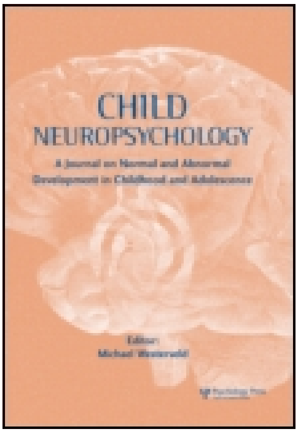

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Executive functioning and psychological adjustment in children and youth with spina bifida

Natalie C. Kelly [a], Robert T. Ammerman [b], Joseph R. Rausch [b], M. Douglas Ris [c], Keith O. Yeates [d], Sonya G. Oppenheimer [b] & Benedicta G. Enrile [d]

[a] Department of Supportive Care Medicine, City of Hope National Medical Center, Duarte, California, USA

[b] Department of Pediatrics, Cincinnati Children's Hospital Medical Center, Cincinnati, Ohio, USA

[c] Department of Pediatrics, Texas Children's Hospital, Baylor College of Medicine, Houston, Texas, USA

[d] Department of Pediatrics, Nationwide Children's Hospital, The Ohio State University College of Medicine, Columbus, Ohio, USA
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CE Executive functioning and psychological adjustment in children and youth with spina bifida

Natalie C. Kelly[1], Robert T. Ammerman[2], Joseph R. Rausch[2], M. Douglas Ris[3], Keith O. Yeates[4], Sonya G. Oppenheimer[2], and Benedicta G. Enrile[4]

[1]Department of Supportive Care Medicine, City of Hope National Medical Center, Duarte, California, USA
[2]Department of Pediatrics, Cincinnati Children's Hospital Medical Center, Cincinnati, Ohio, USA
[3]Department of Pediatrics, Texas Children's Hospital, Baylor College of Medicine, Houston, Texas, USA
[4]Department of Pediatrics, Nationwide Children's Hospital, The Ohio State University College of Medicine, Columbus, Ohio, USA

Children and adolescents with spina bifida are at risk for poor neuropsychological functioning and psychological outcomes. The relationship between executive functioning and psychological adjustment is an area worthy of investigation in this population. The current study assessed executive functioning and psychological outcomes in a group of children and adolescents with spina bifida (SBM) ($n = 51$) and nondisabled controls ($n = 45$). A mediation model was hypothesized, such that Metacognition, as measured by the Behavior Rating Inventory of Executive Function (BRIEF), mediated the relationship between group status (spina bifida versus nondisabled controls) and psychological outcomes. Results indicated that metacognitive skills fully explained the relationship between group and internalizing and depressive symptoms as reported by mothers. In particular, specific components of the BRIEF Metacognition composite were most responsible for this relationship, including Initiate, Working Memory, and Plan/Organize. The study limitations include its cross-sectional nature that precludes drawing conclusions about causality. The results have implications for treatment interventions for children and adolescents with spina bifida and typically developing individuals.

Keywords: Executive functioning; Spina bifida; Children; Adolescents; Psychological.

Epidemiology and Neuropathology of Spina Bifida/Myelomeningocele

Spina bifida/myelomeningocele (SBM) is a congenital malformation that occurs due to failure of neural tube closure between the third and fourth weeks of gestational

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Address correspondence to Robert T. Ammerman, Cincinnati Children's Hospital Medical Center, 3333 Burnet Avenue, ML 3015, Cincinnati, OH 45229, USA. E-mail: robert.ammerman@cchmc.org

age (Frey & Hauser, 2003). SBM occurs at a rate of approximately 1.90 per 10,000 live births in the United States. The rate of neural tube defects is higher in individuals of Hispanic descent than in African Americans in the United States (Centers for Disease Control and Prevention, 2009). Prevalence also varies internationally, with higher rates observed in Northern Ireland, England, and China (Wills, 1993). SBM can be associated with a variety of associated neuropathological features. One of the most common is the Arnold Chiari-II (ACII) malformation, which involves herniation of a compressed cerebellum that is displaced through the foramen magnum and obstructs cerebrospinal fluid (CSF) flow through the fourth ventricle and in some cases the third ventricle, often resulting in hydrocephalus. Additional associated features of SBM include callosal agenesis, tectal "beaking" (stretched tectum), polymicrogyria, and heterotopias (Fletcher, Dennis, & Northrup, 2000; Stevenson, 2006).

Neuropsychological Outcomes in SBM

Children with SBM and hydrocephalus have demonstrated a generally consistent pattern of intellectual functioning across research studies. Wills, Holmbeck, Dillon, and McLone (1990) reviewed the IQ scores of children and adolescents with SBM and compared them to norms. Results indicated that the SBM group scored below the population average but largely within the normal range across most tests. Deficits were apparent in Performance IQ (PIQ), which was significantly lower than Verbal IQ (VIQ) for children with SBM (Wills et al., 1990).

The neuropsychological profile of children with SBM often reveals a distinct pattern of relative strengths and weaknesses across domains. Children with SBM demonstrate impairments in language functioning. Specifically, the overall structure of language is generally preserved, whereas the actual content appears to be compromised (Fletcher et al., 2000). In addition, visual-spatial/perceptual skills are an area of difficulty for children with SBM. For example, in a study by Fletcher and colleagues (Fletcher et al., 1992), children with SBM performed more poorly than controls on measures of visuoperceptual skills. These difficulties are evident on tasks even when the motor component is minimized (Fletcher et al., 2000). Additional areas of cognitive impairment in children with SBM include verbal learning and retrieval and working memory (Boyer, Yeates, & Enrile, 2006; Scott et al., 1998; Yeates, Enrile, Loss, Blumenstein, & Delis, 1995).

One of the most consistent areas of impairment in children with SBM is executive functioning. The term "executive function" is generally used to describe a group of abilities that are responsible for controlling goal-directed cognitive, behavioral, and emotional functioning (Gioia, Isquith, Retzlaff, & Espy, 2002). Studies have consistently demonstrated that children with SBM have difficulty with executive skills, and this is often most clearly evident in everyday behavioral activity. In a study by Rose and Holmbeck (2007), parents of children with SBM reported more everyday executive functioning impairment on the Behavior Rating Inventory of Executive Function (BRIEF; Gioia, Isquith, Guy, & Kenworthy, 2000). Specifically, results indicated that children with SBM had more impairment on the Initiate, Sustain, and Working Memory subscales. The Sustain subscale was no longer significant after controlling for IQ. In addition, children with SBM showed significantly more impairment than a comparison group on measures of planning ability after IQ was controlled. In another study using the BRIEF, children with SBM were rated by their caregivers as having more difficulties than controls (Burmeister et al., 2005). These findings were significant for the Emotional Control subscale. Mahone and colleagues (Mahone, Zabel, Levey, Verda, & Kinsman, 2002) found that parents of children with SBM rated their children as having more problems on the BRIEF when compared to norms. These differences were significant for four of the five Metacognition Index subscales (i.e., Initiate, Working Memory, Planning/Organization, and Monitor) and one Behavioral Regulation Index subscale (i.e., Emotional Control). Taken together, these results suggest that executive functioning difficulties in children and youth with SBM interfere with their day-to-day living as observed by parents and represent an area of concern in this population.

Attentional control has also been identified as an area of impairment in children and youth with SBM. Brewer and colleagues applied Mirsky's neuropsychological model of attention in children with SBM (Mirsky, Anthony, Duncan, Ahearn, & Kellam, 1991). They compared them to children with ADHD only and controls on measures of focused, sustained, and shifting attention. The groups displayed differing patterns of attention functioning. Specifically, children with SBM demonstrated more difficulty on tasks placing greater demands on posterior attention networks (e.g., focus and shifting). In contrast, children with ADHD had more difficulty on tasks that draw on anterior attention systems (e.g., sustained attention) (Brewer, Fletcher, Hiscock, & Davidson, 2001). Loss and colleagues (Loss, Yeates, & Enrile, 1998) also used Mirsky's model as the basis for a study showing that children with SBM and shunted hydrocephalus performed more poorly than siblings and children without shunts on measures of attention. The magnitude of the difference between groups was most significant again for tasks that emphasized posterior attention networks. These results demonstrate that children with SBM exhibit difficulties in attention, but the nature of their difficulties may differ from those seen in other populations because of the greater involvement of posterior brain regions. This would be consistent with the disruption of systems in those regions that can occur due to stretching and thinning of the cortex secondary to hydrocephalus.

Attentional control is essential to executive functioning. Impairment in posterior attention networks has direct implications for executive functioning given that connections between this region and the frontal lobes are central for conducting higher order cognitive tasks (Jurado & Rosselli, 2007). Prefrontal cortex and parietal regions are fundamentally linked and proper functioning relies on their integration.

Psychological Factors in SBM

Emotional regulation and behavioral difficulties are common in chronic illness populations and this is true for SBM as well. Children and adolescents with SBM are at an increased risk for internalizing disorders such as depression, anxiety, and somatic difficulties (Coakley, Holmbeck, & Bryant, 2006). Research has shown that children and young adolescents with SBM are at a greater risk for depressed mood, low self-worth, and suicidal ideation when compared to typically developing controls (Appleton et al., 1997). In addition, this population has evidenced elevated levels of psychiatric symptoms on the Children's Symptom Inventory (CSI): 43% of a sample of children with SBM met the cutoff score for one psychiatric diagnosis and 13% for two or more (Ammerman et al., 1998). Fletcher and colleagues (Fletcher et al., 1995) found that even young children ages 5–7 exhibited clinically significant maladjustment; 26% of the sample was above the cutoff score ($T \geq 64$) for clinically significant impairments on the CBCL. A number of other studies have also found evidence of adjustment problems on the CBCL in children and adolescents with SBM (Wallander, Varni, Babani, Banis, & Wilcox, 1988, 1989). The difficulties are likely related to a number of factors, including cognitive influences such as verbal IQ, restricted socialization, stigma, inadequate coping, and poor self-regulation (Ammerman, Nortz, Ris, & Walz, 2006; Coakley et al., 2006). Although some of these factors have been explored, the variables that contribute to the psychological outcomes in children and adolescents with SBM are not fully understood.

One promising avenue of research that may lend clarity to understanding the psychological outcomes of SBM would be to further explore the role of cognitive and neuropsychological variables. Neuroanatomical organization provides a rationale for why a relationship between these variables might be expected. Specifically, neural substrates physiologically link prefrontal areas that are largely involved in modulating executive skills with structures that regulate emotional functioning in the brain (e.g., amygdala). Functional and structural imaging work suggests that these systems are implicated in mood disorders (Price & Drevets, 2010). In addition to direct neuroanatomical connections, other authors have suggested that events that occur in the early stages of development have the potential to increase children's risk of psychological impairments. Hellemans and colleagues (Hellemans, Sliwowska, Verma, & Weinberg, 2010) proposed an idea built on the model of stress-diathesis involving another neurodevelopmental disorder, fetal alcohol syndrome. In their description, they suggest that neuroadaptive mechanisms mediate the stress response, therefore, increasing an individual's vulnerability to depression and anxiety. Similarly in the current study, executive functioning is conceptualized as the "neuroadaptive mechanism" that mediates the environmental stress response of children with SBM. We propose that when these skills are impaired, the children are at increased risk for poor psychological adjustment.

Other authors within the field of neuropsychology have suggested similar models but with a focus on different outcomes. For example, Fletcher and colleagues (Fletcher, Brookshire, Landry, Bohan, & Davidson, 1996) proposed that cognitive functions (e.g., attention, initiation) mediate the influence of hydrocephalus and its associated physiological abnormalities on social outcomes. Landry and colleagues (Landry, Robinson, Copeland, & Garner, 1993) also suggested a similar role for neuropsychological variables in the relationship between biological factors and social behaviors. Rose and Holmbeck (2007) tested such a mediation model. The authors used a prepublication version of the BRIEF as the measure of executive functioning and several measures of social adjustment. Their analyses indicated that executive functioning (as reported by the teacher on the BRIEF and subscales from the Cognitive Assessment Battery) mediated the associations between SBM status and social adjustment (Rose & Holmbeck, 2007).

The current study builds on and extends these studies in several ways. First, the study uses a mediation analysis, but the focus is on psychological functioning rather than social outcomes, given that this population is at risk for developing internalizing and externalizing symptoms. Additionally, the study uses maternal report on the BRIEF as a measure of executive functioning, specifically the Metacognition Index score. This outcome has been shown to be impaired in previous research with children with SBM, in contrast to the Behavioral Regulation Index (Brown et al., 2008; Mahone et al., 2002). The first hypothesis is that young adolescents with spina bifida will exhibit higher levels of psychological maladjustment in comparison to those without SBM. The second hypothesis is that executive functioning (i.e., Metacognition from the BRIEF) will mediate the relationship between neurodevelopmental status (i.e., with or without SBM) and psychological adjustment outcomes. The third hypothesis is that subscales of the Metacognition Index will also mediate the relationship between neurodevelopmental status and psychological outcomes in this sample. For the analyses, it is hypothesized that higher levels of executive functioning impairment will be associated with poorer psychological adjustment.

METHODS

Participants

Participants in the study were 51 children and adolescents with SBM ages 10–17 years (M = 13.0, SD = 2.4) and 45 nondisabled children ages 10–16 years (M = 11.8, SD = 1.9). Inclusion criteria for the groups were as follows: (a) between the ages of 10–17 years and (b) Verbal or Performance IQ $\geq$ 70 on the Wechsler Intelligence Scale for Children, Third Edition (WISC-III; Wechsler, 1991) or Wechsler Adult Intelligence Scale, Third Edition (WAIS-III; Wechsler, 1997). Children and adolescents with lipomeningocele or sacral meningocele were excluded.

Procedure

Children with SBM were recruited from the Spina Bifida Clinics of both the Division of Developmental Disabilities at Cincinnati Children's Hospital Medical Center in Cincinnati, Ohio and the Nationwide Children's Hospital in Columbus, Ohio. Controls were recruited from local pediatricians' offices and general advertisements and matched to children with SBM on socioeconomic status (SES). Individuals that wanted to participate in the study completed a consent form and children gave assent. The study was approved by the Institutional Review Boards of both hospitals.

Measures

Executive Functioning. Executive functioning was measured by having mothers complete the Behavior Rating Inventory of Executive Function (BRIEF; Gioia et al., 2000). On the BRIEF, raters are asked to indicate whether a specific behavior is "Never," "Sometimes," or "Often" a problem. The BRIEF is comprised of eight clinical scales that form two composite index scales: Behavioral Regulation Index (Inhibit, Shift, and Emotional Control) and the Metacognition Index (Initiate, Working Memory, Plan/Organize, Organization of Materials, and Monitor). The Inhibit scale assesses the ability to stop a behavior appropriately and to resist impulses. It includes items such as "interrupts others." The Shift scale assesses mental-flexibility and the ability to transition and problem solve creatively (e.g., "Tries the same approach to a problem over and over even when it does not work"). Emotional Control is the ability to manage emotions and to execute control over one's feelings and includes items such as "overreacts to small problems." The Initiate scale on the BRIEF refers to the ability to begin a task without external prompting or motivation (e.g., "is not a self-starter"). The Working Memory scale assesses the capacity to hold information in the mind long enough to carry out goals or to complete a task (e.g., "when sent to get something, forgets what he/she is supposed to get"). Plan/Organize measures a child's ability to set a goal and to determine how to obtain it and includes items such as "underestimates time needed to finish tasks." This scale also assesses the ability to bring order to information and understand main ideas and concepts when learning (e.g., "gets caught up in details and misses the big picture"). Organization of Materials refers to the ability to maintain order of personal belongings in the environment and to keep track of one's materials needed for a given task (e.g., "cannot find things in room or school desk"). The Monitor scale assesses the ability to check one's own performance (task-monitoring) and one's awareness or insight about how their behavior affects others (self-monitoring). An item example for this scale is “Does not realize that certain actions bother others.”

The Metacognition Index scale is the specific measure of executive functioning used in the current study. Higher scores on the measure indicate more difficulty with executive functioning skills. The BRIEF has established validity and has been demonstrated to be a psychometrically robust tool (Gioia et al., 2000, 2002).

Psychological Adjustment. The Behavior Assessment System for Children, second edition (BASC-2; Reynolds & Kamphaus, 2004) is a multidimensional assessment of children’s behavior and emotional problems. Raters are asked to report if specific behaviors occur “Never,” “Sometimes,” “Often,” or “Almost Always.” The BASC-2 Parent Rating Scale (BASC-2 PRS) was used in the current study and ratings were obtained from mothers. The following composite scales from the BASC-2 PRS were used: Externalizing Problems (Hyperactivity, Aggression, and Conduct Problems), Internalizing Problems (Anxiety, Depression, and Somatization), and Adaptive Skills (Adaptability, Social Skills, Leadership, Activities of Daily Living, and Functional Communication).

The Child Depression Inventory (CDI; Kovacs, 1992) is a measure that assesses the cognitive, affective, and behavioral symptoms of depression in children and adolescents ages 7 to 17 years old. It contains 27 items each consisting of three statements. For each item, the individual is asked to select the statement that best describes his or her feelings for the past 2 weeks. The youth were administered the child version and mothers were given the parent-report version of the measure. Total T-scores were used in analyses.

The Children’s Loneliness Scale is composed of 16 items assessing the child’s feelings of loneliness, social inadequacy, and subjective estimations of peer status. The response format utilizes a 5-point Likert scale indicating the degree of truthfulness in describing the respondent. It displays excellent internal consistency and moderate test-retest reliability (Asher & Wheeler, 1985).

Data Analysis Plan

The first step of data analysis was to run descriptive statistics followed by correlations of the BRIEF and BASC-2. Next, multivariate analyses of covariance (MANCOVA) were run to determine whether there were differences between the SBM and control groups on psychological adjustment outcomes. The next planned step was to conduct a mediation model analysis to determine whether metacognition explained the relationship between neurodevelopmental status and psychological adjustment outcomes. Finally, another mediation analysis was conducted to determine whether subscales of the Metacognition Index would also explain the relationship between neurodevelopmental status and psychological adjustment.

RESULTS

Analyses were conducted using PASW Statistics 17.0 analysis software. Descriptive statistics for the sample are presented in Table 1. The groups demonstrated a significant age difference, with the SBM group being older than the control group. The SBM group also had significantly lower Full Scale IQ (FSIQ), Verbal IQ (VIQ), and Performance IQ (PIQ) scores. Child age and mother age were covaried in the MANCOVA and the mediation model. IQ was not considered as a covariate because IQ is an outcome of neurodevelopmental conditions and is intimately connected to the disorder itself (Dennis et al., 2009).

Table 1 Descriptive Statistics in Spina Bifida (SBM) and Nondisability Control Groups.

	SBM ($n = 51$)	*Controls* ($n = 45$)
Child Age	13.0 ($SD = 2.4$)*	11.8 ($SD = 1.9$)*
Parent Age	41.3 ($SD = 6.1$)	39.3 ($SD = 5.0$)
Gender		
Male	22 (43.1%)	21 (46.7%)
Female	29 (56.9%)	24 (53.3%)
VIQ	89.4 ($SD = 20.1$)*	110.5 ($SD = 13.0$)*
PIQ	78.1 ($SD = 16.1$)*	107.0 ($SD = 11.9$)*
FSIQ	83.1 ($SD = 17.9$)*	110.0 ($SD = 11.8$)*

Note. VIQ – Verbal IQ, PIQ – Performance IQ, FSIQ – Full Scale IQ.
*$p < .001$ for the group difference.

Table 2 Correlations between Variables for Spina Bifida (SBM) and Nondisability Control Groups.

Variable	Metacognition	Externalizing	Internalizing	Adaptive Skills	Loneliness	CDI-Child
Metacognition						
Externalizing	.42**/.31					
Internalizing	.48**/.53**	.46**/.42**				
Adaptive Skills	−.19/−.51**	−.50**/−.46**	−.33**/−.38**			
Loneliness	.36*/.14	.15/.01	.22/−.01	−.17/−.09		
CDI-Child	.46*/.39*	.21/.19	.27/.18	−.26/−.37	.68**/.64**	
CDI-Mother	.64**/.56**	.47**/.48**	.64**/.68**	−.18/−.45**	.30/.12	.43**/.24

Note. Results of the nondisability controls are presented as the first number in the correlation pairs.
*$p < .05$. **$p < .01$.

Correlation Analyses

Within-group correlations between the BRIEF Metacognitive Index and psychological variables are presented in Table 2. The Metacognitive Index had a significant positive correlation with Internalizing Symptoms and with depressive symptoms as reported by the CDI-Child and CDI-Mother in the SBM group. It was negatively correlated with Adaptive Skills, such that poorer metacognitive skills were correlated with worse adaptive functioning in children and adolescents with spina bifida. Externalizing Problems and the Loneliness Scale were not significantly correlated with the Metacognitive Index in the SBM group. The results were largely comparable in the control group, with the exception of Externalizing Problems and Loneliness, both of which had a significant positive correlation with the Metacognitive Index. Adaptive skills were not significantly correlated with the Metacognitive Index in the control group.

MANCOVA

A multivariate analysis of covariance contrasting SBM and control participants and controlling for child's age and mother's age was performed on the six psychological adjustment outcomes: Externalizing Problems, Internalizing Problems, and Adaptive Skills from the BASC-2, the Loneliness Scale, and the CDI-Child and CDI-Mother reports. The groups differed significantly, $F(6, 72) = 7.37$, $p < .001$. Follow-up univariate comparisons

Table 3 Univariate Comparisons between Spina Bifida ($n = 51$) and Nondisability Controls ($n = 45$).

Variable	SBM *M* (*SD*)	Controls *M* (*SD*)	*F*	*d*
Externalizing Problems	50.1 (12.7)	45.3 (9.9)	6.1	0.55
Internalizing Problems	57.4 (11.7)	48.7 (9.7)	21.7*	1.03
Adaptive Skills	41.6 (8.1)	52.3 (8.0)	38.3*	1.37
Loneliness Scale	29.6 (9.6)	27.6 (7.7)	0.7	0.19
CDI-Child	46.0 (7.5)	44.0 (7.3)	1.4	0.27
CDI-Mother	49.2 (8.9)	42.9 (6.9)	17.8*	0.93

Note. Externalizing Problems, Internalizing Problems, and Adaptive Skills from the BASC-2; CDI-Child Depression Inventory child and mother reports.
$^{*}p < .008$ (Bonferroni Correction).

revealed significant group differences in Internalizing Problems, Adaptive Skills, and CDI-Mother after Bonferonni correction ($p < .008$). In addition, Internalizing Problems was secondarily analyzed to determine if group differences were reflected in each of the subscales of this scale (Depression, Anxiety, Somatization). Results indicated that each of these scales yielded significant group differences. Group was not significantly associated with Externalizing Problems, Loneliness, or the CDI-Child. Table 3 shows the results of the univariate comparisons between the SBM and control groups, including the standardized mean difference (*d*), for each contrast.

Mediation Analyses

A mediation analysis was conducted to determine whether the Metacognitive Index mediated the relationship between Group (SBM vs. controls) and psychological adjustment. Four conditions should be met for complete mediation to be supported: The predictor must be significantly associated with the outcome, the predictor must be significantly associated with the mediator, the mediator must be significantly associated with the outcome after controlling for the predictor, and the predictor and outcome must no longer be significantly associated when the effects of the mediator are controlled (Rose, Holmbeck, Coakley, & Franks, 2004). This latter step is essential for complete mediation; when the last step does not hold, evidence is available for partial mediation if the first three steps hold. These conditions were evaluated in the current study. Sobel tests were also conducted to evaluate the Metacognitive Index as a mediator for outcome variables that were significantly associated with group. First, the MANCOVA reported in the previous section revealed that group was a significant predictor of psychological outcomes, and univariate analyses of covariance (ANCOVAs) demonstrated significant group differences on Adaptive Skills, Internalizing Problems, and CDI-Mother. Second, a univariate ANCOVA was conducted with Group as the independent variable, child's and mother's ages as covariates, and the Metacognitive Index as the outcome variable. The groups differed significantly ($M_{\mathrm{S\,M}} = 63.9$, $M_{\mathrm{Controls}} = 49.7$), $F(1, 82) = 45.9$, $p < .001$, $d = 1.53$. Therefore, the first two conditions of mediation were met for the Metacognitive Index as a mediator of group differences for Internalizing Problems, Adaptive Skills, and CDI-Mother. To evaluate the last two conditions for mediation, multiple regression analyses were performed with Group and the Metacognitive Index entered together as predictors, along with child's and mother's ages as covariates, with each psychological adjustment

Table 4 Multiple Regressions with Metacognition Predicting Psychological Adjustment Outcomes after Controlling for Group, Child Age, and Mother Age.

Variable	β	Adjusted R^2	Effect Size
Internalizing Problems			
Metacognition	0.54*	.40*	0.70
Group	−3.33		
Child Age	−0.20		
Mother Age	−0.48		
Adaptive Skills			
Metacognition	−0.28*	.37*	0.34
Group	7.56*		
Child Age	−0.19		
Mother Age	0.25		
CDI-Mother			
Metacognition	0.47*	.42*	0.86
Group	−1.09		
Child Age	−0.01		
Mother Age	−0.15		

Note. Metacognition from the BRIEF; Externalizing Problems, Internalizing Problems, Adaptive Skills from the BASC-2; CDI-Child Depression Inventory mother report.
*$p < .008$ (Bonferroni Correction).

variable (i.e., Internalizing Problems, Adaptive Skills, and CDI-Mother) treated as an outcome in three separate models. The results are presented in Table 4. The Metacognitive Index was a significant predictor of Internalizing Problems, Adaptive Skills, and CDI-Mother after controlling for group, child's age, and mother's age. Sobel tests based on first and second derivatives for the standard errors were conducted (MacKinnon, 2008) and yielded consistent results with respect to the statistical significance of the mediated effect for each of Internalizing Problems ($p < .001$), Adaptive Skills ($p = .007$), and CDI-Mother ($p < .001$). The group difference was no longer statistically significant in the models with Internalizing Problems and CDI-Mother as the outcomes, providing evidence for complete mediation. However, group differences remained significant for Adaptive Skills, suggesting only partial mediation in this instance. Effect sizes (Table 4) were calculated to determine the strength of the mediation relationship. The particular effect size of interest was the proportion of the total effect explained by the indirect effect and, thus, by the mediator. Larger values correspond to mediators that explain more of the variation in the total effect (MacKinnon, 2008).

To elucidate the relationship of the Metacognitive Index to Internalizing Problems and CDI-Mother, additional analyses were conducted to determine whether mediation occurred with the subscales that constitute the Metacognitive Index. The first step of the mediation analyses was the same as described above (i.e., group differences for each psychological outcome). In the second step, group differences were significant for all of the Metacognitive Index subscales after controlling for child's age and mother's age in separate ANCOVAs where the SBM children were coded as "0" and controls were coded as "1": Initiate ($\beta = -15.1$), $F(1, 82) = 46.5$, $p < .001$, Working Memory ($\beta = -15.7$), $F(1, 82) = 43.2$, $p < .001$, Plan/Organize ($\beta = -14.5$), $F(1, 82) = 39.9$, $p < .001$, Organization of Materials ($\beta = -6.7$), $F(1, 82) = 10.2$, $p < .01$, and Monitor ($\beta = -6.9$), $F(1, 82) = 8.5$, $p < .01$. Again, the first two conditions of mediation were satisfied. In the third step, multiple regression was used, with group and each individual Metacognitive Index subscale entered together as predictors in separate models for the two measures of psychological adjustment (i.e., Internalizing Problems and CDI-Mother). This yielded a total of 10 multiple regressions. The results are presented in Table 5. The Initiate, Working Memory, and Plan/Organize subscales were significant predictors of Internalizing Problems and CDI-Mother when group, child's age, and mother's age were controlled. Group differences were no longer statistically significant in these models, reflecting complete mediation. The Organization of Materials and Monitor subscales also significantly predicted Internalizing Problems and CDI-Mother; however, group remained statistically significant in the models. Consequently, the results provide evidence for only partial mediation for those subscales. Effect sizes are again shown in Table 5 that reflect the extent to which the mediators explain the proportion of the total effects.

DISCUSSION

The current study sought to examine the relationship between neuropsychological functioning and psychological adjustment in children and adolescents with spina bifida. Specifically, we sought to understand how executive functioning was related to these outcomes. The results indicate that executive functioning, in particular metacognitive skills, account for the relationship between group status (i.e., having spina bifida or not) and how these youth function psychologically. Further, particular aspects of metacognitive skills appear to have greater implications for psychological functioning than others.

Group differences were significant on internalizing symptoms, adaptive functioning, and depressive symptoms as reported by mothers. The SBM group had greater difficulties in these areas, consistent with the previous literature. A number of reasons may account for this finding. Youth with SBM face a variety of challenges in multiple spheres of life that can impact their emotional functioning (e.g., anxiety, depression). Measurement issues may also help explain the findings. One of the three subscales that compose the Internalizing composite of the BASC-2 is a Somatization scale. It assesses pain and physical symptoms (e.g., gets sick, complains about health, and complains of pain), which are salient issues for individuals with SBM. The validity of using this scale in populations with neurodevelopmental disorders is called into question. Perhaps the mothers' reports of physical symptoms in children with SBM are contributing to artificial inflation of children's depression scores on this measure. Similarly, the parent version of the CDI includes a Functional Problems subscale, which assesses not only interpersonal issues but also school-related performance (e.g., "has to push himself or herself to do schoolwork," "is showing worse school performance than before"). Youth with SBM often struggle academically. Consequently, difficulties in other domains of functioning may contribute to mothers' reports of internalizing symptoms, based on the specific measures that were utilized in this study.

Youth with spina bifida did not report more depressive symptoms or loneliness than the nondisabled controls. It is possible that they were underreporting their internalizing symptoms at least as compared to maternal reports. This particular finding is curious, given that previous literature suggests that children with spina bifida are at risk for difficulty in these areas. The findings suggest the possibility that children and adolescents with spina bifida have poor insight about their own emotional functioning. In contrast, previous research suggests that children and adolescents (both healthy and those with chronic illness) report *more* internalizing symptoms than are observed by their parents (Martin,

Table 5 Multiple Regressions with Metacognition Subscales Predicting Psychological Adjustment Outcomes after Controlling for Group, Child Age, and Mother Age.

Variable	β	Adjusted R^2	Effect Size
Internalizing Problems			
Initiate	0.45*	.38*	0.65
Group	−3.73		
Child Age	−0.51		
Mother Age	−0.48		
Working Memory	0.35*	.29*	0.58
Group	−5.04		
Child Age	−0.13		
Mother Age	−0.58		
Plan/Organize	0.41*	.37*	0.57
Group	−4.54		
Child Age	−0.16		
Mother Age	−0.63*		
Organization of Materials	0.30	.29*	0.19
Group	−8.46*		
Child Age	−0.08		
Mother Age	−0.67*		
Monitor	0.35*	.34*	0.23
Group	−7.98*		
Child Age	−0.23		
Mother Age	−0.65*		
CDI-Mother			
Initiate	0.38*	.34*	0.74
Group	−1.98		
Child Age	−0.41		
Mother Age	−0.13		
Working Memory	0.37*	.35*	0.74
Group	−2.04		
Child Age	0.13		
Mother Age	−0.19		
Plan/Organize	0.37*	.35*	0.70
Group	−2.32		
Child Age	0.09		
Mother Age	−0.26		
Organization of Materials	0.21	.21*	0.18
Group	−6.34*		
Child Age	0.13		
Mother Age	−0.31		
Monitor	0.34*	.32*	0.30
Group	−5.37*		
Child Age	0.03		
Mother Age	−0.27		

Note. Metacognition from the BRIEF; Externalizing Problems, Internalizing Problems, Adaptive Skills from the BASC-2; CDI-Child Depression Inventory mother report.
*$p < .008$ (Bonferroni Correction).

Ford, Dyer-Friedman, Tang, & Huffman, 2004; Rockhill et al., 2007; Sourander, Helstelä, & Helenius, 1999). The correlation between parent and child CDI reports in this study was not significant in the spina bifida group but was significant in the control group, suggesting greater agreement in the latter group. An alternative interpretation is that parents of children with spina bifida overreported symptoms of depression and focused on physical symptoms that are more related to the disorder rather than to children's emotional state.

Metacognition, as measured in this study, fully accounted for group differences in internalizing and depressive symptoms in mediation analyses. These results suggest that the psychological symptoms these youth experience (as reported by mothers) are perhaps largely a function of their metacognitive skills. Metacognition only partially mediated the relation between group status and adaptive functioning. Therefore, the adaptive deficits shown by children with spina bifida are not accounted for entirely by their deficits in executive functioning.

The findings also suggest that there are particular components of metacognitive skills that are related to psychological adjustment. Specifically, the extent to which a child or adolescent is able to be self-motivated, to mentally manipulate information long enough to carry out a task and to plan and be organized appears to explain their psychological symptoms as reported by mothers. This suggests that if a child has particular difficulty with these skills, their psychological functioning could be compromised. In contrast, organization of personal materials and self-monitoring did not fully account for psychological outcomes; instead a direct effect of group was still evident in these models. It could be that the former skills are more directly evident to others and have a significant impact on how youth function and manage tasks. Children may receive more negative feedback about these particular skills, which in turn perhaps affects their mood. When children and adolescents have difficulty getting started with tasks and procrastinate, have difficulty multitasking and find it hard to think ahead and prepare appropriately, these issues are clearly observed by parents and teachers and interfere with their ability to meet the demands of the environment. This may provide the path by which the internalizing and depressive symptoms begin to manifest themselves.

The current study partially replicates the findings from previous work by Rose and Holmbeck (2007), in that executive functioning mediated the relationship between group status and specific outcomes in children and adolescents with spina bifida. The field as a whole suffers from a lack of replication, and the current findings illustrate how models can provide consistency across conceptual domains. However, the current study differs in that psychological adjustment was the main focus of this investigation. Therefore, the findings also add to the literature by elucidating the relationship between these variables.

The current study has several limitations. First, it is cross-sectional in nature that precludes drawing conclusions about causality. A longitudinal study design would more readily facilitate the ability to make causal links between neurodevelopmental status and psychological outcomes. As it stands, it is unclear whether cognitive factors mediate the relationship between group status and psychological outcomes or whether psychological outcomes mediate the relationship between group status and cognitive factors. Future studies should analyze similar models from a longitudinal perspective, especially given that executive functioning skills are actively developing over the course of adolescence. Another limitation is that parent report was used both for measures of executive functioning and psychological adjustment, which introduces shared rater and method variance. Therefore, this could provide an explanation for why metacognition was related to maternal report on the CDI but not child report. In other words, it is possible that the relationship between metacognition and depression reported by mothers was largely due to the fact that they completed both forms. This same relationship was not found when children reported on their own depression. This study is also limited by the fact that performance measures of executive functioning (e.g., Delis-Kaplan Executive Function System) were not incorporated that would have served to abate the issues regarding common method variance and provided insight to deficits not assessed on the BRIEF. In addition, the BRIEF has been found to correlate with other measures of parent report of psychological functioning and may not be a pure measure of executive function; however, it is a well-standardized measure with established validity (Gioia et al., 2000, 2002; Mahone et al., 2002). In the current study, indicators of illness severity were not incorporated into the analysis. Therefore, it is possible that the model may not hold for youth who, for example, have a higher lesion level or more significant adaptive impairment. Finally, the participants in this study were preadolescents and adolescents. Future studies might attempt to understand the relationships between executive functioning and psychological outcomes in a younger cohort of children.

The results of this current study have implications for psychological treatment not only in youth with spina bifida, but in typically developing individuals as well. The findings suggest that implementing interventions that promote functional executive skills could affect children's emotional functioning and mood. The literature on evidence-based interventions for executive functioning in children and adolescents with neurodevelopmental disorders is limited, and this would be an important area of investigation to pursue in future studies.

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