Attention and Executive Functions in Adolescents with Spina Bifida

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Objective This study was designed to examine attention processes and executive functioning in adolescents with spina bifida, and to explore whether impairment in these domains contributes to problems with social adjustment. **Methods** A sample of adolescents with spina bifida ($n = 68$) and a matched comparison group ($n = 68$) and their families were followed longitudinally. All participants completed questionnaires, and the adolescent participants underwent neurocognitive testing. **Results** The spina bifida sample showed greater impairment on objective and subjective measures of attention and executive functioning, even when differences in intellectual functioning were controlled. Additionally, attention and executive deficits were found to be predictive of social adjustment difficulties. A mediational analysis suggested the neurocognitive deficits mediate associations between spina bifida status and social adjustment difficulties. **Conclusions** Adolescents with spina bifida appear to exhibit clear impairment in attention and executive functioning and this impairment may contribute to their well-established social difficulties.

Key words attention; executive functions; hydrocephalus; neural tube defects; social adjustment; spina bifida.

Introduction

Spina bifida is a birth defect caused by incomplete neural tube development, resulting in a protrusion of spinal cord, meninges, and nerve roots through an opening in the spine (Burmeister et al., 2005). Most individuals with spina bifida develop hydrocephalus, largely due to herniation of the cerebellar tonsils through the foramen magnum (i.e., Arnold–Chiari II malformation). This condition obstructs ventricular flow in the third and/or fourth ventricles, leading to a posterior → anterior progression of ventricular dilation, and causing stretching of nearby white matter tracts and compression of the cortex (Bruner et al., 1999; Fletcher et al., 1996a; Fletcher et al., 2000). Although children with spina bifida tend to score within the low average to average range on measures of general intellectual ability, they are at heightened vulnerability to learning disabilities and other cognitive difficulties (Brewer, Fletcher, Hiscock, & Davidson, 2001; Fletcher et al., 1996b; Wills, 1993), and tend to exhibit increased problems with socialization, academic functioning, and vocational accomplishment as they age (Dise, & Lohr, 1998; Holler, Fennel, Crosson, Boggs, & Mickle, 1995). The role of higher order cognitive abilities (e.g., attention, executive functions) in these functional outcomes is somewhat unclear and has received limited empirical attention.

There is substantial evidence of attention problems associated with spina bifida (Wills, 1993). Fletcher et al. (1996b) reported that children with shunted hydrocephalus performed more slowly on simple tests of attention and processing speed. Loss, Yeates, and Enrile (1998) found clear evidence of problems in multiple attentional domains among children with spina bifida and hydrocephalus compared to their siblings, even on tasks not requiring rapid motor output. Furthermore, they found that these attentional deficits were associated with poor achievement and parent-reported behavioral problems suggestive of attention deficits. In a more recent study, Brewer et al. (2001) compared children with congenital hydrocephalus (predominantly owing to spina bifida), children with Attention Deficit Hyperactivity Disorder (ADHD), and typically developing children on several tests of attention. Using Mirsky's (1996) multidimensional model of attention as a guide, they found

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that children with hydrocephalus exhibited relatively greater difficulty with ''focused attention'', presumably because of its association with corpus striatal and inferior parietal regions of the brain that are particularly vulnerable to the effects of hydrocephalus. In contrast, children with ADHD showed primary deficits on measures of ''sustained attention''. Despite these differences in presentation, researchers have found an ADHD prevalence rate of 31–33% among children with spina bifida, a rate that is considerably higher than the 3–5% prevalence rate for the general child population reported in the DSM-IV (American Psychiatric Association, 1994; Ammerman et al., 1998; Burmeister et al., 2005). Some researchers have suggested that ADHD within the context of spina bifida and hydrocephalus is better characterized by the inattentive subtype of the disorder, rather than the impulsive/hyperactive subtype (Burmeister et al., 2005).

Executive functions are a heterogeneous group of higher order cognitive abilities that include self-regulation, inhibition, planning, mental flexibility, and organization of behavior (Eslinger, 1996). Although executive dysfunction is often associated with frontal lobe damage, it also can result from damage to other brain areas. Damage to the frontal-subcortical white matter circuits, which occurs commonly in congenital hydrocephalus, can disrupt communication between the prefrontal cortex and other areas of the brain, resulting in executive dysfunction (Denckla, 1996; Fletcher et al., 1996b).

Mahone, Zabel, Levey, Verda, & Kinsman (2002) studied a small sample of adolescents with spina bifida, and found high levels of parent-reported problems with initiation, working memory, planning/organization, self-monitoring, metacognition, and emotional control on the Behavior Rating Inventory of Executive Function (BRIEF; Gioia, Isquith, Guy, & Kenworthy, 2000) relative to the published norms. Snow (1999) found evidence of set-shifting and problem-solving difficulties among children with spina bifida, even when IQ was statistically controlled. Fletcher et al. (1996b) designed one of the few comprehensive studies of executive functions and attention in school-aged children with hydrocephalus, and found that hydrocephalus was associated with problem-solving difficulties. However, rather than attributing this to global deficits in executive functioning, they hypothesized that children with hydrocephalus make more errors because of distinct problems with intention/initiation. They reasoned that hydrocephalus and shunt-related damage to the right posterior region of the brain associated with arousal and activation could result in poor initial engagement in a task.

Social and emotional problems also are common within the context of spina bifida. Children and adolescents with spina bifida have higher rates of internalizing behavior and psychiatric diagnoses than the general population, and tend to experience more problems with social relationships and autonomy development (Ammerman et al., 1998; Holmbeck et al., 2003; Landry, Robinson, Copeland, & Garner, 1993). There are likely a number of factors driving these social difficulties, including visible signs of disability (e.g., wheelchair use), lower scholastic achievement, and reduced physical activity (Holmbeck et al., 2003). Fletcher et al. (1996b) proposed an interesting theory: that problems with higher order cognitive functions (such as attention, intention, and initiation) mediate the influence of hydrocephalus and its associated physiological complications on social adjustment. This link between executive functions and social competence also was suggested by Landry et al. (1993), who reasoned that children who show deficits in planning and problem-solving may have difficulty identifying and performing appropriate social behaviors across different social situations. The possibility that executive functions and attention influence social functioning in these children is intriguing, but has not been tested directly.

The current study compared a sample of adolescents with spina bifida and a demographically matched sample of typically developing adolescents on a range of measures of attention and executive functioning, as well as two measures of social adjustment. Based on past literature, there were three specific hypotheses. First, it was expected that adolescents with spina bifida would perform more poorly on measures of visual attention, particularly on tests involving scanning and detection (focused visual attention), as opposed to sustained attention. Second, it was expected that adolescents with spina bifida would perform more poorly than age-matched peers on measures of planning, organization, and initiation, even when a proxy for intellectual ability was held constant. Third, it was expected that performance on measures of attention and executive functioning would correlate positively with social functioning.

Method

Participants

The participants were drawn from a longitudinal study focusing on family relationships and psychological adjustment in children with spina bifida (Holmbeck et al., 1997, 2002, & 2003). Children with spina bifida between the ages of 8 and 9 were recruited for

participation through local children's hospitals and a statewide spina bifida association. Participants in the comparison sample were recruited from the schools where the children with spina bifida were enrolled. Participating families were followed every two years through four waves of data collection. During the first data collection period (Time 1), 68 children with spina bifida and 68 typically developing children and their families participated in the study. The samples were matched across ten major demographic variables, including child age, child ethnicity, and socioeconomic status. By the fourth wave of data collection (Time 4), there were 59 participants in the spina bifida sample and 65 in the comparison sample (91% retention, total $n = 124$). Those who dropped out of the study did not differ significantly from remaining participants in age, gender ratio, intellectual ability (PPVT-R score), or (in the spina bifida sample) spina bifida lesion level. Across both samples, 67 (54%) of the Time 4 participants were male, and 57 (46%) were female. Mean age for the adolescent participants at Time 4 was 14.55 ($SD = .63$).

Among the spina bifida participants at Time 4, 32% had sacral level spinal lesions, 54% had lumbosacral or lumbar lesions, and 13% had thoracic lesions. The majority (71%) was shunted for hydrocephalus. The average number of shunt surgeries (including initial placement and subsequent revisions) was 5.16 ($SD = 8.61$; although there were a few outliers with an extremely high number of surgeries, range = 0 to 41 surgeries). Six (10%) of the spina bifida participants were described by their parents as having seizures or taking medications for seizures. Per parent report, adolescents in the spina bifida sample were significantly more likely to carry a diagnosis of ADHD ($n = 18$, 32% of respondents) than those in the comparison sample ($n = 6$, 10% of respondents; $\chi^2 = 810$, $p = .004$). Refer to Holmbeck et al. (1997, 2002) for complete demographic information, including ethnicity, socioeconomic status, and family structure.

Procedure

For each wave of data collection, trained research assistants conducted home visits with the participating families. During the visits, family members completed questionnaires and participated in videotaped family interaction tasks. Additionally, the child participants were asked to participate in brief neurocognitive testing (described subsequently) at Time 1 and Time 4. Participating families were asked to sign release forms allowing the research team to contact the child's teacher. Questionnaires were mailed to the specified teacher after the home visit. The teacher questionnaire return rate was 90% at Time 1 and 89% at Time 4. Approval was obtained from all relevant institutional review boards prior to initiation of the study, and all participants (including teachers) were compensated for their time at each round of data collection.

Measures: Parent/Teacher Questionnaires

Demographics

At each visit, parents completed a brief measure designed to obtain demographic information, child medical and psychiatric diagnoses (e.g., ADHD diagnosis), educational information, and other general family information.

Questionnaire Measure of Executive Functioning

BRIEF (BRIEF; Gioia et al., 2000) is a multidimensional measure of parent- and teacher-reported executive functioning that correlates well with other measures of attention and behavioral control. With permission from the authors, questions from a pre-publication edition of the measure were administered to parents and teachers at Time 4. This early edition of the BRIEF consists of nine subscales derived through statistical analyses and expert feedback, five of which were selected for use in the current study (the Cronbach's alpha ranges for all three raters are provided in parentheses): Initiate ($\alpha = .80$ to .89), Sustain ($\alpha = .82$ to .85), Organize ($\alpha = .87$ to .93), Plan ($\alpha = .87$ to .94), and Working Memory ($\alpha = .86$ to .93). Alphas for the full measure ranged from .96 to .98 across raters. The five selected scales consisted of 42 items (e.g., ''Underestimates time needed to finish tasks'') that were rated by parents or teachers as ''never,'' ''sometimes,'' or ''often'' a problem for the child. Because the mother- and father-reported BRIEF scores were moderately correlated ($r = .50$ to .63 across subscales), the mean across both parents was used when reports from both parents were available.

Social Adjustment

Parent- and teacher-rated social competence was assessed with the Self-Perception Profile for Children (SPPC; Harter, 1985) at Time 4. The 15-item parent and teacher forms of the measure (i.e., the Rating Scale of Child's Actual Behavior) were used. This measure assesses ratings of a child's competence (compared to peers) on three items within each of five domains. Of the original five scales, only the Social Competence scale was used in the current study (internal consistency $\alpha = .79$ to .94 across reporters). Parent scores were combined

when both parents' ratings were available ($r = .60$ between mother and father report).

A second measure of social functioning, the Social Skills Rating System (SSRS; Gresham & Elliott, 1990) was administered to parents and teachers at Time 4. The measure consists of 30 items that rate a child's social behavior and assertiveness on a scale from 0 (the child never does this behavior) to 2 (the child very often does this behavior). Internal consistency values for the entire combined scale ranged from .92 to .94 across all three reporters. Parent scores were combined ($r = .55$) when two reporters were available.

Measures: Examiner-Administered Neuropsychological Instruments

Intellectual Functioning

At Time 1, each child was administered the Peabody Picture Vocabulary Test, Revised (PPVT-R; Dunn & Dunn, 1981), a test of receptive language that was used as a proxy for general intellectual ability in the current study. The test serves as a particularly useful estimate of intellectual ability with the population under study because its relatively rapid administration does not penalize children who have difficulty sustaining attention for long periods of time and it does not require fine motor output like many other measures of intellectual functioning. The test manual reports that the measure correlates moderately well with the WISC and WISC-R Full Scale IQ and Verbal IQ scores.

Examiner-Administered Measures of Attention and Executive Functioning

At Time 4, the adolescent participants were administered portions of a multidimensional neurocognitive measure, the Cognitive Assessment System (CAS; Naglieri & Das, 1997). Five CAS subscales were used in the current analyses. The first three subscales (i.e., Matching Numbers, Planned Codes, Planned Connections) measure a child's planning ability. The two remaining subscales (i.e., Number Detection and Receptive Attention) measure focused visual attention, described in Mirsky's (1996) model as the capacity to concentrate attentional resources on a specific task while filtering irrelevant stimuli.

Results

As noted above, 59 adolescents with spina bifida and 65 matched comparison adolescents were included in the Time 4 analyses ($n = 124$). At Time 1, the two samples were found to differ significantly in estimated intellectual functioning (PPVT-R score). The spina bifida sample performed in the lower end of the average range ($M = 92.49$) and the comparison sample performed in the upper end of the average range ($M = 108.97$, $t = -5.68$, $p < .001$). Thus, it was decided that PPVT-R score would be used as a covariate in these analyses so that group differences could be attributed more confidently to differences in these higher order abilities, and not to differences in overall intelligence.

Neurocognitive Features of Spina Bifida

To test the first two hypotheses and develop an initial profile of the neurocognitive strengths and weaknesses in early adolescents with spina bifida, the two samples were compared on multiple measures of attention and executive functioning. First, the samples were compared on all CAS subtests using multivariate analysis of variance (MANOVA). Because some families chose for time reasons to complete questionnaires, but not to participate in neuropsychological testing during the fourth wave of data collection, 50 participants with spina bifida and 62 participants from the comparison sample were administered the CAS. Those who opted out of taking the CAS were not found to differ significantly from those who took the test on PPVT-R score or (among spina bifida participants) spinal lesion level. In an omnibus test, adolescents in the comparison sample performed significantly better (i.e., higher average scaled scores) than those in the spina bifida sample on the CAS, $F\ (5,\ 102) = 15.95$, $p = .000$. Follow-up univariate analyses found significant differences across all CAS subtests. Additionally, the mean scaled scores in the spina bifida sample consistently fell more than a standard deviation below those in the comparison sample, often in the range of clinically significant impairment. To ensure that the group differences could not be accounted for by general differences in intelligence, the analyses were repeated with Time 1 PPVT-R score (a proxy for intellectual functioning) entered as a covariate. Group differences remained significant on all subtests, and effect sizes (Cohen's *d*) were large for all univariate comparisons. Table I organizes the results of follow-up univariate analyses by neuropsychological construct.

Using MANOVA, significant group differences also emerged for the BRIEF (parent report) in an omnibus test, $F\ (5,\ 117) = 4.89$, $p = .000$. In univariate analyses, the spina bifida sample was rated as having significantly more impairment (i.e., ''higher'' item mean scores) on the Initiate, Sustain, and Working Memory subscales. However, the groups did not differ significantly on the

Table I. Group Differences on Measures of Attention and Executive Functioning

Construct	Measure	Source	Sample	*M*	*SD*	*n*	*Cohen's d*
Focused Attention	CAS Number Detection[a]	Test data	Spina Bifida	6.06**	3.10	50	1.37
			Comparison	10.15	2.85	61	
	CAS Receptive Attention	Test data	Spina Bifida	7.10*	4.06	50	.98
			Comparison	10.56	2.95	61	
Sustained Attention	BRIEF Sustain[b]	Parents	Spina Bifida	.75	.39	59	.42
			Comparison	.59	.38	64	
	BRIEF Working Memory	Parents	Spina Bifida	.73*	.47	59	.58
			Comparison	.48	.38	64	
	BRIEF Sustain	Teacher	Spina Bifida	.62	.47	48	.35
			Comparison	.46	0.44	62	
	BRIEF Working Memory	Teacher	Spina Bifida	.61	.57	48	.54
			Comparison	.33	.46	62	
Planning	CAS Matching Numbers	Test data	Spina Bifida	6.27**	3.15	49	1.25
			Comparison	10.18	3.11	62	
	CAS Planned Codes	Test data	Spina Bifida	6.02**	3.18	50	1.41
			Comparison	10.21	2.77	62	
	CAS Planned Connections	Test data	Spina Bifida	5.90**	3.23	50	1.45
			Comparison	10.34	2.87	61	
	BRIEF Plan Scale	Parents	Spina Bifida	.76	.41	59	0.17
			Comparison	.69	.41	64	
	BRIEF Plan Scale	Teacher	Spina Bifida	.63	.56	48	.38
			Comparison	.43	.50	62	
Organization	BRIEF Organize	Parents	Spina Bifida	.84	.47	59	.17
			Comparison	.76	.45	64	
	BRIEF Organize	Teacher	Spina Bifida	.54	.58	48	.56
			Comparison	.27	.37	62	
Initiation	BRIEF Initiate	Parents	Spina Bifida	.85*	.42	59	.50
			Comparison	.65	0.38	64	
Initiation	BRIEF Initiate	Teacher	Spina Bifida	.72	.57	48	.46
			Comparison	.47	.50	62	

Note: *Group difference significant at $p < .05$. **Group difference significant at $p < .001$.

[a]CAS subtest means are scaled scores (mean in normative sample = 10, $SD = 3$).

[b]BRIEF means range from 0 (no impairment in this domain) to 2 (significant impairment).

PPVT-R score was entered as a covariate in all analyses.

Organize and Plan subscales as expected. When PPVT-R score was included in the analysis as a covariate, the omnibus test revealed significant differences overall, F (5, 115) = 2.84, $p = .019$. However, in follow-up univariate analyses, group differences on the Sustain subscale were no longer significant, with the groups differing significantly only on the Initiate and Working Memory subscales (Table I). Effect sizes for all BRIEF univariate comparisons were in the small to medium range.

The analyses for both multidimensional measures were repeated using teachers as a collateral information source. As with the parent version, the omnibus test revealed significant differences on the teacher version of the BRIEF, F (5, 105) = 2.53, $p = .033$. Univariate tests found significant differences on all subtests as well. However, when PPVT-R score was entered as a covariate, no significant effects were found, F (5, 103) = 1.29, $p = .276$ (Table I).

It should be noted that within the spina bifida sample, shunt status (i.e., whether or not a participant required shunting for hydrocephalus) was found to be a significant predictor of performance on each of the five CAS subtests in a multivariate test and in follow-up univariate analyses: Matching Numbers, F (1, 44) = 11.54, $p = .001$, Planned Codes, F (1, 44) = 27.48, $p = .000$, Planned Connections, F (1, 44) = 23.11, $p = .000$, Number Detection, F (1, 44) = 29.97, $p = .000$, and Receptive Attention, F (1, 44) = 17.06, $p = .000$. It also was found to be a significant predictor of performance on all five BRIEF subtests, but only when parents were used as informants (shunt status did not predict BRIEF performance as

reported by teachers): Initiate, F (1, 53) = 9.36, p = .003, Sustain, F (1, 53) = 14.64, p = .000, Organize, F (1, 53) = 6.45, p = .014, Plan, F (1, 53) = 9.94, p = .003, and Working Memory, F (1, 53) = 17.88, p = .000. In each case, spina bifida participants who never required shunting performed much better than those who were shunted. Number of shunt revision surgeries and spinal lesion level were not found to be significant predictors of CAS or BRIEF performance.

Neurocognitive Correlates of Social Adjustment

Next, performances on measures of attention and executive functioning (BRIEF, CAS) were tested as cross-sectional predictors of social functioning (SSRS, SPPC) using multiple regression. PPVT-R scores were entered as a covariate in all regression analyses.

Preliminary Analyses

The groups were compared on all Time 4 social outcome measures using a series of *t*-tests. Spina bifida participants were rated as significantly more impaired on three of the four variables: parent-reported SSRS item average score (p = .008), parent-reported SPPC Social Competence score (p = .016), and teacher-reported SPPC Social Competence score (p = .008). Differences were not significant on the teacher SSRS.

Social Skills Rating Scale (SSRS)

Next, measures of attention and executive functioning were tested as predictors of social impairment using multiple regression (Table II). Group status (spina bifida vs. comparison sample) and interactions between group status and the neurocognitive predictors were added in separate steps in the analyses to determine whether the measures predicted social functioning differentially across samples (i.e., whether there were group x measure interactions). Different sources of information were used for the predictor and outcome variables in all regression models (e.g., teacher data predicting parent data) to reduce the risk of biased results due to common-method variance.

In the first multiple regression analysis, parent-reported SSRS scores were entered as the dependent variable. PPVT-R score, group status, BRIEF full scale item average (teacher report), CAS mean score (a mean of all available CAS scaled scores representing an individual's capacity for planning and attention), and interactions among these variables were entered into the equation on separate steps. CAS scores were entered after BRIEF scores to explore whether the CAS contributes to the prediction of social competence above and beyond what the simpler (and easier to administer) BRIEF contributes. When tested in this way, the BRIEF item average (teacher report) was found to be a significant predictor of social skills (parent report), $\beta = -.26$, $F = 6.82$, $p = .010$ (Table II). Specifically, greater teacher-reported executive impairment on the BRIEF was associated with poorer parent-reported social functioning. CAS performance was not a significant predictor of parent-reported social skills, a finding that held even when the CAS was considered independently of the BRIEF. There were no significant interactions between group and either measure of executive functioning to suggest spina bifida status moderates the relationship between the predictor and outcome in these analyses.

Next, teacher-reported social skills (SSRS) was entered as the dependent variable and group status,

Table II. Multiple Regression Main Effects: Neuropsychological Predictors of Social Functioning

Dependent variable (source)	Predictor (source)	β	ΔR^2	F
SSRS (parent)	Group	.227	.040	4.43*
	BRIEF mean (teacher)	−.258	.060	6.82*
	CAS mean scaled score	.060	.002	0.23
SSRS (teacher)	Group	.050	.002	.197
	BRIEF mean (parent)	−.308	.090	10.13**
	CAS mean scaled score	.231	.028	3.25
SPPC Social Competence (parent)	Group	.170	.022	2.27
	BRIEF mean (teacher)	−.049	.002	0.22
	CAS mean scaled score	.348	.067	7.24**
SPPC Social Competence (teacher)	Group	.287	.064	6.74*
	BRIEF mean (parent)	−.283	.075	8.59**
	CAS mean scaled score	.294	.046	5.49*

Note: *p < .05. **p < .01. Beta weights are standardized and indicate the direction of the effect at the step the predictor entered the equation. The BRIEF is scored so that a higher score indicates ''greater'' impairment, whereas on all other measures a higher score indicates better functioning. Thus, all main effects are in the expected direction. None of the tested interaction effects were significant when entered separately, so these results were omitted from the table. PPVT-R scores were entered as a covariate in all equations.

parent-reported BRIEF average score, and the CAS mean scaled score were entered as predictor variables. As hypothesized, BRIEF ratings, $\beta = -.31$, $F = 10.13$, $p = .002$, were found to be significant predictors of social skills (with greater impairment predicting lower-rated social skills; Table II). CAS performance was a significant predictor of teacher SSRS when PPVT-R was not in the equation, but dropped to marginal significance when PPVT-R score was controlled. Group was not a significant predictor, and there were no significant interactions.

Harter Self-Perception Profile for Children (SPPC)

In the next set of regression analyses, associations between attention/executive functioning and social competence were explored. In the first model, parent-reported social competence (SPPC Social Competence scale) was entered as the dependent variable. In separate steps, PPVT-R score, group status, BRIEF average score (teacher report), CAS mean scaled score, and interactions among the variables were entered as predictors. In this analysis, CAS performance was found to be a significant predictor of social functioning (with higher CAS scores predicting better functioning), $\beta = .35$, $F = 7.24$, $p = .008$ (Table II). Group was not a significant predictor in the regression equation and did not moderate any of the predictor-outcome relationships. In analyses involving teacher-reported Social Competence, parent-reported BRIEF scores, $\beta = -.28$, $F = 8.59$, $p = .004$, and CAS performances, $\beta = .29$, $F = 5.49$, $p = .021$, both significantly predicted Social Competence in the expected direction. Group membership also was a significant predictor of teacher-reported Social Competence, but there were no significant interactions (Table II).

Exploratory Analyses

The results described above, specifically those that showed an association between spina bifida and executive impairment, and those that showed an association between executive impairment and social difficulties, lend some support to the theory offered by Landry et al. (1993) and Fletcher et al. (1996b) that problems with planning and other executive functions mediate the relationship between spina bifida status and social functioning (i.e., spina bifida status → executive impairment → social difficulties). Thus, some exploratory mediational analyses were included to test executive functioning as a mediator of the well-established link between spina bifida and social difficulties (e.g., Holmbeck et al., 2003).

For a mediational model to be supported, four conditions must be met: (a) the predictor variable must be significantly associated with the outcome variable, (b) the predictor variable must be significantly associated with the mediator, (c) the mediator must be significantly associated with the outcome variable, after controlling for the predictor, and (d) the previously significant predictor → outcome relationship must be significantly diminished when the effects of the mediator are controlled (Rose, Holmbeck, Coakley, & Franks, 2004).

T-tests found group differences on three social outcome variables (i.e., parent SSRS, parent Social Competence, and teacher Social Competence). Regression analyses found the BRIEF to be a significant predictor of performances on two of these social outcome variables (i.e., parent SSRS and teacher Social Competence), and the CAS to be a significant predictor of parent-rated Social Competence (Table II). Therefore, three mediational analyses were planned (Fig. 1).[1]

First, the teacher-reported BRIEF score was tested as a mediator of the relationship between group and parent-reported social skills (i.e., group → teacher BRIEF → parent SSRS). Multiple regression analyses found group to be a significant predictor of the outcome variable (SSRS), $\beta = 0.24$, $F = 7.30$, $p = .008$, and of the mediator (BRIEF), $\beta = -.26$, $F = 7.68$, $p = .007$, thus meeting the first two conditions for mediation. In a third multiple regression equation, group and teacher BRIEF were tested as predictors of parent SSRS using simultaneous entry, allowing the final two conditions for mediation to be tested. The regression analysis showed that the BRIEF was a significant predictor of SSRS performance when group status was held constant (the third condition for mediation), $\beta = -.26$, $p = .006$. Condition four requires that the strength of the group—SSRS relationship drops significantly when the mediator is controlled. This was tested by using the Sobel (1988) z test, which showed that the mediator (BRIEF) accounted for a significant portion of the variance in the relationship between the predictor (group) and the outcome variable ($z = 1.97$, $p = .049$). Thus, the first mediational model was supported.

[1]The preceding regression analyses (i.e., neurocognitive correlates of social adjustment) were run with and without PPVT-R score as a covariate. The results changed minimally when PPVT-R was in the equation. Therefore, it was not entered as a covariate in the mediational analyses, as it would unnecessarily complicate the analyses and interpretation. Because the CAS was found to be a significant predictor of teacher-reported social competence only when PPVT-R was controlled, this was not tested as a mediator.

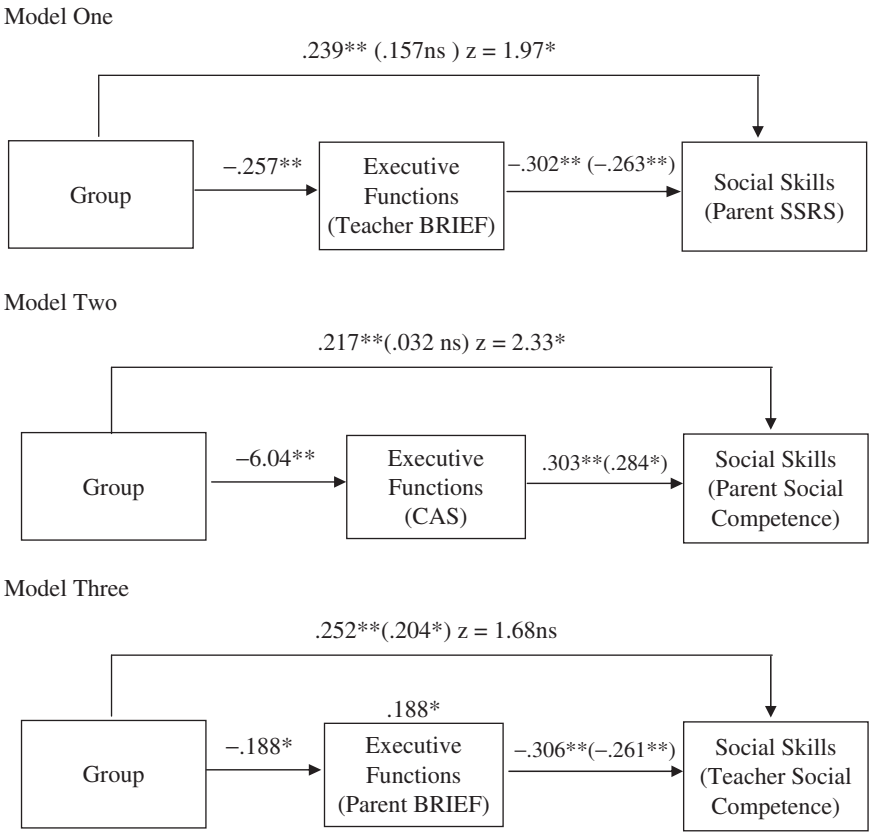

Figure 1. Mediational models.
Note: Groups were coded as spina bifida = 0, comparison = 1. *Group difference significant at $p < .05$. **Group difference significant at $p < .01$.

The second mediational model was tested in the same way. Once again, multiple regression analyses found group to be a significant predictor of the dependent variable (Social Competence) and the mediator (CAS). Additionally, the CAS was found to be a significant predictor of Social Competence when group membership was held constant. Finally, the strength of the group—Social Competence relationship dropped significantly when the mediator was in the equation ($z = 2.33$, $p = .019$), fully supporting the mediational relationship.

The third mediational model (group → parent BRIEF → teacher Social Competence) was not fully supported because the group—Social Competence effect was not significantly reduced upon entry of the mediator ($z = 1.68$, $p > .05$).

Discussion

The objectives of the current study were to identify the aspects of higher order neurocognitive functioning most affected by spina bifida and to understand the social correlates of these functions. Analyses revealed clear differences between the samples on multiple measures of attention and executive functioning, even when controlling for group differences in intellectual ability. There is some indication that the effects of hydrocephalus and/or shunt surgery are a major factor in these group differences, as shunt status (i.e., shunt vs. no shunt) was a significant predictor of CAS and BRIEF performance within the spina bifida sample, and the majority of spina bifida participants have been shunted. Performances on measures of attention and executive functioning were found to predict social adjustment (i.e., poor executive functioning was associated with impaired social competence). Additionally, mediational analyses provided some initial support for the theory that executive functioning mediates the relationship between spina bifida status and social functioning.

With respect to multidimensional measures of attention, analyses revealed statistically and clinically significant group differences on measures of focused visual attention (with the spina bifida sample showing more impairment). However, the groups did not differ

on a measure of sustained attention and differed inconsistently on the BRIEF Working Memory scale (which at the level of face validity overlaps considerably with the Sustain scale). These findings are consistent with past research suggesting that hydrocephalus impacts focused visual attention more than sustained attention because of the posterior–anterior progression of hydrocephalus in this condition (Brewer et al., 2001; Wills, 1993). On the other hand, this effect also could be attributed to differences across outcome measures, because CAS subtests were used to measure focused attention and BRIEF subtests were used to measure sustained attention. The inconsistency of assessment methods across these domains represents a potential confound.

With respect to executive functioning, the spina bifida sample showed significantly more impairment than the comparison sample on all CAS planning subtests, even when controlling for estimated intellectual functioning (PPVT-R). Their parents also reported more impairment on the parent version of the BRIEF Initiate scale, but not on the Plan or Organize subscales. Although teachers of students with spina bifida reported greater impairment on all BRIEF subscales, these differences failed to remain significant when intellectual functioning was held constant.

It was somewhat surprising that the groups would differ significantly on a performance-based measure of planning ability (i.e., all CAS Planning subtests), but not on a questionnaire measure of the same construct (i.e., BRIEF Plan scale). One reason for this discrepancy might be that the two instruments are measuring different aspects of planning ability. Whereas the CAS Planning subtests measure the examinee's ability to develop and rapidly execute problem-solving strategies (e.g., to find an efficient way of completing a coding task), the BRIEF measures social and behavioral manifestations of planning ability. Thus, adolescents with spina bifida may be more vulnerable to difficulties with rapid and efficient development of problem-solving strategies, but may not differ significantly from their peers in their ability to plan for and anticipate consequences in their daily activities. Another possible reason for the CAS–BRIEF discrepancy is that the CAS Planning subtests require rapid visuospatial shifting and a certain degree of fine motor dexterity (e.g., Planned Codes, Planned Connections). Fletcher et al. (1996b) found that spina bifida-related differences on speeded neurocognitive measures failed to remain significant when motor coordination was statistically controlled.

As noted above, no differences were found between groups on either version of the BRIEF Organize scale, and the groups differed on the parent version (but not the teacher version) of the BRIEF Initiate scale. Because organization and initiation tend to be associated with the prefrontal cortex, which remains intact (although some would argue less accessible, e.g., Denckla, 1996; Fletcher et al., 1996b) in the context of hydrocephalus, one possibility is that ''frontal'' functions are not overly impacted in individuals with spina bifida. Another possibility is that the pre-publication version of the BRIEF may not measure these domains as well as the published version, which splits the Organize construct into two new subscales: Plan/Organize and Organization of Materials (Gioia et al., 2000). In their recent study of adolescents with spina bifida and hydrocephalus, Mahone et al. (2002) found evidence of impairment on the Plan/Organize scale but not on the Organization of Materials scale of the BRIEF. Therefore, the published version of the BRIEF might be more helpful in differentiating the types of organizational difficulties associated with spina bifida.

The next major set of analyses focused on the association between attention/executive deficits and social functioning. The spina bifida sample scored lower on parent-reported social skills (SSRS) and parent- and teacher-reported social competence. The BRIEF and the CAS both emerged as significant predictors of social functioning in select analyses (with neurocognitive deficits predicting social impairment), providing further support for the theory that executive functions are associated with adaptive functioning in this population.

The BRIEF emerged as a fairly consistent predictor of social functioning (i.e., in three of four analyses) and the CAS was a predictor of social functioning in two of four analyses. Given grossly comparable predictive ability, the BRIEF (which is administered more quickly and easily than the CAS) may be a somewhat more practical instrument for predicting social deficits among adolescents with spina bifida, particularly those thought most to be at risk (i.e., those shunted for hydrocephalus). However, shared method variance limits the interpretability of these results, as both the BRIEF and the social outcome measures are retrospective questionnaires, unlike the CAS. Because earlier analyses found the CAS to be a more sensitive measure of attention and executive difficulties, both instruments might serve a unique purpose in providing thorough assessment for this population.

At the most complex level of analysis, executive functioning was examined as a mediator of the relationship between group status and social functioning. As hypothesized, executive functioning (as measured by the BRIEF and the CAS in separate analyses) was found to be a significant mediator of the relationship between spina bifida status and parent-reported social skills (i.e., spina bifida status → executive functioning → social functioning) in two analyses. This suggests that executive dysfunction may be one important mechanism by which spina bifida is associated with social adjustment difficulties. Further analysis using a broader range of neurocognitive measures would be helpful in determining the specific aspects of executive functioning that mediate this relationship.

It is important to recognize some of the limitations in this study. First, most of the analyses were cross-sectional, which prevented causal relationships from being tested. The study also might have been stronger if it had included observational measures of social functioning, which would have permitted more qualitative analysis of the aspects of social functioning most impaired in the context of spina bifida and hydrocephalus, and would have reduced the problem of shared method variance in the regression analyses. Finally, as is often the case in the study of pediatric populations, the current study was limited by a rather small sample, which limits statistical power.

The results of this study have important implications for parents, educators, and mental health providers serving individuals with spina bifida. Perhaps the most important finding is that these adolescents appear to be at higher risk for a diagnosis of ADHD and for clinically significant problems with focused attention, planning, and other executive functions. Furthermore, these attention and executive problems may be associated with social skills deficits. Although the correlational nature of the study prevents determination of whether the link between attention/executive deficits and social skills deficits is ''causal'', the observed relationship opens the door for future longitudinal studies.

The current study also has some implications for treatment. First, the current results provide tentative support for the argument made by Brewer et al. (2001) that children with hydrocephalus do exhibit attention problems, but that the attention problems tend to be somewhat different from the attention problems observed within the context of classic ADHD (i.e., they appear to exhibit problems with focused attention rather than sustained attention). The clinical implications of this should be considered. For example, is stimulant therapy (widely used in the treatment of classic ADHD with ~75% response rate; Barkley, 1997) equally effective in individuals with hydrocephalus-related attention problems? Another important issue for consideration is what steps may be taken in the classroom and beyond to address the apparent problems with initiation and planning. Skills training and organizational aids are two examples of compensatory strategies that might be used to help children with spina bifida cope with their executive deficits and ultimately experience better success in the classroom and at home (Mateer, 1999)

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