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ATTENTION PROBLEMS AND EXECUTIVE FUNCTIONS IN CHILDREN WITH SPINA BIFIDA AND HYDROCEPHALUS

Rachel Burmeister,[1] H. Julia Hannay,[1] Kim Copeland,[1] Jack M. Fletcher,[2] Amy Boudousquie,[2] and Maureen Dennis[3]

[1]Department of Psychology, University of Houston, Texas, [2]Department of Pediatrics, University of Texas Health Science Center at Houston, Texas, [3]Brain-Behavior Program, The Hospital for Sick Children, and Department of Psychology, University of Toronto, Canada

This study addressed the incidence of Attention-Deficit/Hyperactivity Disorder (ADHD) subtypes in children with spina bifida meningomyelocele and shunted hydrocephalus (SBH) as well as differences in executive functions among these subtypes. Parent rating scales revealed that 31% of the group with SBH could be identified with AD/HD, mostly the Inattentive type (23%). The group with SBH differed from normal controls on cognitive measures of executive functions, but subtype differences were not significant. Multivariate tests showed that children with SBH were rated with greater difficulties on the Behavior Rating Inventory of Executive Function (BRIEF) compared to controls; those with SBH and any subtype of ADHD differed from those with SBH and no ADHD; and those with ADHD (Combined Type) differed significantly from those with ADHD (Predominantly Inattentive Type). Subtype differences on univariate tests in the latter comparison were significant on the BRIEF Inhibit scale, showing more disinhibition in those with SBH and ADHD (Combined Type), but no significant differences were apparent on the BRIEF Sustain, Shift, and Initiate scales. The results show that the incidence of ADHD in children with SBH exceeds the population rate, is represented by problems with inattention rather than with impulsivity and hyperactivity; and that as with non-brain injured individuals, subtype differences in cognitive function remain to more clearly delineated.

Spina bifida is the most common severely disabling birth defect in North America, with an incidence of approximately 0.5-1.0 per 1,000 births (Charney, 1992). The most prevalent and severe form of spina bifida involves la spinal lesion (meningomyelocele) that accounts for about 90% of cases (Norman, McGillivray, Kalovsek, Hill, & Poskitt, 1995). This form of spina bifida develops in the first trimester as a neural tube defect, representing a disorder of neurulation when the neural folds do not meet in the midline to form the neural tube, but instead remain continuous with the ectoderm at the skin surface. The bones of the vertebral column fail to fuse in the posterior midline, resulting in a

Address all correspondence to Jack M. Fletcher, Department of Pediatrics, University of Texas Health Science Center at Houston, 7000 Fannin—UCT 2478, Houston, TX 77030, (fax) 713-500-3818, E-mail: Jack.Fletcher@uth.tmc.edu.

protrusion of the spinal cord, meninges, parenchyma, and nerve roots through the defect in the spine (Barkovich, 1995; Menkes & Till, 1995).

Spina bifida meningomyelocele is also a disorder associated with a variety of brain abnormalities, usually including a congenital malformation of the cerebellum and hindbrain (Chiari II) and in about half the cases, partial dysgenesis of the corpus callosum (Barkovich, 1995; Hannay, 2000). Hydrocephalus occurs in 95% of children with this disorder, with 80–90% requiring shunting (Reigel & Rotenstein, 1994). The hydrocephalus leads to stretching and thinning of the cortex, especially in posterior regions and the corpus callosum. As such, spina bifida meningomyelocele with hydrocephalus (SBH) represents a complicated series of neural insults that begins prior to birth, with persisting effects on development, including problems in the orthopedic, cognitive, and behavioral domains (Anderson, 1975; Fletcher, Dennis, & Northrup, 2000; Fletcher, Dennis, et al., 2004; Wills, 1993).

Although the effects of SBH have been observed in a variety of neurobehavioral domains, a modal profile can be described (Fletcher, Dennis, et al., 2004). Despite the severity of the neural insults, children with SBH are not typically mentally retarded, although deficiencies on intellectual tasks are present in many, especially on Performance IQ measures. Thus, children with SBH are stronger in specific language domains and weaker on many perceptual and motor tasks, demonstrated in many comparisons of Verbal and Performance IQ scores (Dennis et al., 1981; Donders, Canady, & Rourke, 1990; Fletcher et al., 1992, Raimondi & Soare, 1974). Not all areas of language develop well: children with SBH are proficient in using and understanding single words, but many have problems producing and understanding discourse level language (Barnes & Dennis, 1992, 1998; Dennis, Jacennik, & Barnes, 1994). Math and writing are commonly impaired relative to stronger word recognition skills (Barnes et al., 2002), and the discourse level difficulties lead to parallel weaknesses in reading comprehension (Barnes & Dennis, 1998). Verbal learning and memory impairments are common, but rote memory is usually stronger (Scott et al., 1998; Yeates, Enrile, Loss, & Blumenstein, 1995). Attention skills may also be impaired (Brewer, Fletcher, Hiscock, & Davidson, 2001; Fletcher et al., 1996; Loss, Yeates, & Enrile, 1998). Like memory, skills within this domain are not uniformly impaired, with focusing and shifting of attention more impaired than sustained attention (Brewer et al., 2001; Loss et al., 1998). Similar conclusions are appropriate for assessments of executive functions, in which children with SBH tend to be impaired, especially on measures requiring planning and organization, goal-directed behavior, and problem solving (Fletcher et al., 1996; Snow, 1999).

Less well understood are behavioral outcomes in children with SBH. Behavioral studies generally show higher rates of adjustment difficulties, but specific types of adjustment difficulties have not been identified (Donders, Rourke, & Canady, 1992; Fletcher et al., 1995; Greenley, Holmbeck, Zukerman, & Buck, in press; Zurmohle et al., 1998). Most studies focus on children with hydrocephalus and are not specific to children with SBH. Anecdotally, children with SBH are often described as having specific behavioral characteristics, including excessive and often inappropriate sociability, excessive verbosity, and other examples of poor social skills (Greenley et al., Taylor, 1961; in press). As adolescents, they are often described as withdrawn and isolated. Many children with SBH have been observed to exhibit behavioral problems similar to children with attention-deficit/hyperactivity disorder (ADHD), such as difficulties with distractibility, organizing material, and staying on task, which can also be associated with the types of social difficulties attributed to children with SBH or a nonverbal learning disability (Rourke, 1989).

The presence of ADHD in children with SBH has not been well documented. In the only study of which we are aware, Ammerman et al. (1998) evaluated children ages 6-18 years of age with spina bifida for ADHD using the Child Symptom Inventory (Gadow & Sprafkin, 1987). In this sample of 54 children, 18 (33%), obtained a screening cutoff score consistent with a diagnosis of ADHD. These children had higher scores on the Inattention Scale (M = 2.33, SD = 0.69) than the Hyperactivity/Impulsivity Scale (M = 1.56, SD = 0.66). No differences between children with SBH classified with ADHD and those not classified with ADHD were apparent in age, gender, ambulation status, or lesion level. However, the type of spinal dysraphism and status of hydrocephalus were not completely documented. It is frequently observed that children with upper level meningomyeloceles and hydrocephalus have more adverse neurobehavioral outcomes (Fletcher, Copeland, et al., 2005).

Documenting the presence of ADHD in children with SBH is important because many struggle in school and with social functions. Interventions commonly recommended for children with ADHD may be useful for children with SBH. Moreover, studies of attention and executive functions suggest that children with SBH do have difficulties in these overlapping domains (Fletcher et al., 2000). In the attention domain, Loss et al. (1998) compared children with SBH, spina bifida and no history of shunted hydrocephalus, and sibling controls on neuropsychological assessments of four elements of attention derived from the Mirsky (1996) model of attention. These components included Encode (assessed by Digit Span and Arithmetic subtests from the Wechsler Intelligence Scales for Children-III; Wechsler, 1991), Sustain (continuous performance test; Gordon, 1983), Focus/Execute (WISC- III Coding, Trail Making Tests A and B; Reitan & Wolfson, 1985), and Shift (conceptual responses and perserverative errors from the Wisconsin Card Sorting Test; Heaton, Chelune, Talley, Kay, & Curtis, 1993). Parents completed the Attention Problems scale of the Child Behavior Checklist (Achenbach, 1991). Comparisons of the group with SBH and sibling controls revealed significant differences on all four elements, although the group differences were larger for Encode, Focus/Execute, and Shift than for Sustain. There was a statistically significant, but weak relation of the Attention Problems scale and the four elements of attention across the different groups that was strongest for the parent rating and Focus/Execute element within the group with SBH.

In a more recent study, Brewer et al. (2001) compared children with congenital hydrocephalus (predominately SBH), ADHD (combined type), and normal controls on measures of focused attention (Posner Visual Orienting and Detection Task), sustained attention (Continuous Performance Task), and attention shifting (Wisconsin Card Sorting Test). Components from these tasks have been linked to attention systems mediated by anterior or posterior brain networks (Mirsky, 1996; Barkley, 1997). The anterior system is related to the sustaining and maintenance functions of attention, whereas the posterior system mediates attention functions related to focusing and shifting. The results showed that children with congenital hydrocephalus displayed predominant difficulties on task components involving the ability to focus and shift attention, which specifically implicated impairment of the posterior brain attention system. Children with ADHD displayed *patterns* of performance that were in the same direction as controls on measures of focused attention once their difficulties with sustained attention were taken into account. However, this group showed problems sustaining attention across trials, so that performance was more variable and slower over time. Difficulties sustaining attention are commonly associated with the anterior brain attention system. In contrast, children with hydrocephalus responded at the same level over time despite their difficulties focusing and shifting. Thus, performance of children with hydrocephalus was different from that of children with ADHD. Other pieces of evidence support the view that children with SBH have deficits of the posterior attention system. Children with SBH show problems disengaging from exogenous cues (Dennis, Edelstein, Frederick, et al., in press) and also attenuated inhibition of return (Dennis, Edelstein, Copeland, et al., in press), the latter a posterior attention system problem that children with ADHD do not show (Li, Chang, & Lin, 2003). Thus, there are similarities, but important differences, in the attention skills of children with SBH and children with ADHD.

Just as with tests of attention, measures of executive function show a distinctive profile of performance in children with SBH. In the executive function domain, Fletcher et al. (1996) found that school-age children with SBH displayed difficulties on a number of executive function tasks, including the Stroop task, Tower of London, and Wisconsin Card Sorting. However, patterns of performance reflected speed of processing and difficulties with arousal and attention orientation as opposed to classic "frontal" patterns. Performance on the Stroop task, for example, reflected problems in naming speed rather than cognitive inhibition. On the Tower and Wisconsin tasks, the number of trials to solve the first problem or category was deficient, but there were no differences relative to controls in the total number of problems solved or categories achieved; errors were not consistent with specific patterns involving disinhibition or perseveration. Similarly, Snow (1999) found that children with SBH exhibited severe deficits in visual planning and sequencing on the Trail Making Tests and problem solving on the Wisconsin Card Sorting Test compared not only with typically developing children, but also when compared with children with learning disabilities and/or ADHD.

A number of issues are still unclear about the role of ADHD in children with SBH and those with developmental ADHD. Brewer et al. (2001) did not attempt to characterize ADHD in this population of children with hydrocephalus. Loss et al. (1998) correlated parent ratings with neuropsychological assessments but also did not subdivide children with SBH into ADHD subtypes. It may be that few children with SBH displayed characteristics of the hyperactive-impulsive or combined type of ADHD. It is also possible that children with SBH are more like those with the predominantly inattentive type of ADHD. Behaviorally these children are often described as distractible, unfocused, lethargic, and passive (Barkley, 1998), but without the impulsivity characteristic of children with the other forms of ADHD. At the same time, cognitive assessments often do not robustly differentiate these forms of ADHD (Barkley, DuPaul, & McMurray, 1990; Klorman et al., 1999, 2002), raising questions about the validity of differentiating these types.

The primary purpose of our study was to examine the incidence and types of ADHD in a large sample of well-characterized children with SBH. The prevalence of ADHD in the normal population varies by definition and method of diagnosis. Barkley (1998) reported a 2%–9.5 % rate of ADHD for clinical interviews compared to 7%–17% for behavioral rating scales. Because we used behavior rating scales in our research protocol, we were interested in the incidence of ADHD subtypes using the latter method of identification. According to Barkley (1998), the prevalence of ADHD-combined type (ADHD-C) and ADHD-predominantly hyperactive/impulsive type (ADHD-HI) ranged from 4%–10%, whereas the rate for ADHD-predominantly inattentive type (ADHD-I) ranged from 9%–10%. We hypothesized that the number of children with SBH meeting ADHD criteria, based on behavior rating scales, would be higher than the upper level of 17% expected in the general population. We also predicted that children with SBH would be more likely to show ADHD-I based on behavioral descriptions and the results of cognitive studies of SBH, which generally do not show results consistent with more impulsive subtypes of ADHD.

The secondary purpose of our study was to examine the relation of ADHD classification and executive function performance in children with SBH. We do not address the overarching question of the validity of ADHD subtypes. However, subdividing children with SBH into ADHD subtypes may help explain variability in performance on executive function tasks. Thus, we selected measures of cognitive function as well as behavior ratings of executive function. The cognitive tasks measured constructs that have been interpreted as assessments of executive functions in other studies (Barkley, 1997) and were selected from a large test battery given to the sample in this study. Based on prior research, and because these types of measures are typically factorially confounded (i.e., tap multiple dimensions of cognitive functions), we hypothesized that that as a group, children with SBH would perform more poorly than non-SBH controls on all measures associated with executive functions, including assessments of working memory, concept formation, and semantic organization. More importantly, we predicted that children with SBH meeting criteria for ADHD (regardless of subtype) would perform more poorly than children with SBH who were not classified as ADHD on all measures of executive function/attention. Thus, we hypothesized that children with SBH classified as ADHD-C would perform more poorly than children with SBH classified as ADHD-I on all the cognitive measures because they assess executive functions often associated with anterior brain systems and specifically implicated in ADHD-C (Barkley, 1997).

The behavior rating measure of executive function may be more sensitive to differences between ADHD subtypes because it attempts to differentially assess different components of attention and executive functions (Barkley, 1998). Mahone, Zabel, Levey, Verda, and Kinsman (2002) included the Behavior Rating Inventory of Executive Function (BRIEF) in a study of 28 adolescents with SBH. Parents rated these adolescents as experiencing more difficulties on BRIEF subscales involving executive functions relative to the normative sample, but the small sample was not subdivided into subtypes of ADHD. Interestingly, scores on the Behavioral Assessment System for Children (Reynolds & Kamphaus, 1998), which includes attention problem scales, were not significantly elevated and were not correlated with the BRIEF ratings. A *DSM-IV* checklist for ADHD yielded mean percentiles that were above average, with some children clearly showing elevations in the clinically significant range, although these scores were not correlated with the other rating scales. Thus, we hypothesized that children with SBH classified with ADHD (regardless of subtype) would have higher ratings than either controls or children with SBH not classified as ADHD on the BRIEF. We also hypothesized that children with SBH classified as ADHD-C would have higher ratings than the ADHD-I group on scales assessing sustained attention and inhibition, whereas children classified as ADHD-I would have higher ratings than the ADHD-C group on scales assessing initiation and shifting attention. Finally, we explored differences in sociodemographic, medical, and intellectual characteristics related to ADHD subtype to establish whether any differences in cognitive functions and behavior ratings were due to differences in these characteristics.

METHOD

Participants

The participants were 205 children (7–16 years old), including children without SBH ($n = 41$) and children with SBH ($n = 164$) recruited as part of a large study of the genetic and neurobehavioral characteristics of spina bifida (Fletcher, Dennis, et al., 2004).

Children in the neurologically normal comparison group had normal MRI scans and no history of congenital or acquired neurological disorder. The diagnosis of SBH was based on a review of medical history and in most cases, magnetic resonance imaging (MRI) of the brain. All the children with SBH in this study were born with a meningomyelocele and had shunts placed during the first 3 months of life (usually in the 1st month) to control progressive hydrocephalus. Children were not included in this study if their hydrocephalus was associated with other neurological anomalies (e.g., tumor, trauma, stroke) or genetic syndromes. No child was included with uncontrolled epilepsy, primary sensory loss, severe behavioral disorder, evidence of abuse or neglect, severe spasticity, or problems with hand use so severe that the child could not manipulate test materials. Children were required to have a Verbal Reasoning or Visual/Abstract Reasoning Standard Area Score above 69 on the Stanford Binet Intelligence Scale: Fourth Edition (Thorndike, Hagen, & Sattler, 1986) to ensure that findings did not reflect generalized mental deficiency. This resulted in the exclusion of 22% of the overall sample (Fletcher, Dennis, et al., 2004).

Assessment of ADHD

The group with SBH was divided into subgroups based on scores on the Swanson Nolan Achenbach Pelham-IV (SNAP-IV) Parent Rating Scale (Swanson, 1992). The SNAP-IV is a parent and teacher rating scale addressing inattention, impulsivity, hyperactivity, oppositional behavior, and other dimensions of childhood behavioral adjustment. It was developed specifically to assess ADHD and comorbid conditions in children. We used the SNAP-IV because it is based on *DSM-IV* criteria and has been used in several studies of ADHD, including the NIMH ADHD multimodal clinical trial (MTA Cooperative Group, 1999). In the latter trial, there was good concordance of the SNAP-IV and the structured interview used to identify children with ADHD. The scale consists of 90 items consistent with ADHD and oppositional defiant disorder (ODD) criteria in *DSM-IV* (American Psychiatric Association, 1994) as well as items from the Conners Index Questionnaire (Conners, 1984), IOWA Conners Questionnaire (Loney & Milich, 1982), and items from *DSM-IV* disorders that may overlap with criteria for ADHD. Children are rated on a 4-point scale (0 = Not at all, 1 = Just a little, 2 = Quite a bit, 3 = Very much) on each behavioral item. An average response per question was determined for each dimensional scale by summing the item scores for each scale dividing by the number of items. For this study, we focused only on the inattention and hyperactivity/impulsivity scales in an effort to identify children with attention problems. Based on a normative sample of 847 children in 15 schools in Irvine, California, cutoff scores for the Inattention (1.78) and Hyperactivity/Impulsivity (1.44) Scales were determined by using the average response per item for this normative population + 1.65 SD. This cutoff level was proposed to identify 5% of the population as having attention deficit/hyperactivity disorder, but may identify up to 8% because the distribution of scores on these types of scales tends to be skewed (Swanson et al., 2002).

Procedures

Each child was administered the Stanford Binet and three cognitive tests assessing executive functions as a part of a larger neurobehavioral assessment. Two subtests from the Woodcock Johnson Test of Cognitive Abilities–Revised (Woodcock & Johnson, 1989) were used to assess working memory (Numbers Reversed) and problem solving (Concept Formation). Learning efficiency and semantic clustering were assessed with the

California Verbal Learning Test–Children's Version (CVLT-C; Delis, Kramer, Kaplan, & Ober, 1994). A parent completed a prepublication form of the BRIEF (Gioia, Isquith, Guy, & Kenworthy, 1998) and the SNAP-IV (Swanson, 1992). In some instances, a cognitive test was not completed because of fatigue or inability to understand instructions. Some parents declined to complete the BRIEF or forms were not returned to the examiner. Therefore, the sample sizes vary slightly for each procedure. The four cognitive tests and the rating scale are described next.

Woodcock Johnson Test of Cognitive Abilities–Revised (WJR; Woodcock & Johnson, 1989). The Concept Formation and Numbers Reversed subtests from the cognitive batteries of the WJR were used. Reliability coefficients for all these tests exceed 0.9, with good evidence of concurrent validity. Fletcher, Copeland, et al. (2005) showed that on each of these measures, children with SBH performed more poorly than controls.

Numbers Reversed is a measure of working memory and requires children to listen to serially presented numbers and repeat them backward. It is a span task, so the child begins with two numbers, with an additional number added until the child fails the three highest trials consecutively by test page. Concept Formation is a controlled-learning task assessing categorical reasoning ability and problem solving. Performance on this task is determined by response style (impulsivity), cognitive flexibility, problem-solving ability, and ability to use feedback to modify performance (McGrew, 1994). The subject is presented with a series of problems in which a matching rule using shape, color, size, or number is to be determined. For all but the most difficult items, the child is given feedback regarding the accuracy of the answer.

California Verbal Learning Test–Children's Version (CVLT-C; Delis et al., 1994). The CVLT-C evaluates strategies and processes involved in verbal learning and memory. Children are presented with a 15-item "shopping" list and asked to recall list items after each of a series of five learning trials. This is followed by a second 15-word interference list, short- and long-delay free and cued recall trials, and recognition memory trial of the original list. Median 1-month test-retest reliabilities for six recall measures on samples of 8-, 12-, and 16-year-old children were 0.64, 0.66, and 0.69, respectively. Although the CVLT-C yields many indices of memory and learning, a detailed analysis is beyond the scope of this paper. We used the standardized score representing the number of words learned on the Trials 1 and 5 and the semantic clustering score, which have been shown to be sensitive to memory and learning difficulties in children with SBH (Fletcher, Copeland, et al., 2005; Yeates et al., 1995).

Behavior Rating Inventory of Executive Function (BRIEF; Gioia et al., 1998). The BRIEF is a parent and teacher rating scale assessing executive functions as they are displayed in the everyday environment of the child. In the prepublication version used in this study, there were 129 items, which fall under nine domains: Initiate, Sustain, Inhibit, Shift, Organize, Plan, Self-Monitor, Working Memory, and Emotional Control. A parent was asked to rate behavioral statements on a 3-point scale (1 = Never, 2 = Sometimes, 3 = Often). The average item response for each domain scale was calculated. We used the prepublication version of the BRIEF because the study began prior to the completion and standardization of the current version of the BRIEF. Thus, there are differences in specific items and scales from the published version. However, the psychometric characteristics should parallel those of the published version, which are excellent (Baron, 2000). Internal consistency across different scales ranges from 0.80–0.98 (Gioia et al., 2000). A variety of validity studies reported in a special issue of *Child Neuropsychology* (2002, 4) provide evidence that the BRIEF is related to other measures of executive functions.

RESULTS

Incidence of ADHD subtypes

Parent rating scores on the SNAP-IV exceeded the cutoff criteria for ADHD in 31% of children with SBH. Thirteen children (8%) were classified as ADHD-C and one child (0.5%) was classified as ADHD-HI. Thirty-eight children (23%) were classified as ADHD-I. Two children (5%) of the 41 control children met criteria for ADHD. Using a z test for analysis of proportions, children with SBH had a significantly higher rate of ADHD than the highest previous estimates in the normal population (17%), $z = 4.77$, $p < .0001$, or in our comparison group (5%), $z = 15.28$, $p < .0001$. Table 1 presents means and standard deviations on the SNAP-IV scales for the SBH groups and controls, showing the expected patterns of ratings. As expected, comparisons of the four groups conducted separately for the two scales (alpha = .05/6 = .0083) were all significant except for the comparison of the two ADHD subtypes on the Inattention scale.

Sociodemographic Characteristics

Table 2 provides the age, gender, ethnicity and socioeconomic status (SES; Hollingshead, 1975) for the subgroups with SBH and controls. There were no significant differences across the groups in ethnicity, run as a comparison of Hispanic and non-Hispanic subgroups due to low sample size in some cells, χ^2 (3, $N = 205$) = 4.95, $p = 0.18$; gender, $\chi^2(3, N = 205) = 1.70$, $p = .64$; or SES, $\chi^2(9, N = 205) = 12.83$, $p = .17$. There was a significant difference in age, $F(3, 201) = 3.14$, $p < .03$, reflecting the younger ADHD-C group. As the comparisons of cognitive functions were based on age-adjusted scores, we did not regard this difference as a problem for the study. In fact, studies of developmental ADHD tend to show more impulsivity and hyperactivity in younger children that recedes over time, with persistence of the difficulties with inattention (Barkley, 1998), paralleling the pattern observed in Table 2.

Intelligence

Table 3 provides the Stanford Binet scores for the subgroups with SBH and controls. One-way ANOVAs for the Verbal and Visual/Abstract Reasoning scales were conducted. There was a main effect of group for both Verbal Reasoning, $F(3, 201) = 17.46$, $p < .0001$,

Table 1 Means and Standard Deviations for the SNAP-IV Parent Rating Scale by Group.

Rating Scale SNAP-IV Parent Rating	Control	SBH Non-ADHD	SBH ADHD-I	SBH ADHD-C
N	41	112	38	14
Inattention				
M	0.53	0.91	2.19	2.29
SD	0.19	0.49	0.30	0.43
Hyperactivity/Impulsivity				
M	0.19	0.29	0.50	1.91
SD	0.29	0.32	0.41	0.41

Note. SBH = Spina bifida meningomyelocele and hydrocephalus; ADHD = Attention deficit/hyperactivity disorder; I = inattentive type; C = combined type; SNAP-IV = Swanson Nolan Achenbach Pelman-IV.

Table 2 Means and Standard Deviations for Sociodemographic Variables by Group.

Variable	Control	SBH Non-ADHD	SBH ADHD-I	SBH ADHD-C
N	41	112	38	14
Age (months)				
M	139.2	129.7	142.0	119.0
SD	33.8	30.8	27.0	22.0
Race *n* (%)				
Caucasian	32 (78)	73 (65)	31 (81)	10 (71)
African American	1 (3)	8 (7)	1 (3)	1 (7)
Hispanic	5 (12)	27 (24)	4 (11)	3 (21)
Asian/other	3 (5)	4 (3)	2 (5)	0
Gender				
Female *n (%)*	22 (54)	60 (54)	19 (50)	5 (36)
Socioeconomic status *n* **(%)**				
Higher (I)	10 (24)	18 (16)	7 (18)	3 (21)
Upper Middle (II)	22 (54)	41 (36)	19 (50)	3 (21)
Middle (III)	4 (10)	16 (14)	5 (13)	3 (21)
Lower (IV-V)	5 (12)	37 (33)	7 (18)	5 (36)

Note. SBH = spina bifida meningomyelocele and hydrocephalus; ADHD = attention deficit/hyperactivity disorder; I = inattentive type; C = combined type.

Table 3 Means and Standard Deviations for the Stanford-Binet Intelligence Test–Fourth Edition by Group.

Subtest	Control	SBH Non-ADHD	SBH ADHD-I	SBH ADHD-C
N	41	112	38	14
Verbal Reasoning				
M	110.10	89.27	89.62	94.14
SD	13.65	17.55	14.44	16.83
Visual/Abstract Reasoning				
M	112.10	87.26	88.76	91.14
SD	11.05	16.17	15.81	16.87

Note. SBH = spina bifida meningomyelocele and hydrocephalus; ADHD = attention deficit/hyperactivity disorder. The total for ADHD-C includes one child who met diagnostic criteria for ADHD, hyperactive- impulsive type; I = inattentive type; C = combined type.

and Visual/Abstract Reasoning, $F(3, 201) = 27.70$, $p<.0001$. Follow-up tests using Tukey's honestly significant difference (HSD) procedure controlling alpha at $p<.05$ showed that the children with SBH (regardless of ADHD subtype) had significantly lower scores than the control group on both subtests, as expected. No other comparison yielded statistically significant differences, including the differences in mean values for the ADHD subgroups. Thus, findings reflecting differences in SBH ADHD subgroups cannot be attributed to intellectual differences.

Medical Variables

Table 4 summarizes data on a variety of medical history variables. The ADHD subgroups did not differ on number of shunt revisions, $F(2, 160) < 1$ or history of seizures, $\chi^2(4, N= 160) = 2.34$, p = .67. The groups did differ significantly in ambulatory status, $\chi^2(4, N= 164) = 10.21$, $p = .04$, and lesion level, $\chi^2(2, N= 164) = 8.09$, $p< .05$, reflecting the absence of nonambulatory participants or thoracic-level lesions in the ADHD-C subtype. This association of ambulatory status and lesion level is expected because those with higher lesions have restricted movement.

Cognitive Measures

A one-way ANOVA, with group as the between-participants variable, was conducted on each of the dependent measures for the various cognitive tasks. The dependent variables were the standard scores for Numbers Reversed and Concept Formation subtests (M = 100; SD = 15) and *z*-scores for the CVLT-C. Using the between-group error term, preplanned comparisons were made to directly examine the validity of the hypotheses with the data from each cognitive test. The means and standard deviations for the groups on each cognitive dependent variable appear in Table 5.

The hypothesis that children with SBH would perform significantly poorer on all the cognitive measures relative to the normal comparison group was supported. Specifically there was a significant difference on the Numbers Reversed subtest, $F(1, 54) = 3.97, p < .05$, $\eta^2= .07$, the Concept Formation subtest, $F(1, 196) = 96.01, p < .0001, \eta^2= .32$, Trial 1 on the CVLT-C, $F(1, 200) = 32.15, p < .0001, \eta^2=.13$, Trial 5 on the CVLT-C, $F(1, 200) = 45.47, p < .0001, \eta^2= .18$, and the Semantic Clustering variable from the CVLT-C, $F (1, 200) = 3.87, p < .05, \eta^2= .02$. No significant differences were found involving any ADHD subgroups on the cognitive measures.

Table 4 Medical Variables Associated with SBH by Group.

	SBH		
Variable	Non-ADHD	ADHD-I	ADHD-C
Lesion level ***n* (%)**			
N	112	38	14
Lower (< T12)	77 (69)	27 (71)	14 (100)
Higher (> L1)	35 (31)	11 (29)	0 (0)
Shunt Revisions			
M	2.05	2.79	2.50
SD	3.41	3.06	2.60
Seizures *n* **(%)**	24 (22)	7 (20)	3 (21)
Ambulation *n***(%)**			
Independent	18 (16)	11 (29)	3 (21)
Partial	37 (33)	11 (29)	9 (64)
Unable	57 (51)	16 (42)	2 (14)

Note. SBH = spina bifida meningomyelocele and hydrocephalus; ADHD = attention deficit/hyperactivity disorder; I = inattentive Type; C = combined type. The total for ADHD-C includes one child who met diagnostic criteria for ADHD, hyperactive- impulsive type.

Table 5 Means and Standard Deviations for Executive Function Measures by Group.

Variable	Control	SBH		
		Non-ADHD	ADHD-I	ADHD-C
WJ-Numbers Reversed				
N	41	111	37	14
M	108.37	89.20	93.16	95.21
SD	16.62	25.44	14.92	16.09
WJ-Concept Formation				
N	41	108	37	14
M	111.12	83.78	77.73	80.36
SD	13.66	18.14	13.54	15.10
CVLT-C Trial 1 (*z* scores)				
N	41	113	36	14
M	0.62	-0.59	-0.47	-0.57
SD	1.00	1.11	0.98	1.22
CVLT-C Trial 5 (*z* scores)				
M	0.40	-1.25	-1.61	-1.00
SD	1.43	1.43	1.20	1.40
CVLT-C Semantic Cluster (raw scores)				
M	0.24	-0.31	-0.21	0.00
SD	0.95	1.17	1.00	1.27

Note. SBH = spina bifida meningomyelocele and hydrocephalus; ADHD = attention deficit/hyperactivity disorder; I = inattentive Type; C = combined type; CVLT-C = California Verbal Learning Test–Children. The total for ADHD-C includes one child who met diagnostic criteria for ADHD, hyperactive- impulsive type.

Parent Rating of Attention/Executive Functions

To analyze the BRIEF parent rating scales, the average item response for each of the nine domains was used as the dependent variable. A one-way multivariate analysis of variance (MANOVA) using preplanned comparisons (normal controls vs. SBH, SBH–ADHD vs. SBH–no ADHD, SBH–AD/H-C vs. ADHD-I) was used to analyze the nine domains with group as the between participants variable. Univariate follow-up analyses were conducted on specific domain scores with a Bonferonni correction (0.05/3 = .0167) applied for multiple comparisons.

Table 6 provides BRIEF scale scores for each group. All three hypotheses were supported by the multivariate test. The group effect of SBH was significant, $F(27, 552)$ = 7.34) = 12.94, $p < .0001$, $\eta^2 = .79$. There was also a significant effect of ADHD group membership, $F(9, 182) = 10.72$, $p < .0001$, $\eta^2 = .35$. For the third comparison, parents rated the ADHD-C group as different from the ADHD-I group on the BRIEF, $F(9, 182) = 4.82$, $p < .0001$, $\eta^2 = .19$.

To investigate the impact of the main effect on specific domain scales, univariate follow-up analyses were conducted at $p < .05$ for each scale, but controlling for the number of preplanned comparisons (.05/3 = .0167). The hypothesis that children classified as ADHD-C would have higher ratings than those classified as ADHD-I on the Sustain and Inhibit Scales was supported for the Inhibit Scale, $F(1, 190) = 24.11$, $p<.0001$, $\eta^2 =.19$, but not for the Sustain Scale $F(1, 190) < 1$. The hypothesis that children classified as ADHD-I

Table 6 Means and Standard Deviations for the BRIEF Parent Rating Scale by Group.

		SBH		
Variable	Control	Non-ADHD	ADHD-I	ADHD-C
N	39	106	36	13
Emotional Control				
M	1.31	1.65	1.84	2.20
SD	0.41	0.48	0.64	0.44
Inhibiting				
M	1.25	1.42	1.63	2.33
SD	0.37	0.47	0.45	0.41
Initiating				
M	1.44	1.80	2.50	2.49
SD	0.43	0.44	0.48	0.46
Organization				
M	1.62	1.82	2.59	2.47
SD	0.51	0.50	0.51	0.46
Planning				
M	1.37	1.77	2.55	2.53
SD	0.40	0.48	0.49	0.51
Shifting				
M	1.29	1.71	2.01	2.32
SD	0.37	0.46	0.57	0.52
Self-Monitoring				
M	1.45	1.77	2.32	2.42
SD	0.39	0.47	0.44	0.51
Sustaining				
M	1.38	1.83	2.45	2.56
SD	0.42	0.45	0.47	0.55
Working Memory				
M	1.45	1.77	2.57	2.70
SD	0.52	0.56	0.54	0.68

Note. SBH = spina bifida meningomyelocele and hydrocephalus; ADHD = attention deficit/hyperactivity disorder; BRIEF = Behavior Rating Inventory of Executive Function.

would have higher ratings than the children classified as ADHD-C on the Shift, $F(1, 190) = 3.92$, $p < .05$, and Initiate, $F(1, 190) < 1$, scales was not supported as the Shift Scale difference did not meet the critical level of alpha (.0167) and was not in the predicted direction. Finally, although not directly related to the hypothesis, comparisons of children with SBH (collapsing ADHD subgroups) vs. controls, and SBH–no ADHD and SBH–ADHD were not significant in a post hoc analysis of the other five scales. No comparisons of the two ADHD subtypes met the critical level of alpha (0.167). Only the Emotional Control scale showed a tendency for higher ratings in those with SBH and ADHD-C, $F(1, 190) = 4.90$, $p < .03$.

Relation of the SNAP-IV and BRIEF

The SNAP-IV and BRIEF are behavioral rating measures that are assumed to measure similar but not identical behaviors and also share variance because parents complete both (i.e., method variance). Significant correlations between ratings on these scales are therefore expected (See Table 7). These correlations were all significant, but generally in the low to moderate range for the SNAP-IV Impulsivity/Hyperactivity Scale

Table 7 Pearson Correlations of the SNAP-IV Parent Rating Scale and the BRIEF Parent Rating Scale.

	SNAP-IV Parent Rating Scale	
BRIEF Parent Rating Scale	Inattention*	Hyperactivity/Impulsivity*
Emotional Control	0.41	0.41
Inhibiting	0.40	0.60
Initiating	0.67	0.33
Organization	0.66	0.26
Planning	0.70	0.34
Self-Monitoring	0.59	0.33
Shifting	0.53	0.34
Sustaining	0.70	0.45
Working Memory	0.72	0.40

Note. *$p < .003$; SNAP-IV = Swanson, Nolan, Achenbach, Pelham-IV; BRIEF = Behavior Rating Inventory of Executive Function.

and in the moderate to high range for the SNAP-IV Inattention Scale. Of interest is the magnitude of the correlations of the SNAP-IV Inattention with BRIEF Organization, Initiating, Planning, Sustaining, and Working Memory scales (range = 0.66–0.72). With the exception of Initiating, these scales would be commonly interpreted to represent executive functions thought to be mediated by anterior brain systems.

DISCUSSION

In terms of the primary objective of the study,the incidence of ADHD in this sample of children with SBH (31%) was significantly higher than the prevalence of ADHD in the general population (upper limit of 17%), the comparison group in this study (5%) based on parent rating scales, and the normative sample for the SNAP-IV (about 8%). We also found that most participants with SBH and ADHD met criteria for the Predominantly Inattentive Type of ADHD (23%). Specifically, the incidence of ADHD-I is much greater than expected from prevalence studies, but the incidence of ADHD-C and ADHD-HI are comparable to lower-bound estimates in the general population. The overall increased rate of ADHD in children with SBH (33%) was consistent with the incidence found by Ammerman et al. (1998). Although this latter study did not examine prevalence of ADHD subtypes, children classified as ADHD were rated as displaying more inattentive behaviors than hyperactive or impulsive behaviors, similar to the present study. More generally, these results suggest that behaviors associated with distractibility, lack of focus, and disorganization are more associated with SBH than hyperactive, impulsive behaviors. Examining responses to the SNAP-IV Hyperactive-Impulsive scale, appropriate because the subtypes did not differ on the Inattention scale, showed that the few children with elevations on the Hyperactive-Impulsive scale had parental endorsements involving excessive verbal behavior as opposed to true hyperactivity or impulsivity. Hyperverbosity is commonly attributed to children with SBH and reflects in part the discourse level difficulties many experience with language pragmatics (Dennis et al., 1994).

Ammerman et al. (1998) found that children with spina bifida who met criteria for ADHD did not differ on age, gender, ambulation status, or lesion-level variables from their spina bifida–no ADHD sample. In contrast, we found significant differences in lesion level and ambulation among the ADHD subtypes. Specifically, the ADHD-C children were more likely to have spinal lesions below the thoracic level and, as would be expected, were more independent in their ambulatory capabilities than those with ADHD-I or without ADHD. The small number of children with SBH and ADHD-C also tended to have higher levels of intellectual functioning. Thus, the children in the ADHD-C category may have more opportunity to display hyperactive/impulsive behaviors and more capacity for verbosity than those in the ADHD-I subtype. Children with SBH and ADHD (regardless of subtype) were similar in other medical characteristics to the sample of children with SBH with no ADHD, so the treatment of hydrocephalus and seizures do not explain these differences. Lower spinal lesions are generally associated with higher levels of cognitive functioning and less severe physical and neurological deficits (Fletcher, Copeland, et al., 2005; Hunt & Holmes, 1975; Northrup & Volcik, 2000; Shaffer, Friedrich, Shurtleff, & Wolf, 1985). The difference in our study and Ammerman et al.'s (1998) may reflect the larger size of our sample and the inclusion of a large Hispanic cohort in our study; Hispanics are more likely to experience upper level (thoracic) lesions (Fletcher, Copeland, et al., 2005).

Children with SBH showed deficiencies on measures of executive functions compared to the participants with no neurological abnormalities. Several previous studies have found differences between children with SBH and typically developing comparison children on measures of executive functioning (Fletcher et al., 1996; Snow, 1999) and the CVLT-C (Yeates et al., 1995). In general, effect sizes were in the small-to-moderate range for all executive function measures, replicating previous studies that reported smaller differences on these types of measures than on measures of motor and spatial skills, math, and other domains (Fletcher et al., 1996). These differences do not establish that SBH is specifically associated with cognitive deficits in executive functions because no control tasks were included. Fletcher et al. (1996) found that other task requirements (e.g., motor speed, attention) typically accounted for differences in executive functions. It may well be that the executive function difficulties manifested by children with SBH are actually problems stemming from difficulties with posterior attention systems and the cerebellum, which would be consistent with the nature of the brain anomalies associated with SBH (Dennis, Edelstein, Copeland, et al., in press; Dennis, Edelstein, Frederick, et al., 2003; Fletcher Copeland, et al., 2005). More experimental research is necessary to evaluate these hypotheses.

The main question of interest for the cognitive tasks was whether children with SBH would differ in performance when divided into groups based on the ADHD classification, which was not the case. Due to the retrospective nature of this study, the measures used to assess executive functions were not specifically designed to test for differences among ADHD subtypes, so the null results may reflect selection of the tasks. However, recent research suggests that while non-brain-injured ADHD subtypes are difficult to differentiate on a wide range of executive functions (Chhabildas, Pennington, & Witcutt, 2001; Houghton et al., 1999), they may differ on more specific tasks of executive functioning, such as response inhibition (Nigg et al., 2002) and planning (Klorman et al., 1999). Thus, other cognitive measures not used in this study may discriminate AD/ HD subtypes with SBH, but we obtained essentially null results that may reflect lack of sensitivity of the tasks to executive function differences.

Consistent with our findings, behavior rating measures have consistently been better than cognitive measures at revealing differences between children who have ADHD and those who do not (Barkley, 1998). This observation is not surprising given that these measures directly assess relevant criterion behaviors and are completed by the same parent who completes the criterion rating scales used to assess ADHD behaviors. Shared method variance, however, does not explain the weak patterns of subgroup differences. Children with SBH were rated as significantly worse than control children on the BRIEF on the first multivariate comparison. The ratings also differentiated the children classified as ADHD and non-ADHD on the second multivariate comparison. This suggests that children with SBH classified as having ADHD show greater executive-functioning deficits than SBH children not classified as ADHD. The third multivariate comparison also showed that parents also rated children with ADHD-C as having more executive-functioning problems than children with ADHD-I. However, only the Inhibition scale met the critical level of alpha, showing that children with SBH and ADHD-C were rated as more disinhibited than those with SBH and ADHD-I. Table 6 shows a consistent trend for the group with SBH and ADHD-C to obtain higher ratings on scales associated with impulsivity and disinhibition (Emotional Control, Self-Monitoring, and Sustaining), a pattern that helps to account for the significant multivariate effect.

The correlations of the SNAP- IV and BRIEF suggest that behaviors subsumed under the attention problems category and under the executive functions rubric are related. This raises the question of how best to account for the difficulties with planning, organization, and other executive functions commonly observed in children with SBH. Are these difficulties products of frontal impairment, lack of access to the frontal lobes (Wills, 1993), the posterior attention system (Dennis, Edelstein, Copeland, et al., in press; Dennis, Edelstein, Frederick, et al., in press), or loss of white matter related to a nonverbal learning disability (Rourke, 1989)? Of interest is the evidence from structural neuroimaging studies showing relative preservation of frontal areas with loss of white and gray matter posteriorly (Fletcher, Copeland, et al., 2005) and the increasing evidence from experimental studies of attention in children with SBH specifically implicating the posterior attention system (Brewer et al., 2001; Dennis, Edelstein, Copeland, et al., in press; Dennis, Edelstein, Frederick et al., in press). Although resolution of this question is beyond the scope of the present study, impairment of the posterior attention system is consistent with the major finding that attention problems are relatively specific in form in children with SBH, largely involving behaviors associated with distractibility, reduced arousal, and disorganization seen in ADHD-I.

There are several limitations to the present study. Despite a large sample size, there were a relatively small number of children classified as ADHD-C and ADHD-HI in the sample. This affected the power to detect differences between this group and the other groups, especially as effect sizes were generally in the small range at best. Another limitation was the method used for classification of ADHD children. Using parent rating scales as a means of identifying ADHD has been shown to identify more children than using structured interview methods (Barkley, 1998). In addition, due to the non-normality of ADHD rating scales, the prevalence of ADHD in this sample may be overestimated (Swanson et al., 2000), though normative studies are subject to the same limitations and the rate in the normal control group was exactly as expected (5%). This problem would not lead to subtype differences. Finally, the control group tended to perform in the upper part of the average range, which may lead to larger effect sizes on the cognitive measures. The critical question, however, involved comparisons of the SBH subgroups, which were largely comparable on IQ and demographic measures, and yielded patterns of largely small to negligible effect sizes.

The results of this study suggest several avenues for further research. It would be interesting to compare children with SBH identified as having ADHD-I and ADHD-C to children in the non-brain-injured population identified with these ADHD subtypes on cognitive and behavioral measures. Specifically, a comparison of the types of hyperactive, impulsive, and inattentive behaviors reported would be helpful in determining whether these children have similar behavioral profiles. The use of attention tasks more directly related to neuropsychological and cognitive theories of attention and the brain, as in Brewer et al. (2001) and Dennis et al. (Dennis, Edelstein, Copeland, et al., in press; Dennis, Edelstein, Frederick, et al., in press), would be interesting. This research would be enhanced by exploration of the neuroanatomical correlates associated with ADHD in the SBH population. Regardless of these limitations and future directions, these results document what practitioners have reported: children with SBH commonly show evidence for attention difficulties. Future research should systematically explore the efficacy of interventions, including stimulant medication, commonly used to treat attention problems. These interventions may be especially important given the significant learning and social difficulties experienced by many children and adults with SBH.

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