity Disorder (ADHD). Parents and teachers rate how much each statement describes youth behaviors on a scale from 0 (*Not at All*) to 3 (*Very Much*). Higher scores indicate increased symptom severity, and items are subdivided into an inattention subscale and a hyperactivity/impulsivity subscale.

Youth Neurocognitive Functioning: Performance-Based Measures

Executive Functioning. Working memory was evaluated via youth completion of the Digit Span Forward and Digit Span Backward tasks on the Wechsler Intelligence Scale for Children – Fourth Edition (WISC-IV; Wechsler, 2003). As a whole, the WISC-IV is an assessment battery designed to evaluate cognitive function, specifically verbal comprehension, visual spatial processing, fluid reasoning, working memory, and processing speed, in youth ages 6-16 years of age. For Digit Span Forward, the child is asked to repeat numbers in the same order they are presented by the examiner. For the Digit Span Backward task, the child is asked to repeat the numbers in the reverse order from which they are presented by the examiner. Higher age scaled scores indicate better working memory function.

Attention. Attentional capacities were assessed with youth completion of subtests from the Test of Everyday Attention for Children (TEA-Ch; Manly et al., 2001). The TEA-Ch is an assessment battery that consists of tasks that evaluate three attentional capacities, including Selective/Focused Attention, Attentional Control/Switching, and Sustained Attention. The Sky Search task assesses selective/focused attention by asking the child to circle matching pairs of items as quickly as possible. The Score subtest assesses sustained attention, and the child must count the number of “scoring sounds” on an audiotape. The Sky Search DT task assesses sustained-divided attention, requiring the child to circle matching pairs of items while also counting the number of “scoring sounds” on an audiotape. Finally, the Score DT task assesses sustained-divided attention and requires children to count the number of “scoring sounds” on an audiotape, while also listening for the name of an animal in a news broadcast. TEA-Ch subtests demonstrate adequate test-retest reliability, construct validity, and concurrent validity (Manly et al., 2001).

Youth Knowledge of Spina Bifida

Youth completed the Knowledge of Spina Bifida (KOSB; Greenley et al., 2006) questionnaire at T1, T2, T3, T4, and T5 to indicate condition-related knowledge across three domains: (a) etiology of spina bifida, (b) functional impairments associated with spina bifida, and (c) shunt functioning problems. The questionnaire consists of 18-items and is answered in a free-response format. Six of these items did not assess youth factual knowledge regarding spina bifida and associated complications and, thus, were not included in present analyses. The remaining twelve items address the previously mentioned factual domains and are categorized by trained coders as correct (2 points), inconclusive (some elements of a correct response, but vague

or somewhat inaccurate; 1 point), or incorrect (0 points), based upon the clinical presentation of SB. Psychometric analyses indicate high levels of inter-rater agreement on all items (kappa mean = .79, range = .61-.96; (Greenley et al., 2006).

Self-Management Skills

Spina Bifida Skills. Youth with spina bifida completed the Spina Bifida Independence Survey (SBIS) at T5. This measure consists of 50 items assessing acquisition of youth spina bifida-related skills, which has been adapted from the Diabetes Independence Survey (DIS; Wysocki et al., 1996). Youth respond no (0 points), yes (1 point), not sure, or not applicable, to each spina bifida-specific skill. A ratio of independently mastered skills (i.e., the total of “yes” responses) to all skills that were deemed relevant by the respondent (i.e., the total of “yes” and “no” responses) was used as a measure of self-management skills. The DIS has acceptable internal consistency, construct validity, and concurrent validity (Wysocki et al., 1996).

Planned Analyses

Preliminary Analyses

Prior to conducting the primary analyses, the psychometric properties of all included measures were evaluated. Internal consistency data for questionnaire data and inter-rater reliability for the knowledge of spina bifida questionnaire was calculated. Pearson correlations were used to examine associations between reporters on measures with multiple informants (i.e., parent- and youth-report). Measures were collapsed across informants for measures in which data from two or more informants are significantly correlated with $r \geq .40$ (Holmbeck et al., 2002). Analyses for measures not significantly correlated with one another were conducted separately for each measure.

Primary Analyses

Some families who participated in the study at T1 declined to participate in some or all of the subsequent time points, which has led to some sample attrition across the five time points. Participants with incomplete data were retained for the current study, to preserve the largest sample possible for analyses. This sample of 130 participants provided adequate statistical power (.80) to detect large effects for the planned analyses ($\mu_s = .30$; Múthen & Múthen, 1998). More specifically, power analyses reveal generally adequate statistical power (.80) to detect large effects for analyses of predictive ability of neurocognitive variables on condition-related knowledge (Faul et al., 2007). Notably, parent composite scores had limited sample sizes following data consolidation, reducing statistical power for these analyses. Power analyses reveal adequate statistical power (.80) to detect large effects for predictive ability of condition-related knowledge on medical self-management skills (Faul et al., 2007). Finally, adequate statistical power (.80) to detect large effects for analyses of predictive ability of neurocognitive variables on medical self-management skills, was revealed (Faul et al., 2007). Again, parent composite scores limited sample size and thus statistical power.

Analytic Plan for Objective 1. Analyses were conducted in *Mplus* Version 8 (Múthen & Múthen, 1998). Full information maximum likelihood was used to address missing data, which accommodates missing data such that any participant with at least one time point will be included in analyses (Enders & Bandalos, 2001; Schafer & Graham, 2002). This approach to missing data allows for inclusion of as many participants as possible and reduces biases in analyses as compared to alternative approaches (Arbuckle, 1996; Collins et al., 2001; Little & Rubin, 1989). Latent growth curves were used to characterize changes in condition-related

knowledge over time. The observed variables were the repeated measure of condition-related knowledge, obtained at five different time points: baseline (T1), two years after baseline (T2), four years after baseline (T3), six years after baseline (T4), and eight years after baseline (T5). IQ was examined as a covariate in the models. Model fit was assessed after the addition of each covariate to determine whether inclusion of IQ significantly improves or worsens the fit.

Three variants of the Latent Growth Model were compared using likelihood ratio chi-square statistics: one with intercept only (no growth), intercept and slope (linear growth), and intercept, linear parameter, and quadratic parameter (non-linear growth), as would be expected in the case of a specific period of accelerated growth. Best fit was determined using the bootstrap likelihood ratio test (BLRT) and the Bayesian Information Criterion (BIC). Growth parameters and trajectories of this model were then examined after controlling for appropriate covariates.

Analytic Plan for Objective 2: Attention and executive functioning variables from Time 1 were included as predictors of trajectories of growth in condition-related knowledge across time points. Again, analyses were conducted in *Mplus* Version 8 (Múthen & Múthen, 1998). Best fit was determined using the Bayesian Information Criterion. Growth parameters and trajectories of predictive models were examined both with and without IQ as a covariate.

Analytic Plan for Objective 3: The best fitting trajectory of growth in condition-related knowledge was examined as a predictor of medical self-management skills at Time 5, both with and without IQ as a covariate. Regression analyses were conducted in *Mplus* Version 8 (Múthen & Múthen, 1998). Because of sample size constraints, weighted least square mean and variance adjusted estimators were utilized and models were bootstrapped to estimate model fit. Best fit was determined using the Bayesian Information Criterion.

Analytic Plan for Objective 4. Finally, mediation analyses were conducted to examine the mediating role of SB condition-related knowledge intercept and growth on associations between executive dysfunction and medical self-management skills in youth with SB. Due to sample size constraints, it was not feasible to use growth in condition-related knowledge as a mediating variable. As an alternative, condition-related knowledge at T3 was used as the mediating variable in models, as a proxy for level of condition-related knowledge over T1-T5. Because of sample size constraints, weighted least square mean and variance adjusted estimators were utilized and models were bootstrapped to estimate model fit. Separate models were used to examine whether condition-related knowledge mediated the association between each executive functioning variable and medical self-management skills.

CHAPTER FOUR

RESULTS

Preliminary Analyses

To limit the number of analyses conducted, associations among multiple reporters (e.g., mother, father, and teacher report) were examined for measures of attention and executive function to determine whether composite scores could be created.

Bivariate Correlations

Composite scores were generated based on planned aggregation criteria for parent ratings of inattention (see Table 1; $r = .41$). For measures of inattention, mother and father report of youth inattention was aggregated to generate a parent inattention variable ($r = .41$) at T1 (see Table 2). For measures of executive function, subscale scores on the BRIEF were highly correlated with one another ($r = .40 - .76$), so the general executive composite was used instead of individual subscales to reduce the number of analyses. Mother and father report of youth general executive functioning were aggregated to generate a parent general executive function variable ($r = .56$) at T1 (see Table 3). After aggregating parent ratings, parent composite scores were calculated for inattention and executive dysfunction, while teacher ratings remained separate. As such, youth performance on a working memory task and a sustained attention task, parent report of inattention, teacher report of inattention, parent report of executive dysfunction, and teacher report of executive dysfunction were used as predictor variables in the following analyses.

Table 2. Parent and Teacher Measures of Inattention

	1	2	3
1. Mother Report of Inattention	1		
2. Father Report of Inattention	.41***	1	
3. Teacher Report of Inattention	.15	.03	1

Note. Table 2 presents correlations for measures of inattention across reporters. * $p < .05$, ** $p < .01$, *** $p < .001$.

Table 3. Parent and Teacher Measures of Executive Dysfunction

	1	2	3
1. Mother Report of Executive Dysfunction	1		
2. Father Report of Executive Dysfunction	.56***	1	
3. Teacher Report of Executive Dysfunction	.35***	.21	1

Note. Table 3 presents correlations for measures of general executive function across reporters. * $p < .05$, ** $p < .01$, *** $p < .001$.

Hypothesis Testing

Growth models using *Mplus* Version 8 (Múthen & Múthen, 1998) were used to characterize changes in condition-related knowledge over time in youth with SB (objective 1). Additionally, growth models were used to examine whether attention and executive functioning variables predicted growth in condition-related knowledge (objective 2). Further, regression analyses were used to determine whether growth in condition-related knowledge predicted medical self-management skills in adolescence (objective 3). Finally, growth models were used to determine whether growth in condition-related knowledge mediated the association between attention and executive functioning and medical self-management skills in adolescence (objective 4). Given variability in cognitive functioning among individuals with SB, analyses

were run with and without IQ included as a covariate. Unless otherwise specified, variables did not significantly predict the slope.

Objective 1. Characterize the average trajectory of growth in condition-related knowledge in youth with SB across the transition from childhood to adolescence

Linear and Quadratic Growth Models

First, using data from five time points (T1–T5), growth models were conducted to examine change over time in KOSB (see Table 4). As expected, youth condition-related knowledge significantly increased with age across childhood and adolescence. Participants' knowledge of spina bifida significantly increased (i.e., knowledge was gained) over time in a linear manner at a rate of 1.48 units per year from a mean score of 2.10 at T1. The linear model fit was excellent (CFI = 0.99, RMSEA = 0.04). The linear model demonstrated better fit (BIC = 3018.25) than the quadratic model (BIC = 3034) (see Table 4).

Table 4. Condition-Related Knowledge Growth Models

Knowledge of Spina Bifida	No Predictor			
		CFI	BIC	RMSEA
Linear Model		.99	3018.25	.04
Intercept	2.10***			
Slope	1.48***			
Quadratic Model		.99	3034.00	.05
Intercept	1.92***			
Slope	-0.11 ^{n.s.}			

Note. Table 4 presents coefficients from growth models and indicates change in slope for each unit change of time point. ***p < .001, ^{n.s.}not significant

Objective 2: Examine attention/executive functioning variables as predictors of growth in condition-related knowledge

Next, the six attention and executive functioning variables (i.e., working memory, attention, parent report of inattention, teacher report of inattention, parent report of executive

dysfunction, teacher report of executive dysfunction) were entered as predictors into the models. Separate growth models were used to examine the relationship between each attention/executive functioning variable and growth in condition-related knowledge.

Neurocognitive Predictors of Growth in Condition-Related Knowledge, No Covariates

Working Memory

Without controlling for IQ, youth working memory as measured by digit span performance on the WISC-IV significantly predicted the intercept for knowledge of spina bifida at T1 (Table 5). Specifically, higher working memory performance predicted a higher intercept for youth condition-related knowledge at T1. The inclusion of working memory improved the fit of the linear model (BIC = 2857.33).

Attention

Youth attention as measured by the TEA-Ch significantly predicted the intercept for knowledge of spina bifida at T1 without controlling for IQ in the model (Table 5). Specifically, better performance on attention tasks predicted a higher intercept for youth condition-related knowledge at T1. The inclusion of attention as a predictor improved the fit of the linear model (BIC = 2747.40).

Inattention

Youth inattention as measured by parent report on the SNAP-IV, without controlling for IQ, significantly predicted neither the intercept nor the slope of growth in youth condition-related knowledge but did improve the fit of the model (see Table 5; BIC = 2128.25).

As measured by teacher report of on the SNAP-IV, however, youth inattention significantly predicted both the intercept for youth condition-related knowledge at T1 and the

slope over T1-T5. Specifically, more inattention predicted a lower intercept for youth condition-related knowledge at T1. Further, teacher report of more inattention significantly predicted an increased slope for youth condition-related knowledge over T1-T5, which was contrary to stated hypotheses. The inclusion of teacher report of inattention as a predictor improved the fit of the linear model (see Table 5; BIC = 2675.07).

Executive Dysfunction

Without controlling for IQ, youth general executive function as measured by parent report on the BRIEF did not significantly predict the intercept of condition-related knowledge at T1 (see Table 5). Parent report of executive function also did not improve the fit of the model (BIC = 4282.35).

Youth general executive function as measured by teacher report on the BRIEF, however, did significantly predict both the intercept of condition-related knowledge at T1 and the slope over T1-T5. Specifically, poorer executive function was associated with a higher intercept for youth condition-related knowledge at T1, contrary to hypotheses. In addition, poorer executive function at T1 significantly predicted a reduced slope for youth condition-related knowledge over T1-T5. Including teacher report of executive function as a predictor did not improve the fit of the model (see Table 5; BIC = 4617.88).

Table 5. Predictors of Growth in Condition-Related Knowledge (No Covariates)

Predictor	No Covariates			
		CFI	BIC	RMSEA
Working Memory		.98	2857.33	.06
Intercept	.40***			
Slope	-.03 ^{n.s.}			
Attention		.97	2747.40	.08
Intercept	.36***			
Slope	-.12 ^{n.s.}			
Inattention (parent report)		1.00	2128.25	.00
Intercept	-.07 ^{n.s.}			
Slope	.14 ^{n.s.}			
Inattention (teacher report)		1.00	2675.07	.03
Intercept	-.25**			
Slope	.33*			
Executive Dysfunction (parent report)		.00	4282.35	.47
Intercept	- ^a			
Slope	1.57 ^{n.s.}			
Executive Dysfunction (teacher report)		.00	4617.88	.92
Intercept	1.14***			
Slope	-.70***			

Note. Table 5 presents coefficients from growth models and indicates change in slope for each unit change of time point. * $p < .05$, ** $p < .01$, *** $p < .001$, ^{n.s.}not significant, ^aIntercept could not be computed due to very poor model fit.

Neurocognitive Predictors of Growth in Condition-Related Knowledge, Controlling for IQ

IQ

Youth IQ as measured by the WASI-IV Vocabulary and Matrix Reasoning subtests at T1 significantly predicted the intercept for condition-related knowledge at T1. Specifically, higher IQ predicted a higher intercept for youth condition-related knowledge at T1 but did not significantly predict the slope of gains in condition-related knowledge across T1-T5. The inclusion of IQ as a predictor improved the fit of the linear model (see Table 6; BIC = 2933.15).

Working Memory

Controlling for IQ, youth working memory as measured by digit span performance on the WISC-IV significantly predicted the intercept for knowledge of spina bifida at T1. Specifically, higher working memory performance predicted a higher intercept for youth condition-related knowledge at T1. The inclusion of working memory did not improve the fit of the linear model (see Table 6; BIC = 3995.50).

Attention

Youth attention as measured by the TEA-Ch did not significantly predict the intercept for knowledge of spina bifida at T1 when controlling for IQ in the model. The inclusion of attention as a predictor did not improve the fit of the linear model (see Table 6; BIC = 3851.82).

Inattention

Youth inattention as measured by parent report on the SNAP-IV, controlling for IQ, did not significantly predict the intercept for knowledge of spina bifida at T1. The inclusion of parent-reported inattention improved the fit of the linear model (see Table 6; BIC = 2944.03).

As measured by teacher report of on the SNAP-IV, youth inattention did not significantly predict the intercept for youth condition-related knowledge at T1. The inclusion of teacher report of inattention as a predictor did not improve the fit of the linear model (see Table 6; BIC = 3697.39).

Executive Dysfunction

Parent report of youth executive dysfunction on the BRIEF did not significantly predict the intercept for knowledge of spina bifida at T1 when controlling for IQ. The inclusion of parent

report of youth executive function as a predictor improved the fit of the linear model (see Table 6; BIC = 2831.34).

As measured by teacher report of youth executive dysfunction on the BRIEF did not significantly predict the intercept for knowledge of spina bifida at T1. The inclusion of teacher report of executive function as a predictor did not improve the fit of the linear model (see Table 6; BIC = 3567.64).

Table 6. Predictors of Growth in Condition-Related Knowledge (IQ as a Covariate)

Predictor	IQ Covariate			
		CFI	BIC	RMSEA
IQ		.96	2933.15	.09
Intercept	.47***			
Slope	-.20			
Working Memory		.86	3995.50	.15
Intercept	.42***			
Slope	-.09 ^{n.s.}			
Attention		.84	3851.82	.16
Intercept	.20 ^{n.s.}			
Slope	-.03 ^{n.s.}			
Inattention (parent report)		.95	2944.03	.09
Intercept	-.02 ^{n.s.}			
Slope	.15 ^{n.s.}			
Inattention (teacher report)		.91	3697.39	.12
Intercept	-.10 ^{n.s.}			
Slope	.27 ^{n.s.}			
Executive Dysfunction (parent report)		.94	2831.34	.09
Intercept	-.16 ^{n.s.}			
Slope	.23 ^{n.s.}			
Executive Dysfunction (teacher report)		.90	3567.64	.13
Intercept	-.18 ^{n.s.}			
Slope	.25 ^{n.s.}			

Note. Table 6 presents coefficients from growth models and indicates change in slope for each unit change of time point. * $p < .05$, ** $p < .01$, *** $p < .001$, ^{n.s.}not significant.

Objective 3: Examine growth in condition-related knowledge as a predictor of medical self-management skills in adolescence

Thirdly, regressions were used to assess the predictive utility of growth in condition-related knowledge on medical self-management outcomes at T5.

Effect of Condition-Related Knowledge on Medical Self-Management Skills

The direct effect of condition-related knowledge on youth medical self-management with IQ as a covariate was estimated by regressing the slope growth factor on the outcome variable. Results indicated that growth in condition-related knowledge over T1-T5 was a significant predictor of medical self-management skills at T5 ($\beta = .73, p < .001$). In other words, as rate of growth in condition-related knowledge across T1-T5 increases, independent medical self-management skills at T5 also increase. That said, the model fit was extremely poor (CFI = .00, RMSEA = .47, BIC = 5411.75).

Without IQ as a covariate, results of the regression analysis indicated that growth in condition-related knowledge over T1-T5 was a significant predictor of medical self-management skills at T5 ($\beta = .73, p < .001$). Similarly, as rate of growth in condition-related knowledge increases across T1-T5, independent self-management skills at T5 also increase. Model fit improved (CFI = .00, RMSEA = .51, BIC = 4223.55). Importantly, the small sample size of this study, especially at later time points, makes accuracy of predictive models particularly challenging, as fit statistics are based upon chi-square statistics, which are highly impacted by small sample sizes.

Objective 4: Characterize growth in condition-related knowledge as a mediator of the association between attention/executive function and independent self-management skills

Finally, mediation models were used to determine the mediating effect of growth in condition-related knowledge on the association between attention and executive functioning variables and medical self-management skills. Unfortunately, due to sample size constraints, growth in condition-related knowledge could not be used as a mediating variable. Instead, condition-related knowledge levels at T3 were utilized as the mediating variable, as a proxy for level of condition-related knowledge over T1-T5. Again, separate growth models were used to examine whether condition-related knowledge mediated the association between each executive functioning variable and medical self-management skills.

Working Memory

A mediation analysis was performed with youth working memory as measured by digit span performance on the WISC-IV as the independent variable, T3 condition-related knowledge as the mediating variable, and T5 medical self-management skills as the outcome variable. The indirect effect of youth working memory on medical self-management skills was not significant ($b = -0.06$, $SE = 0.08$, $CI = [-0.21 - 0.08]$). Although the indirect effect was not significant, model fit was good ($CFI = 1.00$, $RMSEA = .00$, $BIC = 1642.90$).

Attention

When a mediation analysis was performed with youth attention as measured by performance on the TEA-Ch as the independent variable, T3 condition-related knowledge as the mediating variable, and T5 medical self-management skills as the outcome variable, the indirect effect was significant ($b = 0.29$, $SE = 0.04$, $CI = [0.22-0.36]$). Model fit was good ($CFI = 1.00$,

RMSEA = .00, BIC = 1463.28). Better performance on an attention task at T1 predicted more condition-related knowledge at T3, which, in turn, predicted more self-management skills at T5.

Inattention

A mediation analysis was run with parent report of inattention as the independent variable, T3 condition-related knowledge as the mediating variable, and T5 medical self-management skills as the outcome variable. The indirect effect of parent report of inattention on medical self-management skills via condition-related knowledge was significant ($b = 0.29$, $SE = .05$, $CI = [0.20-0.38]$). Model fit was good ($CFI = 1.00$, $RMSEA = .00$, $BIC = 1565.55$). More problems with inattention per parent report at T1 predicted more condition-related knowledge at T3, which, in turn, predicted more medical self-management skills at T5.

When a similar analysis was conducted with teacher report of inattention as the independent variable, T3 condition-related knowledge as the mediating variable, and T5 medical self-management skills as the outcome variable, the indirect effect was significant ($b = 0.34$, $SE = .005$, $CI = [0.26 - 0.43]$). Model fit was good ($CFI = 1.00$, $RMSEA = .00$, $BIC = 1593.61$). Similarly, more problems with inattention per teacher report at T1 predicted more condition-related knowledge at T3, which, in turn, predicted more medical self-management skills at T5.

Executive Function

A mediation analysis was performed with parent report of executive function as the independent variable, T3 condition-related knowledge as the mediating variable, and T5 medical self-management skills as the outcome variable. The indirect effect of parent report of youth executive functioning on medical self-management skills via condition-related knowledge was significant ($b = 0.26$, $SE = .05$, $CI = [0.17, 0.35]$). Model fit was good ($CFI = 1.00$, $RMSEA =$

0.00, BIC = 1587.85). Parent report of more executive dysfunction at T1 predicted more condition-related knowledge at T3, which, in turn, predicted more medical self-management skills at T5.

When a mediation analysis was performed with teacher report of executive function as the independent variable, T3 condition-related knowledge as the mediating variable, and T5 medical self-management skills as the outcome variable, the indirect effect of teacher report of youth executive functioning on medical self-management skills via condition-related knowledge was not significant ($b = -0.12$, $SE = 0.08$, $CI = [-0.27, 0.03]$). Model fit was comparatively poor ($CFI = 1.00$, $RMSEA = .00$, $BIC = 1639.53$).

CHAPTER FIVE

DISCUSSION

Youth with SB may experience physical, medical, and neuropsychological complications as a result of their congenital CNS malformation (Copp et al., 2015). In order to prevent secondary complications, individuals with SB must conduct daily medical management tasks, including catheterization, bowel program management, and skin checks for pressure injuries. Self-management skills are typically gained during adolescence, but attention and executive dysfunction may disrupt gains in this area (Dennis et al., 2006; Greenley, 2010). Evidence from other chronic illness populations has also identified poor attention and executive functioning as a predictor of poor medical self-management in adolescents (Modi et al., 2012; Psihogios et al., 2015). Additionally, condition-related knowledge, which involves condition etiology, potential complication awareness, and understanding of functional limitations of a condition, has been identified as a contributor to independent medical self-management (Greenley et al., 2006; Lansing & Berg, 2014). Understanding of these constructs and their associations in the SB population is limited, however.

This study attempted to address these gaps in the literature via a longitudinal design by examining changes in knowledge of spina bifida across late childhood and adolescence. Considering prior research, it was hypothesized that youth with SB would gain condition-related knowledge over time (Greenley, 2006). In addition, this study examined associations between neuropsychological function (i.e., attention and executive function) and growth in condition-

related knowledge. Because SB may involve cognitive and executive functioning deficits, and these may impact learning processes in terms of gaining condition-related knowledge, it was hypothesized that poorer attention and executive functioning would predict less growth in condition-related knowledge over time. Further, extant literature suggests that improved condition-related knowledge may improve several factors, including treatment adherence and medical self-management (Greenley et al., 2006). Thus, this study sought to examine associations between growth in condition-related knowledge and medical self-management skills. It was hypothesized that greater growth in SB-related knowledge would predict increased medical self-management skills in adolescents. Finally, condition-related knowledge was examined as a mediator, such that attention and executive function deficits were expected to limit gains in condition-related knowledge, which were, in turn, expected to limit medical self-management skills.

Changes Over Time in Condition-Related Knowledge

Consistent with our hypothesis, findings indicated that youth with SB gain condition-related knowledge in a linear fashion as they age. These findings are consistent with extant literature in other pediatric populations (i.e., diabetes) that demonstrates gains in condition-related knowledge in late childhood and adolescence (Johnson, 1984). Further, these findings echo those demonstrated in one of the few longitudinal studies of condition-related knowledge in youth with SB, which also found that condition-related knowledge increases with age, but that such knowledge is lacking with regard to several important condition-related factors (Greenley, 2006).

Condition-related knowledge grew in a linear fashion across T1-T5, with better model fit than a quadratic model: knowledge grew at a consistent rate over time for youth with SB and there did not seem to be point at which condition-related knowledge increased more rapidly. This may be due to repeated exposure to condition-related information over time and the experience of secondary complications. Importantly, adherence to medical regimens tends to be variable during adolescence in the context of SB (Psihogios et al., 2015). These varying patterns may contribute to development of secondary complications, possibly adding to a youth's fund of condition-related knowledge through experience. In addition, there is currently no known program of intervention focused specifically on increasing condition-related knowledge in youth with SB, and psychoeducation regarding condition-related knowledge is not standard practice in many medical institutions. Instead, knowledge and perceptions about one's condition is typically gained through experience and pattern identification over time (Asnani et al., 2017), consistent with results from this study.

Impact of Attention and Executive Functioning on Condition-Related Knowledge

This study evaluated the impact of attention and executive functioning on growth in condition-related knowledge, both with and without controlling for IQ. Findings were partially consistent with hypotheses that neuropsychological functioning variables would predict growth in condition-related knowledge. Specifically, without accounting for IQ, better working memory and attention performance predicted a higher intercept for youth condition-related knowledge at T1 but were not predictive of slope of growth of condition-related knowledge. Interestingly, teacher report of more inattention predicted a lower intercept for youth condition-related knowledge at T1, but also predicted an increased slope of youth condition-related knowledge

across T1-T5. This was contrary to hypotheses and will be explored further in the discussion. Also contrary to hypotheses, teacher report of more executive dysfunction was predictive of a higher intercept for youth condition-related knowledge at T1. Aligned with hypotheses, however, teacher report of more executive dysfunction was predictive of decreased slope of youth condition-related knowledge over T1-T5. Parent ratings of inattention and executive dysfunction were not predictive of intercept or slope of youth condition-related knowledge.

The impact of attention and executive functioning on growth in condition-related knowledge was also assessed including IQ as a covariate in the model. Higher IQ predicted a higher intercept for youth condition-related knowledge at T1 but was not predictive of the slope of youth condition-related knowledge over T1-T5. When including IQ as a covariate in analyses, working memory performance was the only significant predictor of youth condition-related knowledge. Specifically, better working memory performance was predictive of a higher intercept for youth condition-related knowledge at T1 but did not significantly predict the slope of growth in condition-related knowledge. No other executive functioning variables were significantly predictive of either intercept or slope of youth condition-related knowledge when controlling for IQ. Because models including IQ did not demonstrate good fit compared to those without IQ as a covariate, models without IQ as a covariate will be interpreted.

The results of this study are partially consistent with previous work that has demonstrated variable results with regard to links between executive functioning and condition-related knowledge (Greenley, 2006). Importantly, aspects of executive dysfunction common in youth with SB may be instrumental in gaining condition-related knowledge. In particular, youth with SB often demonstrate challenges with planning, working memory, explicit memory, focus, and

attention shifting (Dennis et al., 2006; Rose & Holmbeck, 2007). Deficits in these processes may hinder acquisition of condition-related knowledge via disruption of learning processes. Further, in youth with SB, Strömfors et al. (2014) identified an important lack of motivation to learn more about their condition, which may be reinforced by disruptions in attention and executive functioning processes.

In the present study, better working memory and attention performance predicted higher intercepts of condition-related knowledge at T1, as expected. The fact that performance in these domains was not predictive of knowledge growth is consistent with some theories of developmental disorders in which youth with developmental disorders perform at a lower level at baseline and gain knowledge at a similar rate to peers (Francis et al., 1996). Importantly, executive function is thought to play a significant role both in concurrent learning and in trajectories of learning (Mazzocco & Kover, 2007). The finding that teacher report of more inattention was predictive of a lower beginning level of knowledge, but a higher rate of growth was contrary to hypotheses but aligns with a deficit model of learning disabilities, in which youth with learning disabilities may begin at a lower level and catch up to peers over time (Francis et al., 1996). It is also possible that inattention improves over time as children age and the measure of inattention at T1 does not fully capture functioning at later time points, which should be a focus of future research.

Teacher report of more executive dysfunction was predictive of a higher intercept for youth condition-related knowledge at T1 but a decreased slope of youth condition-related knowledge over T1-T5. This may be due to additional parent/caregiver support of youth with higher levels of executive dysfunction. Specifically, parents/caregivers of youth with more

executive dysfunction may (intentionally or not) provide additional hands-on support and scaffolding, artificially elevating condition-related knowledge when youth are younger. The beneficial effects of parent involvement and scaffolding may decline as children get older, resulting in reduced gains in condition-related knowledge over time (Treble-Barna et al., 2016). Finally, differences between parent and teacher report of inattention and executive dysfunction in predicting condition-related knowledge factors are striking. That said, parents and teachers often provide discrepant reports regarding children's executive dysfunction (e.g., Mares et al., 2007). This may be attributable to differences in expectations at home versus at school, as well as differences in perceptions of normative behavior according to parents versus teachers.

Impact of Condition-Related Knowledge on Medical Self-Management Skills

Growth in condition-related knowledge across T1-T5 was a significant predictor of independent medical self-management skills at T5, such that increased growth in condition-related knowledge was predictive of more independent medical self-management skills at T5. This finding was consistent with hypotheses. However, model fit statistics indicated that this regression model had poor fit to the data: the RMSEA was larger than typically acceptable values, indicating that the hypothesized model deviates from a perfect model (Xia & Yang, 2018). Because model fit statistics such as the RMSEA are based on the chi-square statistic, which is highly susceptible to changes in sample size, the small sample size for this study makes predicting outcomes accurately particularly challenging.

Importantly, condition-related knowledge seems to play a role in independent medical self-management skills in adolescents with SB, in line with extant literature. Specifically, condition-related knowledge has been shown to be linked to successful condition management in

cystic fibrosis (i.e., Ievers et al., 1999). This finding highlights the need for larger samples when examining questions of important factors that predict independent medical self-management.

Condition-Related Knowledge as a Mediator

When examining whether growth in condition-related knowledge mediated the association between attention and executive functioning and medical self-management skills, the number of participants was insufficient to conduct these analyses as planned. Instead, this study examined T3 condition-related knowledge as a proxy mediator of this association (instead of the growth variable). Importantly, this approach maintained the prospective components of the model, which was a strength of the study. Notably, this change contributed to the fact that mediation findings do not exactly echo findings from previous analyses evaluating predictive ability of neurocognitive variables. Still, when examining whether condition-related knowledge mediated the association between neuropsychological functioning and independent medical self-management skills, several significant results emerged.

First, T3 condition-related knowledge partially mediated the association between youth attention performance and independent medical self-management, such that better performance on an attention task predicted higher T3 condition-related knowledge, which predicted more medical self-management skills at T5. This finding aligns with the hypotheses and current research. Children who are better able to pay attention in childhood may be better able to learn about their condition over time by attending to relevant information, thus supporting more medical self-management skills in adolescence.

Second, T3 condition-related knowledge partially mediated the association between parent report of inattention at T1 and medical self-management skills at T5, such that more

inattention at T1 predicted more T3 condition-related knowledge, which predicted more medical self-management skills at T5. Similarly, T3 condition-related knowledge partially mediated the association between teacher report of inattention at T1 and medical self-management skills at T5, such that more inattention at T1 predicted more condition-related knowledge at T3, which, in turn, predicted more medical self-management skills at T5. Finally, T3 condition-related knowledge partially mediated the association between parent report of youth executive dysfunction at T1 and medical self-management skills at T5, such that more executive dysfunction at T1 predicted more condition-related knowledge at T3, which predicted more medical self-management skills at T5. These findings were unexpected and contrary to hypotheses but may suggest important insights regarding parent and teacher support for youth with neurocognitive dysfunction across contexts. As previously discussed, parent and teacher reports of executive dysfunction often vary due to differing behavioral expectations across contexts, which may partially explain the fact that T3 condition-related knowledge did not significantly mediate the association between teacher report of executive dysfunction and medical self-management skills (Mares et al., 2007). With regard to parent and teacher report of inattention and parent report of executive dysfunction and their impact on condition-related knowledge and medical self-management, while contrary to hypotheses, it is possible that additional parent/teacher scaffolding in the context of attentional deficits and executive dysfunction supports learning about one's condition, which may facilitate gains in independence in adolescence (Treble-Barna et al., 2016; Winning et al., 2020).

Interestingly, T3 condition-related knowledge did not mediate the association between youth performance on a working memory task at T1 and medical self-management skills at T5.

Working memory does play an important role in learning, but it is possible that working memory improves significantly over development, which is an important process of cognitive development and may be supported by normative brain maturation and/or the learning of additional strategies to support expansions in working memory capacity (Fitamen et al., 2019). Future research should examine the effect of working memory performance at different time points on gains in condition-related knowledge.

Strengths, Limitations, and Future Directions

This study had several strengths. It addressed important gaps in the pediatric literature, added to knowledge about changes in condition-related knowledge in youth with SB, and identified risk factors for limited growth in knowledge. Notably, this study used data collected from multiple sources (i.e., youth with SB, parents, teachers) to capture youth functioning across multiple environments (i.e., home, school). In addition, this study incorporated neuropsychological performance data for the purpose of evaluating youth attention and executive functioning. Finally, this study's longitudinal design allowed for examination of changes in condition-related knowledge over time, as well as chronological associations among attention and executive functioning variables and medical self-management outcomes. Notably, this study made use of five time points, spanning eight years, increasing the number of timepoints historically used in these analyses.

Importantly, this study also had a few limitations. The data analysis techniques utilized maximized the number of individuals included in analyses, even if they only had data at one time point. That said, analyses were generally underpowered, and we were unable to detect small and medium effects. As such, it was difficult to discern whether associations among variables truly

do not exist or whether they could not be detected due to a limited sample size. This was especially relevant in the mediation models, which were the most underpowered. Notably, because of limited sample size, T3 condition-related knowledge was used as a mediator rather than growth in condition related knowledge. Further, an examination of other potential predictors, such as spinal lesion level, was beyond the scope of this study, though these variables have been shown to be important in functional independence in youth with SB (Fletcher et al., 2005). A dataset collected from multiple sites would provide a larger sample size and would likely further our understanding the nuanced associations among condition-related and cognitive factors that may impact condition-related knowledge and independent medical self-management.

In addition, this study did not assess changes in specific domains of condition-related knowledge and instead evaluated knowledge as a whole, due to constraints associated with limited sample size. SB is an extremely complex disorder, with several varied areas of difficulty. Because of the potential risk for secondary complications as a result of lack of information and understanding of condition nuances, variability across knowledge in specific domains is important to characterize. Specifically, knowledge of spina bifida was assessed in the same manner across study participants, regardless of shunt status, due to sample size constraints. Future research should examine condition-related knowledge separately for youth with and without shunts and should examine growth in specific domains in order to more fully understand particular areas that may be more difficult for youth with SB to fully comprehend.

While important to evaluate, the present study used youth report of independent medical self-management skills at T5, which may have introduced some confirmation bias. Adolescents are generally reliable reporters (Herjanic et al., 1975), but may have reported aspirational levels

of medical self-management skills, rather than current levels, thus obscuring the study's results. It is also possible that bidirectional relationships exist between youth condition-related knowledge and independent medical self-management skills, such that youth self-management skills affect gains in condition-related knowledge. These reciprocal associations were not examined in this study and would be an important topic of future research.

This study also did not examine the effects of cultural factors, including language and acculturation status. For the SB population, cultural considerations are particularly important, as incidence rates are highest in Hispanic women, with roughly 3.80 of every 10,000 live births with SB (Data & Statistics on Spina Bifida, 2020). Language barriers and cultural expectations may contribute to challenges with doctor-patient and doctor-family communication and may result in differing expectations for adolescent autonomy. These contextual factors are the subject of much ongoing research, but implications for condition-related knowledge and medical self-management skills are largely unknown.

Conclusions and Clinical Implications

Even considering these limitations, the current study addressed several gaps in the SB literature. Findings suggest that youth with SB gain knowledge related to their condition over time, and that these gains are impacted by attention and executive functioning factors. Further, findings suggest associations among executive dysfunction, condition-related knowledge, and independent medical self-management skills over time. This study further elucidates the impact of executive dysfunction on independent medical self-management and brings to light several important areas of future research.

Executive functioning supports, including tutoring and digital programs, have been demonstrated to improve executive functioning in children with executive dysfunction unrelated to chronic illness conditions (Diamond & Lee, 2011). Some studies point to EF training as a potential intervention for youth with SB and associated executive dysfunction (Tuminello et al., 2012), but the evidence base is limited. Moreover, intervention strategies that support executive functioning and have been shown to be effective in the SB population should be considered. For example, goal management training (GMT), which addresses challenges with sustained attention in order to improve executive functioning, has been shown to lead to improved EF, coping, and quality of life, and decreases in mood and anxiety symptoms (Stubberud et al., 2015). Importantly, because of the variability in symptoms and severity among youth with SB, interventions must be tailored to individual needs and abilities. Further, a careful assessment of appropriate autonomy goals for individuals with SB should be carried out when beginning an intervention strategy.

Importantly, executive dysfunction can be monitored regularly with ongoing neurocognitive screenings. While historically limited by access to neuropsychological assessment professionals and resources, these services are becoming more common and accessible, particularly in the context of specialized medical clinics and academic medical centers. According to the results of this study, keeping healthcare providers informed regarding individual developmental course and specific executive functioning weaknesses may be important in assessing risk for challenges with gains in condition-related knowledge and establishing independent medical self-management. This study emphasizes the important and multifaceted role healthcare providers can play in supporting youth with SB, especially during

childhood and adolescence, around the time of expected gains in autonomy and the transition to adult healthcare. Executive dysfunction may also impact communication with providers and the ability to encode and recall condition-related information. Providers should be well-equipped to provide information about SB in multiple modalities and should reinforce information across multiple visits, consistent with models of effective transition to adult healthcare. Moreover, these considerations should go hand-in-hand with evaluations of health literacy and cultural considerations that may impact communication with medical providers and understanding of important condition-related information. Additional research examining the influence of family and cultural contexts is needed to provide the best possible care for youth with SB and their families.

APPENDIX A
MEASURES

Questionnaire Measures (Alphabetized):

Behavior Rating Inventory of Executive Function (BRIEF)

Knowledge of Spina Bifida Questionnaire (KOSB)

Spina Bifida Independence Survey (SBIS)

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

Direct Assessment Measures (Alphabetized):

Test of Everyday Attention for Children (TEA-Ch)

Wechsler Abbreviated Scale of Intelligence (WASI)

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

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VITA

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