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Development of Inattention and Executive Dysfunction in Youth with Spina Bifida: Condition Severity Variables as Predictors

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LOYOLA UNIVERSITY CHICAGO

DEVELOPMENT OF INATTENTION AND EXECUTIVE DYSFUNCTION IN YOUTH
WITH SPINA BIFIDA: CONDITION SEVERITY VARIABLES AS PREDICTORS

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

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ABSTRACT

Spina bifida (SB) is associated with neurologic impairments that increase individuals' risk for neuropsychological deficits, particularly inattention and executive dysfunction. While extant literature has yet to examine the development of inattention in youth with SB, some research suggests that these youth may not experience the age-related improvements in executive dysfunction seen in the general population. SB is a heterogeneous condition; thus, it is important to consider variability in condition severity when examining outcomes over time in youth with SB. Lesion level and shunt status are commonly used as indicators of SB severity and have been significantly associated with variability in neurocognition and other condition-related outcomes. Shunt revisions, which are also common in these youth and represent additional neurological insult, have also been found to be associated with parent-reported inattention and executive dysfunction in some research. Existing literature highlights a need for additional research on inattention and executive dysfunction and their development across time in youth with SB, as well as careful consideration of condition severity factors, including number of shunt revisions. Thus, this study aimed to characterize the development of inattention and executive dysfunction in youth with SB and examine the relationship between condition severity variables (i.e., lesion level, shunt status, and number of shunt revisions) and inattention and executive dysfunction at age 11.5 years and over time in these youth.

Participants included 140 youth with SB enrolled in a larger study, with data collected over five time points (Holmbeck & Devine, 2010). Medical history, including shunt status, number of lifetime shunt revisions, and lesion level, were collected via parent reports and medical chart review. Parents and teachers reported on youth's inattention and executive function using informant-based measures across the five time points.

Parents and teachers reported linear decreases in inhibition and working memory problems over time and stability in planning/organizing problems. In contrast, the development of inattention and shifting problems varied by reporter. At age 11.5 years, shunt status predicted worse inattention and executive dysfunction according to parents and teachers, while number of shunt revisions predicted worse parent-reported working memory alone. Surprisingly, lesion level predicted better parent-reported inhibition problems at 11.5 years. Lastly, number of shunt revisions and lesion level predicted worse parent-reported inattention and inhibition over time, respectively.

These findings suggest that inattention and executive dysfunction may significantly change over time in youth with SB, though this is dependent on context and reporter. This study also identified a relationship between condition severity variables and these cognitive constructs at age 11.5 years and over time. Early identification of deficits and implementation of interventions for youth with SB, particularly youth with increased condition severity, may result in better longitudinal outcomes. Results also contribute to the expanding literature on shunting practices and highlight the need for advancements in shunting techniques to improve surgical outcomes and, as a result, later neuropsychological functioning.

CHAPTER ONE

INTRODUCTION

Spina bifida (SB) is a common birth defect associated with multisystem deficits, including neurologic, orthopedic, and urologic impairments (Copp et al., 2015). Newborns with SB are surviving at increased rates, but these youth are left with lasting medical and associated complications (Oakeshott & Hunt, 2003; Yun & Kim, 2017). Neurologic problems in particular, such as Chiari II malformation and hydrocephalus, are common and place individuals with SB at increased risk for neuropsychological deficits. Specifically, inattention and executive dysfunction are common in youth with SB and have been associated with various outcomes, such as social and academic functioning, psychosocial adjustment, independence, and condition management (Heffelfinger et al., 2008; Kelly et al., 2012; Rose & Holmbeck, 2007; Stern et al., 2021; Tuminello et al., 2012).

Research conducted with typically developing youth suggests that levels of inattention and executive dysfunction may change across development, but are generally characterized by stability or improvements over time (Hart et al., 1995; Pingault et al., 2014; Vergunst et al., 2019; Vos et al., 2021). Understanding the development of these difficulties over time is essential for informing earlier identification of these deficits, intervention development, and prevention of negative long-term outcomes. While research has yet to examine the development of inattention in youth with SB, extant literature on executive dysfunction development in SB

highlights that these youth may not experience the same age-related improvements that are seen in youth without chronic health illnesses (Tarazi et al., 2008). This study aimed to fill these gaps in the literature by examining inattention and executive dysfunction over time in a sample of youth with SB.

SB is a heterogeneous condition; thus, it is important to consider variability in condition severity when examining outcomes in youth with SB. Condition severity may be understood as contributors of biological risk, or the combined impact of central nervous system (CNS) insults, which is associated with neurobehavioral and cognitive outcomes in many medical conditions (Brown et al., 2008; Dennis, 2000). Extant literature has primarily considered two variables as indicators of SB severity, particularly as they estimate degree of CNS impact: lesion level and shunt status. Both spinal lesion level and shunt status have been found to be associated with cognitive impairments in SB (Bier et al., 1997; Brown et al., 2008; Donders et al., 1991; Fletcher et al., 2005; Rose & Holmbeck, 2007; Snow et al., 1994; Tuminello et al., 2012; Wasserman & Holmbeck, 2016). However, some of the literature on this topic is mixed (Devito et al., 2005; Peterson et al., 2016; Raftopoulos et al., 1994; Roebroek et al., 2006), suggesting additional research is needed to elucidate the relationship between lesion level, shunt status, and neuropsychological functioning in SB.

While shunts have been found to be successful in treating hydrocephalus, shunt revisions are unfortunately common and require additional surgical intervention and neurological insult (Caldarelli et al., 1996; Norkett et al., 2016). Number of shunt revisions has been found to be associated with parent-reported executive dysfunction (Brown et al., 2008), as well as inattention and other attention difficulties (Brewer et al., 2001; Fletcher et al., 1996; Hommeyer et al.,

1999). However, more research is needed to understand the relationship between shunt revision history and neuropsychological functioning in youth with SB.

Overall, the current literature highlights a need for additional research on inattention and executive dysfunction and their development across time in youth with SB, as well as careful consideration of condition severity factors, particularly those capturing biological risk. Furthermore, the impact of shunt revision history on neuropsychological functioning in SB has largely been unexplored, despite high rates of revisions documented in the SB literature (Caldarelli et al., 1996; Norkett et al., 2016). Thus, this study aimed to characterize the development of inattention and executive dysfunction over time in a sample of youth with SB. Lesion level, shunt status, and number of shunt revisions were then examined as predictors of changes in these symptoms. The following sections describe the existing literature on the development of attention problems and executive dysfunction in SB, commonly studied SB condition severity variables, and current shunting practices in SB. Then an overview of the current study is provided, including study objectives and hypotheses.

CHAPTER TWO

REVIEW OF RELEVANT LITERATURE

Neuropsychological Functioning in Youth with SB

Spina bifida (SB) is the most common congenital birth defect, affecting 1 in every 2,758 births (Mai et al., 2019). SB results from failure of neural tube closure during embryonic development, thus significantly impacting the central nervous system (Copp et al., 2015). Individuals with SB experience lasting medical and associated effects that impact their development and quality of life (Oakeshott & Hunt, 2003; Yun & Kim, 2017). Neurologic complications, such as Chiari II malformation and hydrocephalus, are common in youth with SB and leave youth susceptible to neuropsychological deficits (Caldarelli et al., 1996; Dennis et al., 2006). Difficulties with attention and executive functioning in particular represent a core component of the typical neuropsychological profile of individuals with SB (Dennis et al., 2006; Wills, 1993).

Attention

Attention is a complex construct comprised of various domains, such as alertness/arousal, orienting, selective focusing, sustained attention, and shifting and divided attention. Youth with SB demonstrate difficulties with several domains of attention, such as attention orienting, selective focusing, and shifting (Brewer et al., 2001; Burmeister et al., 2005; Dennis et al., 2006; Loss et al., 1998). Sustained attention, on the other hand, remains relatively intact in these youth

(Brewer et al., 2001; Burmeister et al., 2005; Dennis & Barnes, 2010; Loss et al., 1998; Swartwout et al., 2008). Individuals with SB also exhibit deficits in attention orienting to external stimuli, which has been found to be related to dysfunction in posterior brain regions implicated by SB-related Chiari II malformations (Dennis & Barnes, 2010; Dennis et al., 2005a, b, c; Dennis et al., 2006; Rose & Holmbeck, 2007).

Inattention, a hallmark feature of Attention Deficit/Hyperactivity Disorder (ADHD), refers to difficulties in sustained attention, distractibility, and disorganization (ADHD; Larsson et al., 2011). The national prevalence of ADHD is estimated to be between 7-11%, though ADHD occurs at significantly higher rates in individuals with SB (Burmeister et al., 2005; Vissner et al., 2014; Wasserman et al., 2016). Within youth with SB, one study found that youth with SB who met criteria for ADHD presented with significantly more inattentive symptoms than hyperactive/impulsive symptoms (Ammerman et al., 1998). Another study demonstrated that 31% of participants with SB met criteria for ADHD, with 23% being inattentive type (Burmeister et al., 2005). These rates were significantly higher than those found in the comparison group and general population (Burmeister et al., 2005).

Youth with SB share some commonalities with youth with ADHD. For example, children with SB similarly exhibit difficulties with distractibility, organizing material, and staying on task (Burmeister et al., 2005). However, there are important differences between youth with SB and youth with ADHD. While symptoms may be similar across the two groups, implicated attention networks appear to differ. Attention difficulties in SB are associated with the dorsal attention network but are associated with the ventral attention network in youth with ADHD (De la Torre et al., 2017). One performance-based study comparing children with SB to children with ADHD-

only and a control group of youth without chronic illnesses highlighted this difference in attention network activation (Brewer et al., 2001). Specifically, youth with ADHD demonstrated difficulties in sustaining and maintenance of attention (associated with anterior attention networks), whereas youth with SB demonstrated difficulties in focusing and shifting functions (associated with posterior attention networks; Brewer et al., 2001).

Children with SB also generally do not exhibit hyperactive and impulsive behaviors, further differentiating these youth from the general ADHD population (Ammerman et al., 1998; Brown et al., 2008). This difference in prevalence of hyperactive and impulsive behaviors may be a result of mobility limitations present in SB (Ammerman et al., 1998; Brown et al., 2008). Moreover, differing etiologies and implicated brain regions may also contribute to these differences in hyperactive/impulsive symptoms (Ammerman et al., 1998). Despite these differences, it is critical to understand the presence of inattentive symptoms in youth with SB, as these youth may still benefit from interventions recommended for youth with ADHD targeting school and social functioning (Burmeister et al., 2005). Furthermore, inattention has also been found to be associated with medical responsibility, academic fluency, and sleep disturbance in individuals with SB (Cirino et al., 2019; Murray et al., 2016; Stern et al., 2021), and with internalizing and externalizing behaviors, educational attainment, and social functioning in youth without chronic health conditions (Larsson et al., 2011; Pingault et al., 2014; Solanto et al., 2009). These findings thus highlight the need for increased understanding of inattentive symptoms and their development, especially in SB.

Development of Inattention in Youth with ADHD and Youth Without Chronic Health Conditions. Research on the development of inattentive symptoms in youth with ADHD

and youth without chronic health conditions is generally inconsistent—some studies report stability, while others report reductions or increases in these symptoms over time (Döpfner et al., 2015; Vergunst et al., 2019). No existing research has examined inattention over time in youth with SB. However, one study examined sluggish cognitive tempo (SCT; now cognitive disengagement syndrome (CDS; Becker et al., in press)), an inattention-like construct, in youth with SB and found that it increases over time (Smith et al., 2021). These findings point to the relevance of considering inattention specifically over time in this population. While no research has explored inattention in particular over time in SB, examining development in youth with ADHD and in youth without chronic health conditions may provide insight into trajectories of inattention in SB.

Increase. A limited amount of research suggests that inattentive symptoms increase over time. Larsson et al. (2011) aimed to describe the independent and joint development of hyperactivity/impulsivity and inattention from childhood to adolescence and examined 1,450 twin pairs over 12 years. Results from this study demonstrated a general increase in inattention symptoms over time, as measured by a parent-reported symptom checklist. The authors suggest these findings support those from Lahey et al. (2005), which reported a later onset of ADHD-inattentive type and a general shift from ADHD-combined diagnosis to ADHD-inattentive type diagnosis over time in a sample of youth with ADHD.

Reduction. Contrary to literature on the increases in inattention symptoms in the general population, more recent research suggests that inattention symptoms may decrease over time. A recent study (Liu et al., 2019) followed twin pairs over time from age 8 to 16 and found a decrease in inattention using the Conners' Parent Rating Scales-Revised. These findings

replicated those found in an earlier study using the same sample, which demonstrated a linear decrease in inattention over 11 years (Pingault et al., 2015). However, the mean decrease in inattention symptoms in this earlier study was still less pronounced than the decrease in hyperactivity/impulsivity symptoms (Pingault et al., 2015). Similarly, Biederman et al. (2000) found a general reduction in inattention over time in youth with ADHD ages 6 to 20, with the reduction being less pronounced than that of hyperactivity/impulsivity symptoms. While the overall sample studied by Pingault et al. (2015) saw a decrease in inattention symptoms, it is important to note that some participants did report an increase in inattention. This is consistent with the findings presented by Larsson et al. (2011) and Lahey et al. (2005), suggesting an increase in the prevalence of the inattentive subtype later in development.

Nonetheless, other studies have also documented a decrease in parent-reported inattention symptoms over time, including a study examining community-based samples of ADHD ages 7-11 (Musser et al., 2016). Additional studies using other informants, such as teachers, have also reported reductions in inattention, with the largest declines occurring between 9th and 10th grades (Evans et al., 2013). Lastly, using DSM-IV-TR criteria, Döpfner et al. (2015) aimed to identify subgroup trajectories for ADHD symptoms. While inattention symptoms decreased over time in this sample overall, the developmental trajectory of inattention for youth with high levels of inattention was more stable over time (Döpfner et al., 2015).

Stability. Several studies have documented support for a stable course of inattention symptoms across childhood and adolescence. One study examining males with ADHD aged 7 to 12 years at baseline over the course of 4 years found that, despite an initial decline in inattention symptoms between years 1 and 2, inattention symptoms remained relatively stable over the

course of the study (Hart et al., 1995). This initial decline was not hypothesized to be indicative of the developmental course of inattention, as there was no age effect (Hart et al., 1995).

Findings from another study assessing ADHD symptoms from infancy to adolescence suggested an initial increase in inattention between ages 1.5 to 3.5, with symptoms then being stable through adolescence (Vergunst et al., 2019). Other studies using teacher- and parent-report measures have also identified varying trajectories of inattention symptoms, including stable-low, stable-high, rising, and declining trajectories, with stable trajectories describing over 50% of the samples' inattention development in these studies (Pingault et al., 2014; Vos et al., 2021).

These findings overall suggest that while there is variability in the developmental trajectories of inattention symptoms, particularly earlier in development, inattention is likely best characterized as remaining stable or decreasing through childhood and adolescence and into young adulthood. While these findings may provide some insight as to how inattention develops over time in SB, differences in brain activation between youth with ADHD and youth with SB may result in important distinctions in the development of inattention between these two populations. One study investigated the temporal development of the dorsal attention network (DAN; implicated in youth with SB) and ventral attention network (VAN; implicated in youth with ADHD) across 7 to 12-year-olds and 18 to 31-year-olds (Farrant & Uddin, 2015). This study found that children demonstrated increased within-DAN functional connectivity, whereas adults demonstrated greater within-VAN functional connectivity, thus highlighting potential asymmetrical development of the DAN and VAN (Farrant & Uddin, 2015). As these attention networks are differentially implicated in SB and ADHD, individuals with SB may not experience

the same age-related stability or decreases in inattention seen in youth with ADHD and in the general population.

Executive Functioning

Executive function (EF) is an overarching construct used to capture goal-oriented and control functions employed by the prefrontal cortex (Best et al., 2009; Eslinger, 1996). EF generally lacks conceptual clarity in extant literature, resulting in a variety of definitions of these functions (Klenberg et al., 2001). Components typically encompassed within the larger umbrella of EFs include working memory, inhibition, shifting/cognitive flexibility, and planning (Anderson, 2002; Best et al., 2009; Best & Miller, 2010; Miyake et al., 2000). Given these various components, there is a lack of agreement regarding whether EF is a set of independent components versus a unitary construct (Best & Miller, 2010; Miyake et al., 2000). Miyake et al. (2000) proposes acknowledging both the “unity and diversity” of EFs, as EFs are both distinct and interdependent.

While neuropsychological functioning varies within the SB population due to heterogeneous neurological impairment, executive dysfunction is common in individuals with SB (Kelly et al., 2012; Wasserman & Holmbeck, 2016; Zabel et al., 2011). Impairments in EF are well-documented, particularly vulnerabilities in working memory, cognitive flexibility, and planning (Brown et al., 2008; Mahone et al., 2002; O’Hara & Holmbeck, 2013; Rose & Holmbeck, 2007). Several studies have examined these impairments using performance-based measures of EF and have found consistent difficulties with mental flexibility, cognitive abstraction, problem-solving, and planning (Heffelfinger et al., 2008; O’Hara & Holmbeck, 2013; Rose & Holmbeck, 2007; Snow, 1999; Snow et al., 1994; Tuminello et al., 2012). These

deficits are maintained even after controlling for IQ (Rose & Holmbeck, 2007; Snow et al., 1999).

Performance-based measures are effective in examining EFs in a controlled setting but may fail to capture the role of EF in everyday functioning and may not provide a comprehensive understanding of a child's EF abilities (Brown et al., 2008; Gioia et al., 2000a; Gioia et al., 2010; Isquith et al., 2005). On the other hand, rating scales of EF, such as the Behavior Rating Inventory of Executive Function (BRIEF; Gioia et al., 2000a; Gioia et al. 2000b), are better able to measure "real world EF" (Isquith et al., 2005). At the most complex level, existing literature illustrates that performance-based and parent reports of EF capture different aspects of EF (Anderson et al., 2001; Bodnar et al., 2007; Huizinga & Smidts, 2010; Mahone et al., 2002). It is therefore essential to consider impairments in executive function within the context of parent-, teacher-, and self-reported day-to-day EF, as well as performance-based assessments, although the former may be more practical when attempting to track the unfolding of EF over time.

A considerable amount of research has examined executive dysfunction in SB using questionnaire measures, such as the BRIEF. The BRIEF is comprised of 3 indices: Behavioral Regulation Index (BRI; inhibit, shift, and emotional control subscales), Metacognitive Index (MCI; initiate, working memory, plan/organize, organization of materials, and monitor subscales), and Global Executive Composite (all subscales) (Gioia et al., 2000a; Gioia et al., 2000b). Extant literature has found mixed results regarding EF impairments across these indices in SB. Some studies report worse metacognition in youth with SB compared to controls, but fewer to no differences in behavioral regulation (Brown et al., 2008; Kelly et al., 2012; Mahone et al., 2002). Conversely, other studies have reported increased dysfunction in SB across all areas

of EF assessed (Burmeister et al., 2005; O'Hara & Holmbeck, 2013; Tarazi et al., 2008; Tuminello et al., 2012). These mixed findings call for additional research examining these functions in individuals with SB.

Current literature suggests that differences in executive dysfunction in individuals with SB are associated with well-documented impairments in processing speed and a specific profile of inattention rather than deficits in prefrontal abilities (Burmeister et al., 2005; Fletcher et al., 1996), which is consistent with the posterior brain involvement in SB. Moreover, attentional control plays a significant role in the executive profile of SB, particularly given the relationship between affected posterior attention and frontal executive brain regions (Jurado & Rosselli, 2007; Kelly et al., 2012). It is thus not surprising that studies have documented associations between inattention and EF in SB (Burmeister et al., 2005). Nonetheless, given unique associations between EF and various outcomes in youth with SB, it is still important to examine EF specifically in this population. Careful consideration must be taken to determine differing clinical implications of inattention and EF components such as working memory, inhibition, shifting, and planning in SB.

Overall, these findings suggest that youth with SB demonstrate elevated levels of executive dysfunction compared to normative and control samples. Understanding how these difficulties develop and/or change over time is important, given known associations between EF and social and academic functioning, psychological adjustment, functional independence, autonomy, and condition management in SB (Heffelfinger et al., 2008; Kelly et al., 2012; Rose & Holmbeck, 2007; Stern et al., 2021; Tuminello et al., 2012).

Development of Executive Dysfunction in Youth Without Chronic Health

Conditions and Youth with SB. Research on EF development has been inconsistent and disproportionately focused on the emergence of EFs during the preschool years rather than considering long-term trajectories of these functions (Best et al., 2009; Best & Miller, 2010). Childhood and adolescence are widely regarded as important developmental periods for executive functions, due to the maturation of prefrontal brain regions later in adolescence (Anderson et al., 2001; Best & Miller, 2010; Kalkut et al., 2009). Understanding EF development across the school-age years and beyond is therefore of considerable importance, as such research may: (1) contribute knowledge about the development of EF, (2) delineate the significant changes across various domains of EF that occur across adolescence, (3) promote better identification of atypical EF development, and (4) elucidate how developmental changes in experiences across childhood and adolescence impact everyday executive functioning (Anderson et al., 2001; Best et al., 2009).

Much of the research on EF growth has been conducted using performance-based measures. Studies using such measures have demonstrated that EF begins to develop during infancy but is strengthened throughout childhood and adolescence (Best & Miller, 2010; Garon et al., 2008). Specifically, this research suggests that inhibition develops early and quickly compared to other EFs, with rapid improvements during early childhood and more modest increase throughout adolescence (Best et al., 2009; Best & Miller, 2010). Working memory and shifting abilities, on the other hand, are reported to demonstrate a more gradual, linear development over time (Best et al., 2009; Best & Miller, 2010; Kalkut et al., 2009). Planning abilities also appear to develop and mature later in childhood adolescence (Best et al., 2009).

Additional research has documented improvements in performance-based EF across adolescence and into young adulthood, thus supporting the need for more research examining EF development beyond the childhood years (Ferguson et al., 2021). While differences in frontal cortical thickness in youth with SB may place these youth at risk for atypical development of individual executive functions, no research has examined the development of performance-based EF in youth with SB (Juranek et al., 2008).

Limited research has explored the development of executive dysfunction using behavior rating scales, such as the BRIEF, in the general population. One such study, conducted by Huizinga & Smidts (2010), examined changes in EF across age in a Dutch sample of youth aged 5-18. Comparing age groups, this study found significant differences between groups on all scales comprising the Behavioral Regulation Index (inhibition, shifting, and emotion control), as well as working memory and the overall Global Executive Composite (Huizinga & Smidts, 2010). Specifically, elevated behavioral dysregulation was found in 5–8-year-olds compared to 9-11-year-olds, and in 12-14-year-olds compared to 15-18-year-olds (except Shifting in the latter comparison) (Huizinga & Smidts, 2010). Greater working memory problems were also reported in 5-8-year-olds compared to 9-11-year-olds, but not in other age group comparisons (Huizinga & Smidts, 2010). These findings therefore support examination of the development of executive dysfunction over time across childhood and adolescence, as well as considering the components of EF individually.

One study conducted by Tarazi and colleagues (2008) examined EF across adolescence in a sample of youth with SB and shunted hydrocephalus (MMH: myelomeningocele + hydrocephalus) compared to typically developing peers on the BRIEF. Using cross-sectional

data, this study found a significant age-by-group interaction: the comparison group demonstrated age-related improvements on the BRIEF (specifically, BRI subscales), while executive dysfunction remained stable and elevated in the MMH group (Tarazi et al., 2008). This suggests that youth with SB who have shunts may not exhibit age-related improvements in executive dysfunction over time exhibited in youth without chronic health conditions and supports research examining these symptoms over time in these youth (Tarazi et al., 2008). Additionally, a study examining parent- and self-reports on the BRIEF across two time points found that executive dysfunction in MCI and GEC domains persisted from adolescence to adulthood in SB (Zabel et al., 2011). Contrary to Tarazi et al. (2008), BRI difficulties were not maintained into young adulthood, suggesting the possibility of improvements in some aspects of EF over time (Zabel et al., 2011). More research is therefore necessary to elucidate the trajectories of executive dysfunction in this population, which would inform intervention timing and development, as well as promotion of independence in these youth (Tarazi et al. 2008; Zabel et al., 2011).

Spina Bifida Heterogeneity

When examining neuropsychological functioning in SB, it is critical to consider the heterogeneity of SB as a condition. SB is complex and marked by multisystem involvement, including varying levels of neurologic, orthopedic, and urologic deficits (Fletcher & Brei, 2010). The heterogeneity of SB has prompted interest in examining disease parameters, or condition severity variables, as predictors of various SB-related outcomes (Holmbeck & Faier-Routman, 1995; Hommeyer et al., 1999; Wallander et al., 1989a; Wallander et al., 1989b). Such research has indeed found significant associations between condition severity and proximal outcomes in SB, such as cognitive and physical functioning (Hommeyer et al., 1999). Extant literature

typically captures SB severity by considering spinal lesion level or shunt status, both of which indicate the degree of CNS involvement in SB. These variables thus reflect biological risk, or the “cumulative effect of primary and secondary CNS insults,” which has been found to impact neurobehavioral and cognitive outcomes (Brown et al., 2008; Dennis, 2014, p. 325).

Lesion Level

Spinal lesion level has been significantly associated with variability in neurocognition and other condition-related outcomes in SB (Copp et al., 2016; Leger, 2005; Lemanek et al., 2000; Pit-ten Cate et al., 2002). Lesion level refers to the location of the SB lesion on the spine, which has been referred to as a visible indicator of phenotypic diversity (Lemanek et al., 2000; Taylor et al., 2010). Typically, higher lesion level (e.g., thoracic) is associated with increased condition severity and worse outcomes (Fletcher et al., 2005). For example, studies have reported that increased cognitive impairments are more common in individuals with higher lesion levels compared to lower lesion levels (Bier et al., 1997; Brown et al., 2008; Donders et al., 1991; Fletcher et al., 2005; Wasserman & Holmbeck, 2016). However, some studies have conversely reported no relationship between lesion level and cognition (Roebroek et al., 2006).

Additionally, decreased functioning in other SB-related outcomes, including ambulation and bladder and bowel function, has also been associated with higher lesion levels (Lemanek et al., 2000). This is related to the larger amount of spinal cord involvement implicated in individuals with higher lesions compared to lower lesions. Other studies have further outlined relationships between upper lesion level and poorer academic skills, adaptive functioning, satisfaction with self-care, vocational/educational status, and independence (Barf et al., 2007; Fletcher et al., 2005; Verhoef et al., 2006).

Shunt Status and Shunt Revisions

Shunt status, or the presence of a ventricular shunt, has also been identified as a source of variability in SB-related outcomes (Brown et al., 2008; Tew & Laurence, 1975; Yeates et al., 1995). Shunting practices are common in SB, occurring in approximately 80-90% of youth with SB, as most of these youth are born with neurologic complications such as Chiari II malformation (Adzick, 2013; Copp et al., 2015; Dennis et al., 2006). Chiari II malformation is a structural defect of the posterior fossa, brain stem, and cerebellum resulting in hydrocephalus, which involves a build-up of cerebrospinal fluid (CSF) in the brain (Adzick, 2013; Caldarelli et al., 1996; Copp et al., 2015; Norkett et al., 2016). Hydrocephalus occurs in 60-95% of youth with SB (Adzick, 2013) and can be extremely dangerous and is a notable cause of morbidity and mortality in individuals with SB (Norkett et al., 2016). Surgical ventricular shunting was developed to treat hydrocephalus, as this procedure drains excess CSF and releases pressure in the brain (Norkett et al., 2016). While shunts are effective in treating hydrocephalus, complications such as shunt revisions are unfortunately common and represent additional surgical intervention (Caldarelli et al., 1996; Norkett et al., 2016).

Some research in normal pressure hydrocephalus suggests that shunting can improve neuropsychological functions, particularly memory and psychomotor speed (Devito et al., 2005; Peterson et al., 2016; Raftopoulos et al., 1994). However, additional literature points to a negative impact of shunting on other neuropsychological functions in youth with SB, including poorer executive functioning and attention (Rose & Holmbeck, 2007; Snow et al., 1994; Tuminello et al., 2012). One study found that shunt status was a significant predictor of performance-based tasks of planning and attention, as well as parent-reported measures of

sustained attention, working memory, planning and organizing, and initiation compared to typically developing peers (Rose & Holmbeck, 2007). Snow et al. (1994) further identified three subgroups of neuropsychological profiles of individuals with SB. The group with the most severe neurocognitive dysfunction, including deficits with planning and flexibility, also reported the highest rates of shunting (88%; Snow et al., 1994). It is important to note that, while there is some evidence that shunting may be associated with poor neurocognitive outcomes, shunted individuals may indeed demonstrate worse neurocognitive functioning if the hydrocephalus were to be left untreated. Extant literature has not yet examined the effects of shunting vs. non-shunting practices on neuropsychological outcomes, which would elucidate whether outcomes associated with shunting are, in fact, preferable to the impact of non-shunted hydrocephalus, at least neuropsychologically. Regardless, severity of the hydrocephalus and implications for morbidity and mortality would clearly take preference when making decisions on whether to shunt.

While the relationship between shunting and neuropsychological functioning is unclear, complications such as shunt revisions can result in decreased neuropsychological function due to additional surgical intervention (Iddon et al., 2004). Accordingly, number of shunt revisions has been found to significantly predict parent-reported difficulties with metacognition (planning, organizing, and problem-solving) in SB (Brown et al., 2008). These findings, however, are mixed throughout the literature, with some studies finding no significant impact of history of revisions on parent-reported EF (Tuminello et al., 2012). In addition to parent-reported measures, studies have also found that number of shunt revisions predicts dysfunction on performance-based measures of executive function in children with SB (Loss et al., 1998).

With regards to attention difficulties, some research suggests that children with shunted hydrocephalus demonstrate specific impairments with inattention (Fletcher et al., 1996). Studies have also found that children with shunted hydrocephalus have increased difficulties with focusing, sustaining, and shifting attention compared to typically developing peers (Brewer et al., 2001; De la Torre et al., 2017; Hommeyer et al., 1999; Loss et al., 1998). These difficulties with inattention, initiation, and shifting attention may be associated with hydrocephalus- and shunt-related damage to the posterior areas of the brain responsible for arousal and activation (Brewer et al., 2001; Fletcher et al., 1996). Therefore, while there are findings suggesting that a more complicated shunt history is related to impaired attention and executive dysfunction, more research is needed to further understand the impact of shunt revision history in youth with SB (Brown et al., 2008; Snow et al., 1994).

Gaps in Current Literature

While current literature has documented impairments in inattention and executive dysfunction in youth with SB (Brown et al., 2008; Burmeister et al., 2005; Kelly et al., 2012; Mahone et al., 2002; O'Hara & Holmbeck, 2013; Tarazi et al., 2008; Tuminello et al., 2012; Wasserman et al., 2016), notable gaps persist in better understanding these challenges, both cross-sectionally and over time. For example, findings regarding specific impairments within the domain of executive dysfunction has been mixed, thus calling for additional investigations of these difficulties in youth with SB (Brown et al., 2008; Burmeister et al., 2005; Kelly et al., 2012; Mahone et al., 2002; O'Hara & Holmbeck, 2013; Tarazi et al., 2008; Tuminello et al., 2012). With regards to the development of these symptoms, no research to date has examined the longitudinal development of inattention in SB. Some studies have indeed examined executive

dysfunction development in SB, though studies in both youth without chronic health conditions and with SB that are based on rating scale assessments of everyday EF are generally lacking. Further, only one study (Zabel et al., 2011) has examined executive dysfunction across multiple time points in SB, as others have instead used cross-sectional study designs (Huizinga & Smidts, 2010).

Overall, research in normative populations suggests that the developmental course of inattention may predict academic achievement and attainment, social functioning, and internalizing and externalizing symptoms (Larsson et al., 2011; Liu et al., 2019; Pingault et al., 2014; Pingault et al., 2011; Willoughby, 2003;). Studies of executive dysfunction have also documented associations with similar outcomes, as well as autonomy, independence, and condition management in youth with SB in particular (Heffelfinger et al., 2008; Kelly et al., 2012; Rose & Holmbeck, 2007; Stern et al., 2021; Tuminello et al., 2012). Therefore, the early identification of inattention and executive dysfunction concerns, as well as understanding their development over time in youth with SB, is critical and largely missing in existing literature.

Research on associations between condition severity factors contributing biological risk (i.e., lesion level and shunt status) and neuropsychological functioning has also been conducted but gaps are present in this literature as well. Mixed findings have been documented regarding the relationship between lesion level and cognitive functions (Bier et al., 1997; Brown et al., 2008; Donders et al., 1991; Fletcher et al., 2005; Roebroek et al., 2006; Wasserman & Holmbeck, 2016). Research on the effects of shunt status and shunt revision history on neuropsychological outcomes is similarly mixed and perhaps suggests differential impact of shunting on different cognitive functions (Brown et al., 2008; Devito et al., 2005; Peterson et al.,

2016; Raftopoulos et al., 1994; Rose & Holmbeck, 2007; Tuminello et al., 2012; Snow et al., 1994). Furthermore, despite high levels of shunt revisions across individuals with SB, existing literature fails to adequately measure and describe the impact of shunt revision frequency on cognitive and other outcomes in SB. The current study, therefore, aimed to address these gaps by examining the development of inattention and executive dysfunction over time in youth with SB, and considering various indicators of condition severity as predictors of this development.

The Current Study

This study aimed to first characterize the development of inattention and executive dysfunction in youth with SB. This study also addressed the relationship between condition severity variables (i.e., shunt status, number of shunt revisions, and lesion level) and inattention and executive dysfunction over time in these youth. Examining these factors longitudinally will contribute to increased understanding of neuropsychological function, as well as the involvement of CNS insult, or biological risk, in the neuropsychological development of youth with SB. This knowledge will aid with earlier detection of inattention and executive dysfunction and intervention development, which are of critical importance, given known associations of these difficulties with psychosocial, educational, and condition management outcomes.

These findings will also help elucidate the impact of condition severity and numerous shunt revisions on neuropsychological functioning, thus contributing to the expanding literature on shunting practices and implications of spina bifida heterogeneity. In doing so, this study highlights methodological challenges associated with measuring shunt revision history and provides recommendations for how future research might best capture this variable.

Additionally, this longitudinal study utilized five time points, which allowed for the use of

sophisticated statistical procedures to determine the development of inattention and executive dysfunction over time, as well as predictors of this development. This study, therefore, addressed limitations in existing literature, which has failed to examine inattention longitudinally in SB and has largely analyzed age differences in executive dysfunction using cross-sectional designs.

Study Objectives and Hypotheses

This study had three objectives. The first objective was to characterize the development of parent- and teacher-reported inattention and executive dysfunction over time in youth with SB and elucidate whether this development is linear or nonlinear. It was hypothesized that development of inattention and executive dysfunction would be linear. Additionally, based on extant literature in ADHD populations and differences in brain activation networks between youth with ADHD and youth with SB, it was hypothesized that inattention would worsen (i.e., increase) over time.

The second objective was to identify the relationship between measures of condition severity and measures of inattention and executive dysfunction in youth with SB. It was hypothesized that increased condition severity (i.e., presence of a shunt, higher number of shunt revisions, and higher lesion level) would predict more parent- and teacher-reported inattention and executive dysfunction in all domains at 11.5 years.

The third objective was to examine measures of condition severity as predictors of growth in inattention and executive dysfunction over time in youth with SB. It was hypothesized that shunt status, shunt revisions, and lesion level would predict growth in parent- and teacher-reported inattention and executive dysfunction in all domains across age. Specifically, greater

condition severity would predict increasing trajectories of inattention and executive dysfunction over time.

CHAPTER THREE

METHODS

Participants

Participants in this study included youth with SB enrolled in a larger longitudinal study of psychosocial adjustment and related family, peer, and neuropsychological factors in children and adolescents with SB, “The Chicago Healthy Adolescent Transition Study” (CHATS; Holmbeck & Devine, 2010). Participants were recruited from a statewide SB association and four hospitals in the Midwestern U.S. during clinic visits and using recruitment materials (e.g., letters).

Families who were interested in participating underwent initial screening, and inclusion criteria for participation at T1 included: (1) youth with diagnosis of SB, (2) 8-15 years old, (3) residence within 300 miles of Chicago, (4) absence of other comorbid, chronic medical or psychiatric conditions, and (5) ability to speak and read English or Spanish.

One hundred sixty-three children and families initially agreed to participate out of the 246 invited families. Twenty-one families were then excluded due to being lost to follow-up or later declined, and an additional two of the initial 163 families did not meet all inclusion criteria. The final study sample thus included 140 participating families with a child with SB (T1: 53.6% female; 53.5% Caucasian; $M_{\text{age}} = 11.43$ years; Table 1). Of the total sample of 140 participants, 109 were reported to have a shunt. Group differences on demographic and condition-related variables were examined between participants who were shunted and those who were not. No significant differences were found between participants with and without shunts for gender, race,

SES, IQ, and lesion level ($ps>.05$). Additional information regarding child demographic characteristics for the current sample is displayed in Table 1.

Table 1. Youth Demographic and Condition-Related Information Reported 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%)
Biracial	6 (4.2%)
SB 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	1 (0.7%)
Shunt Present	109 (77.9%)
IQ	85.75 (19.54)
Family SES	39.12 (16.09)

This study included data from five time points of the CHATS study (T1-5). T1 was considered the baseline assessment, and T2, T3, T4, and T5 occurred at 2-, 4-, 6-, and 8- year follow-ups, respectively. Parents and teachers completed questionnaires at each time point until the child participant turned 18. A portion (25%) of the sample turned 18 beginning at T3, thus resulting in a decline in parent and teacher reports at T3, which increased to 50% at T4, 75% at

T5, and 100% at T6. As the current study is interested in using parent- and teacher-reported inattention and executive dysfunction data, only T1-T5 were included in this study given no available parent or teacher data at T6. Of the 140 total participants who participated at T1, 110 participated at T2 (78.6%), 102 participated at T3 (72.9%), 93 participated at T4 (66.4%), and 98 participated at T5 (70.0%). Attrition across these time points was examined prior to running proposed analyses to determine whether there were differences between participants who returned for later time points versus those who did not.

Procedure

This project utilized data collected from the CHATS study, which was approved by the relevant university and hospital Institutional Review Boards. Parents and children over 18 years of age gave informed consent, and children between ages 12-17 provided assent prior to participation. At T1 of the CHATS study, participants underwent two three-hour study visits during which trained research assistants visited participants' homes to administer parent questionnaires, neuropsychological assessment, and other study procedures (e.g., videotaped interactions and interviews) as part of the larger study. At least one Spanish-speaking research assistant was present for study visits with predominantly Spanish-speaking families, and questionnaires were also translated into Spanish by native Spanish-speaking research assistants. After completing study visits and obtaining releases of information from families, research assistants conducted medical chart reviews to collect additional medical history data. Teacher-report questionnaires were also obtained. Participants were then recruited for subsequent time points every two years. Only one home visit was conducted beginning at T2. Throughout the course of the study, parents continued to complete questionnaires until the child participant

turned 18. Families received \$150, a t-shirt, and a pen to compensate them for their participation at each study time point.

Measures

Demographics

Parents completed a demographics form, reporting child age, gender, ethnicity, race, and other demographic variables at T1. Socioeconomic status (SES) was assessed using the Hollingshead Index of Socioeconomic Status (Hollingshead, 1975). This index uses parental education and occupation to determine SES, with higher scores reflecting higher SES (Hollingshead, 1975). SES was included as a covariate in analyses, given associations between SES and inattention and executive dysfunction in existing literature (Döpfner et al., 2015; Hampton et al., 2011; Noble et al., 2007; Vergunst et al., 2018).

Shunt History and Additional Medical History

Research assistants extracted information from participants' medical charts to acquire data on various medical variables, including shunt status, whether the participant had ever undergone a shunt revision (yes/no), total number of lifetime shunt revisions, and the dates on which shunt revisions occurred. Information on shunt revision dates was collected to clarify discrepancies on total number of revisions. Additional data on participants' medical history were also extracted from medical chart reviews, including lesion level (i.e., sacral, lumbar, thoracic, or cervical).

In addition to medical chart review, information on shunt history was also reported by mothers and fathers using the Medical History Questionnaire (MHQ; Holmbeck et al., 1998). This questionnaire asks parents to report on items related to numerous domains of medical

history. Data gathered from this questionnaire for the current study includes shunt status, whether their child had ever undergone any shunt revisions (yes/no), total number of lifetime shunt revisions, and shunt revision dates.

Thus, shunt data were collected using 3 sources: medical chart review, mother report, and father report. Given that these data were obtained from 3 sources, it was possible that discrepancies in number of shunt revisions existed among the various reporters. Indeed, upon initial review of the data on number of lifetime shunt revisions, various discrepancies were noted between reporters. These discrepancies included both disagreement among all three reporters and disagreement between 2 out of 3 reporters. Nonetheless, 54 out of the 109 youth with a shunt had agreement among all existing reporters (49.5%). For the remaining 55 participants, rules were established to resolve discrepancies in the reports of number of shunt revisions to determine one final value. Several factors and assumptions were considered when creating these rules: (1) the medical chart would not have reported a shunt revision that did not occur, (2) the medical chart may not reflect total number of shunts, as the child may have undergone a revision at another institution, and (3) it is impossible to be sure whether parents had full understanding of shunt revision versus shunt infection, or other neurological complications. Such rules were intended to prioritize: (1) medical chart data, primarily when the medical chart reported the highest number of shunts, and (2) the largest number reported when there was lack of any agreement among reporters. Table 2 details the rules for resolving these discrepancies, as well as example discrepancies for each rule and the number of participants that fell under each rule when determining a final value.

Table 2. Rules for Resolving Reporter-Based Discrepancies in Number of Shunt Revisions.

Rule	Number of Participants Under Each Rule
1) If all three reporters agree (or there are only 2 reporters and they agree), use that agreed upon value. Rule remains even if one reporter provides a range. <i>e.g., M=3, F=3, Med=3 → 3</i> <i>e.g., M=10, Med=9+ → 10</i>	N=54
2) If there is only one reporter, use that value. If the one reporter indicates a range, use the higher value. <i>e.g., F=6-7 → 7</i>	N=17
3) If any two reporters agree, use the value most agreed upon, even if the discrepant value is a higher value that is reported by the <i>mother or father</i> . Rule remains even if one report provides a range. <i>e.g., M=4, F=3, Med=3 → 3</i> <i>e.g., M=5, F=3-4, Med=3 → 3</i> 3a) However, if mother and father agree, but the <i>medical chart</i> provides a discrepant, higher value, prioritize medical data. <i>e.g., M=1, F=1, Med=2 → 2</i>	N=14 N _{3a} =5
4) If there are only 2 reporters and they disagree, use the higher value, regardless of the reporter. <i>e.g., F=3, Med=6 → 6</i>	N=13
5) If all 3 disagree use the highest value reported., regardless of reporter. <i>e.g., M=10, F=6, Med=5 → 10</i>	N=2
6) If there is ambiguity (i.e., if there appears to be a lack of understanding), two study team members reached consensus to determine the appropriate value. <i>e.g., M and F both reported no to “has your child ever had a shunt revision?” but indicated 2 revisions, Med=5 → 5</i>	N=4

Note. M = mother report, F = father report, Med = medical chart report

Neuropsychological Function

Youth completed a neuropsychological assessment, including a measure of intellectual functioning (IQ) at T1. Parents and teachers also reported on youth's inattention and executive dysfunction using questionnaires at all five time points. Table 3 lists the questionnaire subscales used as outcome variables in the current study.

IQ. Intellectual functioning was measured at T1 using the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999), a well-validated, performance-based measure of intelligence. Full-Scale IQ (Standard Score) was determined using scaled scores from the Vocabulary and Matrix Reasoning subtests. The Vocabulary subtest ($\alpha = .89$) is a 42-item measure of expressive vocabulary and verbal knowledge that captures crystallized and general intelligence (Wechsler, 1999). The Matrix Reasoning subtest ($\alpha = .92$) is a 35-item measure of nonverbal fluid reasoning and general intelligence (Wechsler, 1999).

Inattention. Parents and teachers reported on youth's inattention symptoms using the Swanson, Nolan, and Pehlam Teacher and Parent Rating Scale Version – Fourth Edition (SNAP-IV; Swanson et al., 2001). The SNAP-IV is an 18-item measure based on DSM-IV criteria for Attention-Deficit/Hyperactivity Disorder (ADHD) (American Psychiatric Association, 1994). Response choices are on a three-point Likert scale, with parents selecting 0 (Not at All), 1 (Just A Little), 2 (Quite A Bit), or 3 (Very Much) for each item as they pertain to their child. This measure yields two subscales: Inattention (items 1-9) and Hyperactivity/Impulsivity (items 11-19). Only the Inattention subscale was used for this study, which is calculated by averaging the first 9 items. Higher scores reflect greater parent-reported problems with inattention. Internal consistency for the SNAP-IV Inattention subscale was high across reporters in the current sample ($\alpha = 0.92-0.94$).

Executive Dysfunction. The Behavior Rating Inventory of Executive Function (BRIEF; Gioia et al., 2000a; Gioia et al., 2000b) was used to measure parent- and teacher-reported executive dysfunction. The BRIEF is a parent- and teacher-report questionnaire that yields eight subscales of executive functioning: inhibit, shift, emotional control, initiate, working memory,

plan/organize, organization of materials, and monitor. For all items (85 items for parents, 86 items for teachers), respondents are asked to indicate the frequency with which each item has been a problem for their child in the last 6 months (0 = Never, 1 = Sometimes, 2 = Often). The BRIEF has high internal consistency within a normative sample for parent and teacher reports ($\alpha = 0.80-0.98$), as well as high test-retest reliability ($r = 0.81$ for parents, $r = 0.87$ for teachers) (Gioia et al., 2000a). In the current sample, the BRIEF demonstrated satisfactory internal consistency across reporters and subscales ($\alpha = 0.74-0.94$). Based on review of the SB literature, the following subscales were included in the current study due to their relevance to executive functioning concerns in SB: inhibit, shift, working memory, and plan/organize. Raw scores were used to reflect raw increases or decreases in executive dysfunction over time, and means were calculated for the subscales. Higher scores reflect increased executive dysfunction.

Table 3. Mother-, Father-, and Teacher-Reported Subscales Used in the Current Study.

Domain	Subscale
Inattention	SNAP-IV Inattention
Executive Dysfunction	BRIEF Inhibit Shift Working Memory Plan/Organize

Planned Analyses

Preliminary Analyses

Prior to testing hypotheses, descriptive data were analyzed to check for outliers and skewness. Psychometric properties of the measures were also assessed. Attrition analyses indicated that participants who returned for time points T2-T5 did not differ from participants

who did not return on the following demographic and condition-severity variables: gender ($X^2 = .147-1.675$, $p = .196-.701$), race ($X^2 = 4.004-9.217$, $p = .162-.676$), SES ($t = -1.963-1.460$, $p = .052-.847$), IQ ($t = -1.440-1.229$, $p = .152-.870$), lesion level ($X^2 = .096-2.254$, $p = .324-.953$), SB type ($X^2 = .006-2.101$, $p = .147-.939$), shunt status ($X^2 = .018-1.719$, $p = .190-.894$), and number of shunt revisions ($t = -.435-.756$, $p = .451-.838$).

Primary Analyses

Participants with incomplete data across the five study time points were kept in analyses to maintain the largest possible sample size for primary analyses. Literature addressing statistical power in growth analyses using SAS indicated that the current sample size would have sufficient power (.80) in detecting large effects ($\mu_s = .30$; Zhang & Wang, 2009).

Analytic Plan for Objective 1. To determine the development of parent and teacher-reported inattention and executive dysfunction over time, mixed effects growth curves were estimated using SAS Proc Mixed (SAS Institute Inc.). Both linear and quadratic growth models were considered, and best fit was determined using the Akaike (AIC) and Bayesian (BIC) Information Criteria, as well as the AICC, a small-sample bias-adjusted form of the AIC (Hurvich & Tsai, 1989; SAS Institute Inc.). For these criteria, smaller values represent better model fit. Time was defined using participant age centered at the median age for T1, 11.5 years. Thus, analyses estimated growth in inattention and executive dysfunction across adolescence in the sample, rather than across arbitrary study time points. Using mixed effects models allowed for average intercept and slope estimation (i.e., fixed effects) and individual variability in intercepts and slopes (i.e., random effects).

To capture inattention and executive dysfunction across contexts, mother, father, and teacher reports were included in the current study. When significantly associated (i.e., $r \geq .40$), mother and father reports were combined into a single parent composite to reduce number of analyses. Following guidance from existing literature, executive dysfunction was analyzed separately by subscale. Thus, separate models were examined for the following outcomes: (1) parent-reported inattention, (2) teacher-reported inattention, (3) parent-reported executive dysfunction subscales, (4) teacher-reported executive dysfunction subscales. The Kenward-Roger degrees of freedom adjustment was used for estimating effect parameters to enhance approximations in a small sample (Chawla et al., 2014; SAS Institute Inc.). Restricted maximum likelihood (REML) methods were used to account for data missing at random (SAS Institute Inc.). As age was included in the models, REML used age to predict parameters for missing data, thus allowing data to be missing due to planned attrition (i.e., 25% of participants turning 18 at T3) rather than at random. Thus, the following analyses are still robust despite planned attrition. Models were re-fit to exclude slope random effects for models in which slope random effects variance was estimated to be zero.

Analytic Plan for Objective 2. To identify the relationship between measures of condition severity and inattention and executive dysfunction, shunt status, number of shunt revisions, and lesion level were entered as predictors into the models. For the purposes of objective 2, condition-severity predictors of intercept for the separate models were examined.

Analytic Plan for Objective 3. To examine measures of condition severity as predictors of growth in inattention and executive dysfunction over time, shunt status, number of shunt

revisions, and lesion level were examined as predictors of slope across age for the separate models.

CHAPTER FOUR

RESULTS

Preliminary Analysis

Descriptive analyses were conducted to check for outliers and skewness. Values larger than three standard deviations from the mean were considered outliers. Of the 109 participants with shunts, 82 reported having previous shunt revisions and 25 reported no revisions. This information was missing for 2 participants with shunts. Total number of revisions ranged from 0-40, with a mean of 3.86 and standard deviation of 6.60 revisions. The total number of revisions data were skewed, with a skewness value of 3.768. Specifically, three outliers were identified, with participants reporting 25, 40, and 40 total revisions. Following procedures conducted by Brown et al. (2008), number of total shunt revisions was transformed using a square root transformation prior to running analyses. This procedure reduced the skewness value to 1.291.

Bivariate correlations were conducted to determine whether mother and father reports of inattention and executive dysfunction could be collapsed into parent composites to reduce the number of analyses. Across inattention and all executive dysfunction subscales (inhibit, shift, working memory, plan/organize), mother and father reports were significantly correlated at T1, with Pearson's r values greater than 0.4 (Inattention: $r=.718, p<.001$; Inhibit: $r=.501, p<.001$; Shift: $r=.412, p<.001$; Working Memory: $r=.671, p<.001$; Plan/Organize: $r=.612, p<.001$). Thus, mother and father reports were combined to create composites of parent-reported inattention and executive dysfunction. No parent- and teacher-reported inattention and executive dysfunction

variables were found to be skewed (i.e., skewness values were all less than 2.00). Descriptive statistics for parent and teacher reports across all five time points can be found in Table 4.

Table 4. Descriptive Statistics for Parent- and Teacher-Reported Inattention and Executive Dysfunction Across Five Time Points.

	<u>T1</u> M(SD)	<u>T2</u> M(SD)	<u>T3</u> M(SD)	<u>T4</u> M(SD)	<u>T5</u> M(SD)
<i>Inattention</i>					
Parent	1.00(.59)	.98(.67)	.95(.69)	.84(.65)	.84(.68)
Teacher	1.23(.78)	1.16(.83)	1.11(.87)	.96(.69)	.98(.86)
<i>Inhibit</i>					
Parent	1.45(.38)	1.34(.36)	1.31(.33)	1.30(.31)	1.23(.31)
Teacher	1.32(.45)	1.30(.41)	1.29(.38)	1.23(.32)	1.22(.29)
<i>Shift</i>					
Parent	1.66(.35)	1.60(.39)	1.57(.39)	1.53(.39)	1.63(.46)
Teacher	1.43(.48)	1.40(.45)	1.47(.46)	1.41(.44)	1.56(.58)
<i>Working Memory</i>					
Parent	1.80(.42)	1.79(.48)	1.73(.56)	1.62(.47)	1.66(.49)
Teacher	1.87(.62)	1.82(.58)	1.78(.60)	1.68(.59)	1.69(.58)
<i>Plan/Organize</i>					
Parent	1.82(.42)	1.81(.46)	1.81(.56)	1.75(.46)	1.87(.47)
Teacher	1.88(.57)	1.83(.56)	1.84(.57)	1.81(.55)	1.76(.67)

Hypothesis Testing

Mixed effects growth curves were used to characterize the development of parent- and teacher-reported inattention and executive dysfunction over time in youth with SB (objective 1; SAS PROC Mixed; SAS Institute Inc.). To identify the relationship between measures of condition severity and inattention and executive dysfunction at 11.5 years (objective 2) and over time (objective 3), shunt status, number of shunt revisions, and lesion level were included as predictors in the models. SES was included as a covariate in all analyses examining condition severity variables as predictors (objectives 2 and 3). Due to an estimated slope random effects

variance of zero, the following models were re-fit to exclude slope random effects: (1) parent-reported shift without predictors, (2) parent-reported shift with shunt status as a predictor, and (3) parent-reported inhibit with lesion level as predictor.

Objective 1: Characterizing the Development of Parent- and Teacher-Reported Inattention and Executive Dysfunction Across Age, Without Predictors

To characterize the development of parent- and teacher-reported inattention and executive dysfunction over time, linear and quadratic growth models were compared using AIC, BIC, and AICC fit statistics. Consistent with hypotheses, linear models had better fit across all analyses. Specifically, all linear models had smaller AIC, BIC, and AICC values when compared to quadratic models. Linear models were thus used in all subsequent analyses.

Growth models were conducted for parent- and teacher-reported inattention, inhibit, shift, working memory, and plan/organize. Results of these growth models are presented in Table 5. According to parents, inhibition ($p < .0001$), shifting ($p = .003$), and working memory ($p = .008$) problems decreased over time, while inattention and planning/organizing remained stable ($p > .05$). According to teachers, difficulties with inattention ($p = .0003$), inhibition ($p = .007$), and working memory ($p = .005$) decreased over time, while shifting and planning/organizing remained stable ($p > .05$). These findings are inconsistent with hypotheses, as inattention was hypothesized to worsen (i.e., increase) over time. There was significant variability in individual slopes for parent-reported inattention ($p = .014$) and working memory ($p = .021$), as well as teacher-reported plan/organize ($p = .016$). No significant variability ($p > .05$) in individual slopes were found for parent-reported inhibit, shift, and plan/organize, and teacher-reported inattention, inhibit, shift, and working memory.

Table 5. Development of Inattention and Executive Dysfunction Across Age Without Predictors.

Outcome	Slope Estimate
Parent-Report	
Inattention	-0.016
Inhibit	-0.037***
Shift	-0.016**
Working Memory	-0.015**
Plan/Organize	0.007
Teacher-Report	
Inattention	-0.053***
Inhibit	-0.022**
Shift	-0.003
Working Memory	-0.030**
Plan/Organize	-0.020

Note. Results reflect unit change in slope for each one-year increase in age. * $p < .05$, ** $p < .01$, *** $p < .001$.

Objective 2: Examining Condition Severity Variables as Predictors of Inattention and Executive Dysfunction at 11.5 Years

Shunt status, number of shunt revisions, and lesion level were included as predictors in the growth curve models to examine the relationship between measures of condition severity and inattention and executive dysfunction at 11.5 years. SES was included as a covariate in these analyses. Intercept coefficients for these growth models are presented in Table 6.

Shunt Status. Shunt status was first entered as a predictor for models examining parent- and teacher-reported inhibit, shift, working memory, plan/organize, and inattention. With regards to parent reports, shunt status significantly predicted the intercept for inattention ($p = .011$), working memory ($p = .007$), and plan/organize ($p = .035$). Specifically, and consistent with

hypotheses, shunt status predicted worse inattention, working memory, and planning/organizing abilities at age 11.5. For teacher reports, shunt status significantly predicted the intercept for inattention ($p=.011$) and working memory ($p=.010$), with presence of a shunt predicting worse inattention and working memory at 11.5 years.

Number of Shunt Revisions. Next, number of shunt revisions was included as a predictor into the models. Number of shunt revisions significantly predicted the intercept for parent-reported working memory ($p=.046$). As hypothesized, more shunt revisions predicted increased working memory problems at age 11.5. Although number of shunt revisions did not predict the intercept for any teacher-reported outcomes, the intercept for teacher-reported working memory was also trending towards significance ($p=.065$).

Lesion Level. Lesion level was then included as a predictor into the models. Lesion level significantly predicted the intercept for parent-reported inhibit ($p=.006$) only, though not in the expected direction. Specifically, higher lesion level predicted fewer parent-reported inhibition difficulties at 11.5 years. Lesion level did not significantly predict the intercept for any teacher-reported outcomes.

Table 6. Intercept Coefficients for Growth Curve Models Examining Condition Severity Variables as Predictors of Inattention and Executive Dysfunction at Age 11.5 Years.

Outcome	Condition Severity Predictor		
	Shunt Status	Number of Shunt Revisions	Lesion Level
Parent-Reported			
Inattention	0.308*	0.023	0.008
Inhibit	-0.066	0.013	-0.121**
Shift	0.125	0.022	-0.031
Working Memory	0.242**	0.074*	-0.024

Plan/Organize	0.171*	0.040	-0.016
Teacher-Reported			
Inattention	0.394*	0.063	-0.006
Inhibit	0.028	-0.031	-0.037
Shift	0.155	-0.007	-0.009
Working Memory	0.302**	0.087	0.050
Plan/Organize	0.181	0.036	-0.010

Note. Results reflect unit change in intercept for each one-year increase in age, as predicted by each condition severity variable. * $p < .05$, ** $p < .01$.

Objective 3: Examining Condition Severity Variables as Predictors of Inattention and Executive Dysfunction Across Age

Shunt status, number of shunt revisions, and lesion level were included as predictors in the linear growth curve models (i.e., same models used in objective 2) to examine measures of condition severity as predictors of inattention and executive dysfunction over time in youth with SB. Slope coefficients for these growth models are presented in Table 7.

Shunt Status. Shunt status was first examined as a predictor of parent- and teacher-reported inhibit, shift, working memory, plan/organize, and inattention over time. Shunt status did not predict parent- or teacher-reported inattention or executive dysfunction over time ($p > .05$).

Number of Shunt Revisions. Number of shunt revisions was then examined as a predictor of parent- and teacher-reported inattention and executive dysfunction over time. Number of shunt revisions significantly predicted the slope for parent-reported inattention over time ($p = .038$) but did not predict the slope for any teacher-reported outcomes. Specifically, increased number of shunt revisions predicted more parent-reported inattention across age.

Lesion Level. Lastly, lesion level was considered as a predictor of inattention and executive dysfunction over time. Lesion level significantly predicted the slope for parent-reported inhibit ($p=.034$), with higher lesion level predicting increased inhibition problems with age. Lesion level did not predict the slope for any teacher-reported outcomes ($ps>.05$).

Table 7. Slope Coefficients for Growth Curve Models Examining Condition Severity Variables as Predictors of Inattention and Executive Dysfunction Across Age.

Outcome	Condition Severity Predictor		
	Shunt Status	Number of Shunt Revisions	Lesion Level
Parent-Reported			
Inattention	0.017	0.020*	-0.006
Inhibit	0.003	0.009	0.015*
Shift	-0.006	0.007	0.008
Working Memory	0.002	0.004	0.007
Plan/Organize	0.014	0.004	-0.002
Teacher-Reported			
Inattention	-0.032	0.003	0.017
Inhibit	0.005	0.006	-0.003
Shift	-0.001	0.014	-0.013
Working Memory	0.016	-0.005	0.004
Plan/Organize	0.012	-0.004	-0.001

Note. Results reflect unit change in slope for each one-year increase in age, as predicted by each condition severity variable. * $p<.05$.

CHAPTER FIVE

DISCUSSION

Youth with SB are at increased risk for neuropsychological deficits due to neurologic problems, including Chiari II malformation and hydrocephalus. Two domains of neuropsychological functioning in particular—attention and executive functioning—are especially relevant for youth with SB, given known associations with social and academic functioning, psychosocial adjustment, condition management, and independence (Heffelfinger et al., 2008; Kelly et al., 2012; Rose & Holmbeck, 2007; Stern et al., 2021; Tuminello et al., 2012). While research in the general population suggests that inattention and executive dysfunction change over time, little research has examined the development of executive dysfunction in youth with SB, and no research has considered inattention over time in this population (Hart et al., 1995; Pingault et al., 2014; Tarazi et al., 2008; Vergunst et al., 2019; Vos et al., 2021; Zabel et al., 2011).

As SB is a highly heterogeneous condition, it is necessary to consider variability in condition severity when examining outcomes longitudinally in youth with SB. Literature on youth with SB typically captures condition severity through lesion level and shunt status, which have been found to be valuable indicators of CNS involvement and biological risk in SB. However, the association between these measures of condition severity and cognitive outcomes is mixed throughout the SB literature (Bier et al., 1997; Brown et al., 2008; Devito et al., 2005; Donders et al., 1991; Fletcher et al., 2005; Peterson et al., 2016; Raftopoulos et al., 1994;

Roebroek et al., 2006; Rose & Holmbeck, 2007; Snow et al., 1994; Tuminello et al., 2012; Wasserman & Holmbeck, 2016). In addition, shunt revisions, or surgical repairs of shunts resulting in further neurological insult, are also very common. Despite high rates of shunt revisions in individuals with SB, little research has considered shunt revisions as a measure of condition severity. Additional research is thus necessary to examine shunt revisions as a measure of condition severity and highlight the impact of shunt revision frequency on neuropsychological functioning.

Thus, this study aimed to address gaps in the literature regarding the development of inattention and executive dysfunction across age in youth with SB, and associations between condition severity and inattention and executive dysfunction in this population. Additionally, this study aimed to determine the impact of shunt revisions, in particular, on inattention and executive dysfunction. To accomplish these aims, this study employed longitudinal methods to first characterize the development of parent- and teacher-reported inattention and executive dysfunction across age in a sample of 140 youth with SB. To elucidate the relationship between condition severity variables and inattention and executive dysfunction, measures of shunt status, number of shunt revisions, and lesion level were examined as predictors of these cognitive constructs at 11.5 years (the midpoint for age at Time 1). Lastly, to determine the impact of condition severity variables on the development of inattention and executive dysfunction, the assessment of shunt status, number of shunt revisions, and lesion level were examined as predictors of longitudinal changes in inattention and executive dysfunction across age. It was hypothesized that increased condition severity (i.e., presence of a shunt, higher number of shunt revisions, and higher lesion level) would predict more parent- and teacher-reported inattention

and executive dysfunction at 11.5 years, as well as worse inattention and executive dysfunction over time.

Inattention and Executive Dysfunction Over Time in Youth with SB

The first aim of this study was to examine the development of parent- and teacher-reported inattention and executive dysfunction over time in youth with SB. As hypothesized, linear models best characterized the development of parent- and teacher-reported inattention and executive dysfunction in the current sample. These findings are consistent with longitudinal research conducted in the general population, which has demonstrated linear trajectories of inattention and executive functioning over time (Best et al., 2009; Best & Miller, 2010; Kalkut et al., 2009; Pingault et al., 2015). According to parents, difficulties with inhibition, shifting, and working memory decreased over time, while inattention and planning/organizing remained stable. According to teachers, problems with inattention, inhibition, and working memory decreased over time, while shifting and planning/organizing remained stable.

Due to differences in the maturation of brain regions implicated in youth with SB and youth with ADHD, it was hypothesized that inattention may increase in youth with SB (Farrant & Uddin, 2015). Contrary to this hypothesis, inattention was reported to decrease according to teachers and remain stable according to parents. These results are consistent with findings documented in the general population and in youth with ADHD, as most research with these populations suggests stability or reduction in parent- and teacher-reported inattention problems over time (Döpfner et al., 2015; Evans et al., 2013; Hart et al., 1995; Liu et al., 2019; Musser et al., 2016; Pingault et al., 2014; Pingault et al., 2015; Vergunst et al., 2019; Vos et al., 2021). In contrast, these findings are inconsistent with a recent study examining sluggish cognitive tempo

(SCT; now cognitive disengagement syndrome (CDS; Becker et al., in press)) in this same sample of youth with SB (Smith et al., 2022). Unlike inattention, SCT (CDS) was found to increase over time in the current sample (Smith et al., 2022). This discrepancy illustrates distinctions between symptoms associated with inattention and CDS (Becker et al., in press). Furthermore, the disagreement between parent- and teacher-reported inattention suggests differences in the development and presentation of inattentive symptoms across home and school settings (Burns et al., 2016). Specifically, in the current sample of youth with SB, increased inattention challenges may be identified earlier within the classroom setting, followed by more noticeable declines in these symptoms over time per teacher reports.

Consistent with extant literature on performance- and reporter-based executive functioning, individual elements of executive dysfunction exhibited different trajectories over time (Best et al., 2009; Best & Miller, 2010; Kalkut et al., 2009). According to both parents and teachers, inhibition and working memory difficulties decreased over time, while planning/organizing problems remained stable. These findings are consistent Huizinga & Smidts (2010), which found stable planning/organizing and decreased inhibition and working memory problems on the BRIEF in a Dutch sample of typically developing youth. In a sample of adolescents and young adults with SB and shunted hydrocephalus, Zabel et al. (2011) similarly identified improvements in behavioral dysregulation over time. Conversely, Tarazi et al. (2008) demonstrated elevated, stable executive dysfunction across domains in youth with SB and shunted hydrocephalus. Differences in the findings between these studies involving youth with SB and shunted hydrocephalus highlight the need to consider predictors of executive dysfunction over time.

Despite similarities in findings across parent- and teacher-reported inhibition, working memory, and planning/organizing over time, the findings for shifting were inconsistent between reporters in this study. Specifically, parents reported decreased shifting problems, while teachers reported stability in shifting over time. Though differing from the findings for parent- and teacher-reported inattention, these findings similarly suggest differences in the development of shifting based on setting. One possible explanation is that youth are required to demonstrate cognitive flexibility earlier within the school setting due to programmed shifts in school day schedules, whereas shifting demands at home may increase with age. Therefore, youth may have increased opportunities to demonstrate shifting abilities at school compared to home. This may result in initially greater levels of shifting difficulties reported by parents, followed by greater declines in these problems. Overall, these findings indicate that youth with SB may indeed exhibit age-related improvements in some aspects of behavioral dysregulation and metacognition, such as inhibition and working memory, while planning/organizing challenges remain relatively stable across adolescence. On the other hand, the development of inattention and shifting problems appears to vary by reporter and, relatedly, setting (e.g., school and home).

Relationship Between Condition Severity and Inattention and Executive Dysfunction in Youth with SB at Age 11.5 Years

After characterizing the development of parent- and teacher-reported inattention and executive dysfunction over time, measures of condition severity (i.e., shunt status, number of shunt revisions, and lesion level) were examined as predictors of these cognitive constructs at 11.5 years and across adolescence. Shunt status significantly predicted worse parent- and teacher-reported inattention and working memory at 11.5 years, as well as worse parent-reported

planning/organizing. Number of shunt revisions also significantly predicted worse parent-reported working memory at 11.5 years but did not predict any teacher-reported outcomes. Overall, shunt-specific condition severity factors were more predictive of parent-reported outcomes at 11.5 years compared to teachers. These findings are generally consistent with literature documenting an association between shunt history and difficulties with executive functioning and attention (Rose & Holmbeck, 2007; Snow et al., 1994; Tuminello et al., 2012).

At 11.5 years, shunt status, but not number of shunt revisions, significantly predicted parent- and teacher-reported inattention and parent-reported planning/organizing. Therefore, while shunting may improve certain aspects of neuropsychological functions, such as memory and psychomotor speed (Devito et al., 2005; Peterson et al., 2016; Raftopoulos et al., 1994), youth with SB and shunted hydrocephalus are at increased risk for deficits in “real-life” executive functioning and attention. Furthermore, these findings highlight that general neurologic changes related to shunt status are significantly associated with inattention and planning/organizing challenges at 11.5 years. In contrast, working memory at 11.5 years was predicted by both shunt status and number of shunt revisions. This suggests that working memory abilities in particular may be more sensitive to repeated neurological insult (i.e., more revisions) compared to other domains of executive dysfunction. Overall, these findings indicate that early interventions targeting inattention, planning/organizing, and working memory are especially important for youth with SB with history of a shunt and shunt revisions.

Lastly, lesion level did not predict any teacher-reported outcomes, but significantly predicted parent-reported inhibition at 11.5 years. Contrary to hypotheses, higher lesion level (i.e., worse condition severity) predicted better inhibition at 11.5 years. In the existing literature,

lesion level has generally been associated with greater cognitive impairments (Bier et al., 1997; Brown et al., 2008; Donders et al., 1991; Fletcher et al., 2005; Wasserman & Holmbeck, 2016), though some research has reported no relationship between lesion level and cognitive outcomes (Roebroek et al., 2006). Unlike shunt status and shunt revisions, lesion level is a more global measure of SB condition severity, representing biological risk within both neurological and non-neurological domains. Individuals with higher lesion levels may thus require greater caregiver involvement related to their condition severity. Indeed, studies of associations between family relationships and lesion level have identified that higher lesion level is associated with increased maternal attachment, as well as increased willingness to grant autonomy (Holmbeck & Faier-Routman, 1995). Youth's inhibition at 11.5 years may therefore be influenced by the degree of caregiver involvement based on the child's lesion level.

Relationship Between Condition Severity and Inattention and Executive Dysfunction in Youth with SB Over Time

Shunt status, number of shunt revisions, and lesion level were then examined as predictors of changes in parent- and teacher-reported inattention and executive dysfunction across age. Shunt status did not predict any parent- or teacher-reported outcomes over time. As shunt status predicted various outcomes at 11.5 years, these findings suggest that shunt status may influence early levels of inattention and executive dysfunction but does not impact the developmental course of these constructs throughout adolescence and young adulthood. Number of shunt revisions also did not predict changes in parent- or teacher-reported executive dysfunction over time. These findings were not entirely unexpected, as extant literature regarding

the relationship between shunt revisions and reporter-based executive dysfunction has been mixed (Brown et al., 2008; Tuminello et al., 2012).

On the other hand, number of shunt revisions significantly predicted the slope for parent-reported inattention. Specifically, youth with more shunt revisions exhibited greater inattention problems across age. This suggests that increased neurological insult (i.e., more revisions) may have a unique impact on the developmental course of inattention, accounting for individual variability in the development of inattention symptoms. Possible explanations for this relationship may include the involvement of affected posterior brain regions in youth with SB and the role of connectivity between the dorsal attention network (DAN) and other neural networks (Brewer et al., 2001; Farrant & Uddin, 2015). Specifically, as increased connectivity between DAN and other neural networks reflects better top-down attentional processing abilities acquired later in development, it is possible that repeated neurological insult negatively impacts the connectivity between these neural networks over time and, subsequently, the development of inattention (Farrant & Uddin, 2015).

Lastly, lesion level significantly predicted the slope for parent-reported inhibition, with higher lesion level predicting greater inhibition problems across age. These findings are contrary to those found at 11.5 years, in which higher lesion level predicted fewer parent-reported inhibition problems. This suggests that youth with higher lesion level have greater inhibition abilities at 11.5 years, but these gains are not maintained across development. These findings may reflect the impact of the relationship between atypical brain development exhibited in youth with higher lesion level and additional neurocognitive functions, such as processing speed, on inhibition development in these youth (Burmeister et al., 2005; Fletcher et al., 2005).

Strengths, Limitations, and Future Research

The present study possessed numerous strengths. First, this is the first study to examine informant-reported inattention and executive dysfunction over an eight-year period using longitudinal methods in youth with SB. The statistical and methodological procedures employed in this study were able to expand upon extant literature in this population, which has been limited to cross-sectional designs or studies examining only two time points (Tarazi et al., 2008; Zabel et al., 2011). The consideration of individual components of executive dysfunction (e.g., inhibition, shifting, working memory, and planning/organizing) also provides greater insight into the development of executive functions over time, as existing longitudinal literature in SB has generally grouped individual executive functions into higher order indices (e.g., behavioral regulation and metacognition; Zabel et al., 2011).

Furthermore, this study utilized both parent and teacher reports, which allowed for the examination of inattention and executive dysfunction across multiple contexts—namely, home and school. Discrepancies between parent- and teacher-reported outcomes found in the current study illustrate the importance of considering different reporters and settings. In addition, using informant-based measures of these constructs is a strength of the study, as existing literature examining executive functioning over time has generally relied on performance-based measures. The measures used in this study assess “real-world” inattention and executive dysfunction, thus capturing the impact of these functions in everyday life and providing increased clinical application.

While shunt status is widely used as a measure of condition severity in SB literature, limited research has examined the impact of numerous shunt revisions on neuropsychological

and other outcomes. This study, therefore, contributes to the growing body of research on shunt complications. An additional strength of this study is the consideration of multiple reporters of shunt revision history. Specifically, data for number of shunt revisions were collected via mother and father reports, as well as medical chart review. Examining these data using three different reporters revealed numerous discrepancies across reporters, including disagreements between mothers and fathers or between parents and medical charts. Given the retrospective report of shunt revisions at T1 of this study, it was impossible to ascertain whether discrepancies could be attributed to underreporting on medical charts (e.g., youth undergoing revision procedures at another institution) or parent error when reporting shunt history. As a result, the current study included the development of a decision-making process which helped to resolve discrepancies and obtain a single value for total lifetime number of shunt revisions for each participant. This system attempted to minimize errors by prioritizing: (1) agreement among any two reporters, and (2) medical chart data (over other forms of data), particularly when the medical chart reported the highest number of revisions. Therefore, strengths of this study include highlighting the significance of considering reporter bias when measuring number of shunt revisions, as well as providing a model for how to resolve discrepancies across reporters.

Although the use of shunt revision data is a strength of this study, one limitation is that the procedure established for resolving discrepancies among reporters involved reaching consensus when data did not neatly fit into the predetermined rules (e.g., rule 6; Table 2). Therefore, these data were not necessarily obtained in a precise manner. Future research should anticipate these challenges in obtaining data on shunt revision history and prospectively strategize to collect accurate, precise shunt revision data. It is recommended that researchers

speak with families and medical providers to resolve discrepancies during the data collection process to minimize the need for retrospective data resolution. Future research should also consider shunt revisions beyond 11.5 years, as youth with SB have been found to exhibit an increase in rate of revisions in the early teen years (Dupepe et al., 2016)

Another limitation of the current study is the relatively small sample size. Due to sample size, the current analyses were only able to detect large effects. Some findings were trending towards significance, which indicates that an increase in sample size may have resulted in additional significant findings. Nonetheless, including age in our models allowed for REML methods to estimate parameters for missing data. Therefore, the findings obtained with the current analyses were still meaningful despite planned attrition (i.e., absence of parent data after participants turned 18).

In addition, to reduce the number of analyses, mother and father reports were combined in the current study. Despite significant associations between reporters allowing for the creation of parent-report composites, considering mother and father reports separately may produce unique findings. Furthermore, fathers are generally underrepresented in research in pediatric populations (Phares et al., 2005). Considering fathers' perspectives individually in pediatric psychology research is thus warranted. Future research should also examine the development of self-reported executive dysfunction and inattention, given evidence of differences between parent- and self-reports on the BRIEF in adults with SB (Zabel et al., 2011). Lastly, the current study included geographic limitations, as participants only included youth with spina bifida within the midwestern United States. Future research should be based on institutional

collaborations that enable more geographic diversity, thus improving generalizability and sample size.

Conclusions and Clinical Implications

Overall, findings from this study addressed notable gaps in extant SB literature. Results indicated that elements of inattention and executive dysfunction may improve over time in youth with SB, though this varies based on setting and reporter. This study also identified a relationship between condition severity variables and inattention and executive dysfunction at 11.5 years; however, only number of shunt revisions and lesion level were associated with outcomes over time. This study has several clinical implications, including the significance of early identification of neurocognitive deficits in youth with SB and earlier implementation of interventions.

By examining inattention and executive dysfunction over time and considering condition severity variables as predictors of development, the current study aimed to inform early identification of neuropsychological deficits and implementation of interventions. Findings suggest that youth with SB would benefit from early interventions targeting inattention, planning/organizing, and shifting, as deficits in these domains may persist across adolescence. Youth with increased condition severity, particularly with a history of shunting and shunt revisions, may further benefit from interventions focused on working memory. Additionally, these findings indicate that youth with increased shunt revisions and higher lesion level require supports in the areas of inattention and inhibition, respectively, over time. Early identification of these deficits and early intervention, with special consideration of condition severity factors, may therefore result in better longitudinal outcomes for youth with SB. This is particularly important

given well-documented associations between inattention and executive dysfunction and psychosocial functioning, academic functioning, independence, and condition-related management in this population.

To aid with early identification of executive dysfunction and inattention, medical providers serving youth with SB should consider regularly incorporating reporter-based screenings into clinic visits, using measures such as the BRIEF and SNAP-IV. This information should be routinely examined, as the current study demonstrates that these outcomes may significantly change over time. Furthermore, this information should be collected with special consideration of the youth's condition severity (i.e., shunt status, history of shunt revisions, and lesion level). Importantly, early identification of deficits would promote earlier implementation of effective interventions. Such interventions could include skills training, use of organizational aids, and formal cognitive training targeting executive functions (Rose & Holmbeck, 2007; Tuminello et al., 2012). Goal Management Training (GMT) is an example of one intervention that targets sustained attention and inhibitory control and has been found to improve performance-based and self-reported executive dysfunction, emotional health, and coping in adults with SB (Stubberud et al., 2013; Stubberud et al., 2014; Stubberud et al., 2015). A pediatric protocol for GMT has been developed and found to be acceptable and feasible in youth with SB, though a larger-scale trial is necessary to determine the efficacy of the protocol for changes in neurocognitive and associated outcomes (Stubberud et al., 2020). Regardless of intervention selection, these interventions should be modified based on the youth with SB's developmental level and condition severity to best serve the needs of the child and subsequently result in better outcomes (Yun & Kim, 2017).

Lastly, this study also informs current shunting practices in SB. As described, surgical ventricular shunting is a potentially life-saving practice for youth with SB, though shunts often require surgical revisions across the lifespan. Despite the prevalence of shunt revisions, little research has examined the impact of shunt revision frequency on neurocognitive outcomes, and the research that does exist is mixed (Brown et al., 2008; Loss et al., 1998; Tuminello et al., 2012). No research to date has examined the correlates of shunting practices, which would allow for a direct comparison of neuropsychological and other outcomes based on shunting decisions. Of note, randomized trials of shunting vs. non-shunting decisions are unlikely, as randomization of research participants to shunt vs. no shunt conditions would be unethical due to implications for morbidity and mortality. Therefore, researchers are left to consider how information regarding the relationship between condition severity (i.e., shunt status, shunt revisions, and lesion level) and various outcomes may inform clinical practice following the medical decision to shunt.

Findings from this study therefore contribute to this expanding literature on clinical implications of shunting. First, if medical providers and caregivers have the knowledge that youth with shunts and a high rate of revisions are at increased risk for neurocognitive deficits, they are more able to identify potential deficits and implement interventions to better support these youth. Second, the finding that shunt revisions may contribute to worsening inattention, and perhaps other outcomes not included in the current study (e.g., psychosocial functioning, quality of life, medical self-management factors), indicates that research efforts are necessary to advance shunting techniques and reduce revision rates. Factors that may be associated with shunt revisions, such as age at shunt placement, history of infections, shunt strategy, valve type and

size, and use of alternative methods, such as endoscopic third ventriculostomy (ETV), should continue to be explored in youth with shunted hydrocephalus to promote enhanced surgical outcomes and subsequent neuropsychological functioning (Hatlen et al., 2012; McCarthy et al., 2019; Reddy, Bollam, & Caldito, 2014; Rinaldo et al., 2019; Tervonen et al., 2017).

APPENDIX A
MEASURES

Questionnaire Measures (Alphabetized):

Behavior Rating Inventory of Executive Function (BRIEF)

Medical History Questionnaire (MHQ)

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

BRIEF
Parent Form

Instructions:

On the following pages is a list of statements that describe children. We would like to know if your child has had problems with these behaviors over the past 6 months. Please answer all the items the best that you can. Please **DO NOT SKIP ANY ITEMS**. Think about your child as you reach each statement and circle your response:

N if the behavior is **Never** a problem
S if the behavior is **Sometimes** a problem
O if the behavior is **Often** a problem

N=Never S=Sometimes O=Often

1. Overreacts to small problems	N	S	O
2. When given three things to do, remembers only the first or last	N	S	O
3. Is not a self-starter	N	S	O
4. Leaves playroom a mess	N	S	O
5. Resists or has trouble accepting a different way to solve a problem with schoolwork, friends, chores, etc.	N	S	O
6. Becomes upset with new situations	N	S	O
7. Has explosive, angry outbursts	N	S	O
8. Tries the same approach to a problem over and over even when it does not work.	N	S	O
9. Has a short attention span	N	S	O
10. Needs to be told to begin a task even when willing	N	S	O
11. Does not bring home homework, assignment sheets, materials, etc.	N	S	O
12. Acts upset by a change in plans	N	S	O
13. Is disturbed by change of teacher or class	N	S	O
14. Does not check work for mistakes	N	S	O
15. Has good ideas but cannot get them on paper	N	S	O
16. Has trouble coming up with ideas for what to do in play or free time	N	S	O
17. Has trouble concentrating on chores, schoolwork, etc.	N	S	O
18. Does not connect doing tonight's homework with grades	N	S	O

N=Never S=Sometimes O=Often

19. Is easily distracted by noises, activity, sights, etc.	N	S	O
20. Becomes tearful easily	N	S	O
21. Makes careless errors	N	S	O
22. Forgets to hand in homework, even when completed	N	S	O
23. Resists change of routine, foods, places, etc.	N	S	O
24. Has trouble with chores or tasks that have more than one step	N	S	O
25. Has outbursts for little reason	N	S	O
26. Mood changes frequently	N	S	O
27. Needs help from an adult to stay on task	N	S	O
28. Gets caught up in details and misses the big picture	N	S	O
29. Keeps room messy	N	S	O
30. Has trouble getting used to new situations (classics, groups, friends)	N	S	O
31. Has poor handwriting	N	S	O
32. Forgets what he/she was doing	N	S	O
33. When sent to get something, forgets what he/she is supposed to get	N	S	O
34. Is unaware of how his/her behavior affects or bothers others	N	S	O
35. Has good ideas but does not get job done (lacks follow-through)	N	S	O
36. Becomes overwhelmed by large assignments	N	S	O
37. Has trouble finishing tasks (chores, homework)	N	S	O
38. Acts wilder or sillier than others in groups (birthday parties, recess)	N	S	O
39. Thinks too much about the same topic	N	S	O
40. Underestimates time needed to finish tasks	N	S	O
41. Interrupts others	N	S	O
42. Does not notice when his/her behavior causes negative reactions	N	S	O
43. Gets out of seat at the wrong times	N	S	O
44. Gets out of control more than his/her friends	N	S	O
45. Reacts more strongly than other children	N	S	O
46. Starts assignments or chores at the last minute	N	S	O
47. Has trouble getting started on homework or chores	N	S	O
48. Has trouble organizing activities with friends	N	S	O
49. Blurts things out	N	S	O
50. Mood is easily influenced by the situation	N	S	O
51. Does not plan ahead for school assignments	N	S	O
52. Has poor understanding of own strengths and weaknesses	N	S	O
53. Written work is poorly organized	N	S	O
54. Acts too wild or "out of control"	N	S	O
55. Has trouble putting the brakes on his/her actions	N	S	O
56. Gets in trouble if not supervised by an adult	N	S	O
57. Has trouble remembering things, even for a few minutes	N	S	O
58. Has trouble carrying out the actions needed to reach goals (saving money for special item, studying to get a good grade)	N	S	O
59. Becomes too silly	N	S	O
60. Work is sloppy	N	S	O

N=Never S=Sometimes O=Often

61. Does not take initiative	N	S	O
62. Angry or tearful outbursts are intense but end suddenly	N	S	O
63. Does not realize that certain actions bother others	N	S	O
64. Small events trigger big reactions	N	S	O
65. Talks at the wrong time	N	S	O
66. Complains there is nothing to do	N	S	O
67. Cannot find things in room or school desk	N	S	O
68. Leaves a trail of belongings wherever he/she goes	N	S	O
69. Leaves messes that others have to clean up	N	S	O
70. Becomes upset too easily	N	S	O
71. Lies around the house a lot (“couch potato”)	N	S	O
72. Has a messy closet	N	S	O
73. Has trouble waiting for turn	N	S	O
74. Loses lunch box, lunch money, permission slips, homework, etc.	N	S	O
75. Cannot find clothes, glasses, shoes, toys, books, pencils, etc.	N	S	O
76. Tests poorly even when he/she knows the correct answers	N	S	O
77. Does not finish long-term projects	N	S	O
78. Has to be closely supervised	N	S	O
79. Does not think before doing	N	S	O
80. Has trouble moving from one activity to another	N	S	O
81. Is fidgety	N	S	O
82. Is impulsive	N	S	O
83. Cannot stay on the same topic when talking	N	S	O
84. Says the same things over and over	N	S	O
85. Has trouble getting through morning routine in getting ready for school	N	S	O

Medical History and Adherence Questionnaire

1. What type of spina bifida does your child have?

- lipomeningocele (lipo)
 myelomeningocele (MM)
 not sure

2. What is the level of your child's lesion?

- sacral
 lumbar
 thoracic
 not sure

3. Does your child have a shunt? yes _____ no _____

- a. IF YES, has your child's shunt been infected? yes _____ no _____
 b. IF YES, has your child had a shunt revision? yes _____ no _____
 c. IF your child's SHUNT HAS BEEN INFECTED, how many times? _____
 d. IF your child has had a SHUNT REVISION, how many times? _____

4. Does your child have seizures or take medication to prevent seizures?

yes _____ no _____

5. Is your child able to do independent toileting?

yes _____ no _____

6. Is your child on a catheterization schedule? yes _____ no _____

a. If YES, does your child do the catheterization (check one)?

- independently without reminding
 independently with reminding
 with partial assistance
 with complete assistance

b. Has your child ever had a bladder or urinary tract infection? yes _____ no _____

c. How many times has your child had a bladder or urinary tract infection? _____

d. Has your child had bladder stimulation? yes _____ no _____

7. Is your child on a bowel program?

yes _____ no _____

a. If YES, what type of bowel program (suppositories, diet, enemas, digital manipulation, etc.)?

b. If YOUR CHILD IS ON A BOWEL PROGRAM, does your child do this program (check one)?

_____ independently without reminding
 _____ independently with reminding
 _____ with partial assistance
 _____ with complete assistance

c. Has your child had bowel stimulation? yes _____ no _____

8. Does your child use diapers? yes _____ no _____

a. If YES, where does your child use diapers (please check all that apply)?

_____ school
 _____ home
 _____ on outings
 _____ all the time
 _____ other? _____

9. Does your child use braces? yes _____ no _____

a. If YES, what type (please check all that apply)?

_____ ankle-foot
 _____ knee-ankle-foot
 _____ hip-knee-ankle-foot
 _____ reciprocating brace
 _____ full control brace
 _____ swivel walker
 _____ parapodium
 _____ twister cables
 _____ night splint
 _____ back brace

10. Does your child use crutches? yes _____ no _____

11. Does your child use a walker? yes _____ no _____

a. If YES, where does your child use a walker (please check all that apply)?

- _____ school
- _____ home
- _____ for long distance walking
- _____ on outings
- _____ all the time
- _____ other? _____

12. Does your child use a wheelchair? yes _____ no _____

a. If YES, where does your child use a wheelchair (please check all that apply)?

- _____ school
- _____ home
- _____ for long distance travel
- _____ on outings
- _____ all the time
- _____ other? _____

13. If your child uses more than one mobility device, please write down the percentage of time that your child uses each device (please make sure that the percentages add up to 100%):

- _____ % unassisted walking (no braces)
- _____ % braces alone (no crutches or walker)
- _____ % braces with crutches or walker
- _____ % wheelchair
- = 100 %

14. Please list your child's medications (include NAME OF MEDICATION, AMOUNT, HOW OFTEN TAKEN):

	<u>Name of Medication</u>	<u>Amount</u>	<u>How Often Taken?</u>
1.	_____	_____	_____
2.	_____	_____	_____
3.	_____	_____	_____
4.	_____	_____	_____
5.	_____	_____	_____
6.	_____	_____	_____
7.	_____	_____	_____
8.	_____	_____	_____
9.	_____	_____	_____
10.	_____	_____	_____

15. Please list your child's surgeries, since birth (include year of surgery, reason for surgery; examples include: shunt revision, shunt replacement, leg surgery, back surgery, tethered cord, etc.):

	<u>Year of Surgery</u>	<u>Reason for Surgery</u>
1.	_____	_____
2.	_____	_____
3.	_____	_____
4.	_____	_____
5.	_____	_____
6.	_____	_____
7.	_____	_____
8.	_____	_____
9.	_____	_____
10.	_____	_____
11.	_____	_____
12.	_____	_____
13.	_____	_____
14.	_____	_____
15.	_____	_____

16. What changes have occurred in your child's health OVER THE PAST YEAR?

17. What type of health insurance does your child have?

18. In the past year, how many visits has your child had with a primary care physician (regular family doctor)? _____

Please describe the reason for these visits: _____

19. In the past year, how many visits has your child had with a urologist? _____

Please describe the reason for these visits: _____

20. In the past year, how many visits has your child had with an orthopedist? _____

Please describe the reason for these visits: _____

21. In the past year, how many visits has your child had with a neurologist? _____

Please describe the reason for these visits: _____

22. In the past year, how many visits has your child had with a physical or occupational therapist? (please specify which one)

Please describe the reason for these visits: _____

23. In the past year, on how many occasions has your child visited the emergency room? _____

Please describe the reason for these visits: _____

24. In the past year, how many visits has your child had with any other type of health care professional? _____

Type of health professional seen: _____

Please describe the reason for these visits: _____

25. In the past year, how many times has your child been hospitalized? _____

Length of stay _____

Please describe the reason for these hospitalizations: _____

The SNAP-IV Teacher and Parent Rating Scale
James M. Swanson, Ph.D., University of California, Irvine, CA 92715

For each item, check the column that best describes this child:

	Not At All	Just A Little	Quite A Bit	Very Much
1. Often fails to give close attention to details or makes careless mistakes in schoolwork or tasks	_____	_____	_____	_____
2. Often has difficulty sustaining attention in tasks or play activities	_____	_____	_____	_____
3. Often does not seem to listen when spoken to directly	_____	_____	_____	_____
4. Often does not follow through on instructions and fails to finish schoolwork, chores, or duties	_____	_____	_____	_____
5. Often has difficulty organizing tasks and activities	_____	_____	_____	_____
6. Often avoids, dislikes, or reluctantly engages in tasks requiring sustained mental effort	_____	_____	_____	_____
7. Often loses things necessary for activities (e.g., toys, school assignments, pencils, or books)	_____	_____	_____	_____
8. Often is distracted by extraneous stimuli	_____	_____	_____	_____
9. Often is forgetful in daily activities	_____	_____	_____	_____
10. Often fidgets with hands or feet or squirms in seat	_____	_____	_____	_____
11. Often leaves seat in classroom or in other situations in which remaining seated is expected	_____	_____	_____	_____
12. Often runs about or climbs excessively in situations in which it is inappropriate	_____	_____	_____	_____
13. Often has difficulty playing or engaging in leisure activities quietly	_____	_____	_____	_____
14. Often is "on the go" or often acts as if "driven by a motor"	_____	_____	_____	_____
15. Often talks excessively	_____	_____	_____	_____
16. Often blurts out answers before questions have been completed	_____	_____	_____	_____
17. Often has difficulty awaiting turn	_____	_____	_____	_____
18. Often interrupts or intrudes on others (e.g., butts into conversations/games)	_____	_____	_____	_____

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VITA

Allison d. Payne is a doctoral student in the Clinical Psychology program at Loyola University Chicago, specializing in clinical-child and neuropsychology. Prior to beginning her graduate studies, Payne earned her B.A. in Psychology with minors in Cognitive Science and Philosophy & Bioethics from Georgetown University in 2018. During her junior year, Payne conducted research in the Safe Concussion Outcome, Recovery, & Education (SCORE) Clinic at Children's National Hospital in Washington, D.C, where she worked under Drs. Gerard Gioia, Catherine McGill, Christopher Vaughan, and Alison Burns. During her senior year, Payne continued to expand her research experience at Children's National Hospital under the mentorship of Dr. Karin S. Walsh. Upon graduation, Payne became a full-time research coordinator for Dr. Walsh and was responsible for nine research studies examining children, adolescents, and young adults with Neurofibromatosis Type 1 (NF1), cancer, and hemophilia. It was during this time that Payne's interests in the neuropsychological correlates of pediatric chronic illness, as well as the effects of neurocognitive function, family relations, and culture on medical self-management and adjustment in youth with chronic illness, were strengthened. As a graduate student at Loyola, Payne is a member of Dr. Grayson Holmbeck's CHATS Lab. Through the CHATS lab, Payne has worked on projects examining psychosocial adjustment in youth and young adults with spina bifida and related family, peer, and neuropsychological factors, as well as a camp-based intervention designed to promote psychosocial adjustment and

independence in children, adolescents, and young adults with spina bifida. Payne's master thesis characterized the development of inattention and executive dysfunction over time in youth with spina bifida and examined condition severity variables as predictors. As a result of these various experiences, Payne has presented research in the form of conference abstracts, oral presentations, and peer-reviewed manuscripts.