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Executive functions in adolescents with spina bifida: Relations with autonomy development and parental intrusiveness

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The current study was part of a larger longitudinal investigation and examined the relation of parent-report and performance measures of executive functioning (EF) with measures of behavioral and emotional autonomy and parental intrusiveness in adolescents with and without spina bifida (SB; $n = 65$ in a comparison sample and 61 in an SB sample; M age $= 14.55$, $SD = 0.63$). For both groups, higher levels of *parent-reported* EF problems predicted higher levels of observed child dependency and lower levels of teacher-reported intrinsic motivation. Higher scores on *performance* EF measures predicted lower levels of observed child dependency and observed maternal intrusiveness for both groups. In adolescents with SB only, higher performance EF scores predicted higher intrinsic motivation and emotional autonomy from both mother and father and predicted lower levels of observed paternal intrusiveness. While causal conclusions cannot be drawn, EFs appear to be closely related to autonomy development and parental intrusiveness, particularly for adolescents with SB. These results suggest that the inclusion of EF training in interventions targeting adolescents with SB may be beneficial for autonomy development.

Keywords: Spina bifida; Executive function; Autonomy; Parental intrusiveness; Adolescence.

Spina bifida (SB) is a severe birth defect resulting from a failed closure of the neural tube during fetal development and is associated with several abnormalities in the brain, including hydrocephalus and the Arnold-Chiari malformation (Chiari II malformation). While individuals with SB generally have average to low-average intelligence, these brain abnormalities lead to a specific pattern of cognitive performance (see Dennis, Landry, Barnes, & Fletcher, 2006, for a review), including relative strengths in grammar and

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vocabulary and weaknesses in semantic and pragmatic knowledge as well as attentional shifting and focusing (Brewer, Fletcher, Hiscock, & Davidson, 2001; Burmeister et al., 2005; Swartwout et al., 2008).

Recent research has found SB to be associated with deficits in executive functioning (EF) as well (e.g., Burmeister et al., 2005; Rose & Holmbeck, 2007). EFs are thought to include planning, decision making, judgment, and self-regulation (Han, Delis, & Holdnack, 2008). Research examining EFs in individuals with SB has found impairment on behavioral measures of switching ability (Iddon, Morgan, Loveday, Sahakian, & Pickard, 2004; Rose & Holmbeck, 2008; Snow, 1999), working memory (Burmeister et al., 2005; Iddon et al., 2004), planning, organization, goal-directed behavior, and problem solving (Burmeister et al., 2005; Fletcher et al., 1996; Iddon et al., 2004; Snow, 1999). Some have suggested that such EF deficits are not due to impairments in frontally mediated skills but are instead due to deficits in processing speed and attention, skills that are known to be impacted in SB (Burmeister et al., 2005; Fletcher et al., 1996). Indeed, the posterior attentional networks necessary for executive attention are part of a distributed neural network underlying EF and are also thought to be impacted in SB (Denckla, 1996; Fletcher et al., 1996).

In addition to executive dysfunction, youth with SB have been found to have difficulties associated with the transition to adolescence, including in the area of autonomy development (e.g., Friedman, Holmbeck, DeLucia, Jandasek, & Zebracki, 2009). Autonomy is an interactive process between adolescents and their families in which the adolescent gains independence while continuing connections with the family (Friedman et al., 2009). Two types of autonomy were assessed for the current study: behavioral autonomy, or independent decision-making behavior, and emotional autonomy, which requires adolescents to relinquish their childish dependencies on their parents (Harter, 1980; Hill & Holmbeck, 1986; Lamborn & Steinberg, 1993; Silverberg & Gondoli, 1996). The development of autonomy is likely more challenging for an adolescent with SB due in part to their physical disabilities, which require them to rely on family members for tasks that typically developing adolescents can perform relatively independently (Blum, Resnick, Nelson, & St. Germain, 1991; Friedman et al., 2009). The special medical needs of adolescents with SB also introduce additional areas of responsibility (Friedman et al., 2009). In support, a recent longitudinal study examining trajectories of autonomy development in samples of typically developing adolescents and adolescents with SB found that adolescents with SB consistently exhibited more dependent behaviors than typically developing peers at ages 9 through 15 (Friedman et al., 2009). Consistent with this finding, the sample with SB had a slower growth rate for intrinsic motivation, producing a larger difference from typically developing peers by age 15. On the other hand, adolescents with SB had higher growth rates of behavioral autonomy, thus eliminating any sample differences in this factor by age 15 (Friedman et al., 2009).

Certain parenting behaviors may also alter the transition to adolescence in children with SB. Having a child with a chronic illness has been shown to promote the development of intrusive parenting in some families (Holmbeck et al., 2002). Such behaviors include parental overprotectiveness, which refers to a level of parental protectiveness and intrusiveness that is excessive considering the child's developmental level (Thomasgard, Metz, Edelbrock, & Shonkoff, 1995). Parental intrusiveness tends to occur more often in samples with chronically-ill children in part due to the child's medical needs (Holmbeck et al., 2002). In situations where a child's health depends on the management of a complex medical regimen, it may be adaptive for parents to increase their protectiveness to ensure that all the child's medical needs are met and to modulate their own levels of anxiety (Holmbeck

et al., 2002). Yet, such an environment may also promote the development of less adaptive parenting that may include excessive protectiveness and control (Anderson & Coyne, 1991, 1993). Such a pattern becomes especially problematic during the transition to adolescence, when autonomy development is particularly salient (Holmbeck et al., 2002).

Levels of autonomy development and parental intrusiveness in adolescents with SB may be partially explained by the adolescent's level of EF. The development of EF networks in the brain, which is not completed until early adulthood, mirrors the development of autonomy in adolescence (Tarazi, Zabel, & Mahone, 2008). That is, the higher order abilities subsumed under EF are the skills necessary for a child to successfully achieve autonomy. Indeed, policy makers have already taken this lack of maturity in the EF networks into account when noting adolescents' relative immaturity in the area of autonomous decision making (Johnson, Blum, & Giedd, 2009). Without the abilities to plan, to make decisions, to use judgment, and to think flexibly, adolescents will be less able to assume responsibilities normally attended to by their parents and hence be less able to autonomously navigate the world. For adolescents with SB in particular, whose unique medical needs require a keen ability to plan, to stay organized, and to make healthy decisions, the relationship between EF and the ability to independently care for oneself may be even more robust (e.g., Ries, Zabel, & Mahone, 2003; Sawin, Brei, & Adams, 2003). However, only one study could be located that examined the link between EF and self-care skills in children with SB, finding that parent-report levels of working memory and initiation were negatively associated with self-care skills (Ries et al., 2003). In addition, in a review of the literature, components of EF were associated with autonomy-related outcomes in individuals with SB, such as employment rates, independent living, and management of medical needs (Sawin et al., 2003). Yet, studies with other populations have also found support for the relationship between EF and autonomy. For instance, much research examining decision-making capacity, a specific manifestation of autonomy, has found that higher EF predicts more independence in medical decision making (e.g., Basso et al., 2010; Dreer, DeVivo, Novack, Krzywanski, & Marson, 2008).

In addition, an adolescent's level of executive dysfunction may increase the likelihood of parental intrusiveness through parents' observation of their child's EF deficits. If parents observe their child having difficulties making sound decisions and planning and organizing effectively, they may be less likely to transfer responsibilities to them, particularly for important medical tasks. Somewhat surprisingly, research examining the relationship between parenting and a child's EF has only considered how parenting impacts a child's EF, not how a child's EF determines a parent's tendency to be overly intrusive towards their child. Nevertheless, such research has found that parental autonomy promotion, parental sensitivity, and the utilization of scaffolding enhance a child's EF (e.g., Bernier, Carlson, & Whipple, 2010; Dilworth-Bart, Poehlmann, Hilgendorf, Miller, & Lambert, 2010). Yet, for adolescents with SB who have EF deficits due to characteristics of SB itself, it will be important to examine EF not only as an outcome of parenting but also as a predictor of parenting behaviors. As previously discussed, having a child with a chronic illness is known to produce higher levels of intrusive parenting, and, while this may be due to the high amount of health risks associated with the chronic illness, this increase in intrusive parenting may also be in response to the EF deficits in these children. Such a possibility warrants further exploration than it has received in the literature.

Thus, the purpose of this study was to examine behavioral and parent-report measures of EF in relation to observed and reported levels of child autonomy and parental intrusiveness. The present study was part of a larger longitudinal investigation exploring

the transition to adolescence in children with and without SB. The sample was composed of adolescents with SB and a demographically matched comparison sample of typically developing adolescents. It was predicted that adolescents with SB would have lower levels of executive functioning overall. In addition, it was expected that EF skills would be positively related to behavioral and emotional autonomy development in both samples, but particularly for adolescents with SB. Similarly, it was also predicted that EF would be negatively related to parental intrusiveness, particularly for adolescents with SB.

METHOD

Participants

The participants were drawn from a longitudinal study that focused on family relationships and psychological adjustment in families of children with SB and a comparison sample of families of typically developing children (for a more detailed review of participant selection and methods, see Holmbeck et al., 1997, 2002, 2003). Children with SB between the ages of 8 and 9 were recruited for participation through local children's hospitals and a statewide SB association. Participants in the comparison sample were recruited from the schools where the children with SB were enrolled. Participating families were followed every 2 years through four waves of data collection. During the first data collection period (Time 1), 68 children with SB and 68 typically developing children and their families participated in the study. The samples were matched across 10 major demographic variables, including child age, child ethnicity, and socioeconomic status (see Holmbeck et al., 2003, for a detailed description of the sample matching process). The current study examined parent and child data collected during the fourth wave (Time 4) of data collection, when the youth were 14 or 15 years old. By Time 4, there were 61 participants in the SB sample and 65 in the comparison sample (93% retention, total $N = 126$). Those who dropped out of the study did not differ significantly from remaining participants in age, gender ratio, intellectual ability (Peabody Picture Vocabulary Test–Revised; PPVT-R score), or (in the SB sample) SB lesion level. Across both samples, 69 (55%) of the participants in the Time 4 data collection were male while 57 (45%) were female. Mean age for the adolescent participants at Time 4 was 14.55 ($SD = .63$). Eighty-nine percent ($n = 112$) of the sample was Caucasian, 3.2% ($n = 4$) was African American, 3.9% ($n = 5$) was Asian, 1.6% ($n = 3$) was Latino, and 2.4% ($n = 3$) identified as "other." Ethnicity data was missing from 17% ($n = 26$) of the sample. Participants' socioeconomic status (SES) was measured using the Hollingshead (1975) Four Factor Index of SES, which is based on a composite of maternal and paternal education and occupational status and ranges from 8 to 66. The mean Hollingshead SES score for the sample was 45.00 ($SD = 10.94$), indicating that the average SES of the sample was in the moderate range. Refer to Holmbeck et al. (2003) for complete demographic information for this sample.

Among the participants with SB at Time 4, 29% had sacral level spinal lesions, 57% had lumbosacral or lumbar lesions, and 14% had thoracic lesions. The majority (71%) was shunted for hydrocephalus. The average number of shunt surgeries prior to Time 4 (including initial replacement and subsequent revisions) was 5.16 ($SD = 8.61$; although there were a few outliers with an extremely high number of shunt-related surgeries, range = 0–41 surgeries). Six (10%) of the participants with SB were described by their parents as having seizures or taking medications for seizures. Per parent report, adolescents in the sample with SB 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), $\chi(1)^2 = 810, p = .004$.

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 one hour of videotaped family interaction tasks. Additionally, the children were asked to participate in brief neurocognitive testing 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 89% at Time 4. Approval was obtained from all relevant institutional review boards prior to initiation of the study, and all participants were compensated for their time at each round of data collection.

The following two tasks from the videotaped family interaction session were coded for pertinent autonomy and parenting variables: an unfamiliar board game task and a conflict task. For the board game task, families were asked to establish rules for and to play a novel educational game. The conflict task was based on a procedure used by Smetana, Yau, Restrepo, and Braeges (1991). During the questionnaire portion of the home visit, parents and the child completed a short form of the Issues Checklist (Robin & Foster, 1989), a frequently used measure of parent-child conflict that inquires about parent-child discussions that have taken place over the past 2 weeks across 15 issues. Prior to the beginning of the interaction tasks, research assistants tabulated weighted conflict scores (i.e., Frequency $\times$ Intensity) for each issue for each family member on the basis of questionnaire responses. The five issues that received the highest total weighted conflict score across family members were presented to the family for discussion during the conflict task. Family members were asked to select and discuss three of the five issues for 10 minutes total.

Parent and Child 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.

Behavioral autonomy. To assess behavioral autonomy, we used the Decision-Making Questionnaire (DMQ; Dornbusch et al., 1985, Steinberg, 1987) for which respondents (parents and children) were asked to rate their perception of who makes decisions in the family (Steinberg, 1987). Fifteen nonmedical issues were included in this measure, such as when the child has to do homework and what the child is allowed to watch on television. Items were rated on the following scale: (a) parents decide; (b) parents and child discuss the issue, but parents have the final say; (c) parents and child discuss the issue, but the child has the final say; and (d) the child decides. The average scale alpha across reporters was .81 for the SB sample and .74 for the comparison sample. Because mother and father reports were found to be moderately correlated ($r = .45$ to .47), the mean across reporters was used as the index of behavioral autonomy and was scored in the direction of greater behavioral autonomy for the child.

Emotional autonomy. Children completed a composite of three subscales (Parental Deidealization, Nondependency on Parents, and Individuation) from the 20-item Emotional Autonomy Scale (EAS; Steinberg & Silverberg, 1986). Children were asked to rate how much they agreed on a 4-point Likert scale with 14 statements (e.g., "Even when my mother and I disagree, my mother is always right," "I try to have the same opinions as my mother," "I go to my mother for help before trying to solve a problem myself"; all reverse scored). Each child participant completed this questionnaire twice, once referencing emotional autonomy from the mother ($\alpha = .80$) and once referencing emotional autonomy from the father ($\alpha = .80$).

Parental intrusiveness. To assess the intrusiveness construct, we used items from two questionnaires. These items were selected because they indexed excessive (rather than normative) levels of intrusiveness.

The Child Report of Parental Behavior Inventory (CRPBI; Schaefer, 1965; Schludermann & Schludermann, 1970; Schwarz, Barton-Henry, & Pruzinsky, 1985) is a 108-item scale that assesses maternal and paternal child-rearing behaviors. The scale includes 18 subscales that tap three second-order factors: acceptance-rejection, firm control-lax control, and psychological control-psychological autonomy. Mothers, fathers, and children completed a portion of the 18 subscales by rating parents on a 3-point Likert scale for the following subscales: Acceptance (eight items) and Rejection (eight items, reverse scored) from the acceptance-rejection factor; Control (five items), Enforcement (five items), and Lax Discipline (five items, reverse scored) from the firm control-lax control factor; and Intrusiveness (five items) and Hostile Control (eight items) from the psychological control-psychological autonomy scale. In the current study, only child and parent report on the five-item Intrusiveness subscale were used.

The 25-item Parental Bonding Instrument (PBI) assesses the quality of bonding between parent and child across two dimensions: parental care (12 items) and parental overprotection (OP; 13 items; Parker, Tupling, & Brown, 1979). Mothers, fathers, and children responded to five of the items from the OP scale on a 3-point Likert scale. Sample items from the child report version include "My (mother/father) likes to baby me," and "My (mother/father) lets me decide for myself" (reverse scored). These five items were chosen because other items included words and concepts too complex for 8- and 9-year-old children or because of redundancy with items from the Intrusiveness subscale of the CRPBI.

To construct the questionnaire measures of intrusiveness, we combined mother and child reports of the mother's parenting on the five CRPBI items and the five PBI items to assess maternal intrusiveness (yielding a 20-item scale across the two respondents and two measures). An analogous 20-item scale was constructed for paternal intrusiveness. Alphas for maternal and paternal intrusiveness across the SB and comparison samples ranged from .71 to .80.

Questionnaire measure of executive functioning. The Behavior Rating Inventory of Executive Function (BRIEF; Gioia, Isquith, Guy, & Kenworthy, 2000) is a multidimensional measure of parent-reported executive functioning that correlates well with other measures of attention and behavioral control. With permission from the authors, questions from a prepublication 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, across samples and raters, are provided in parentheses): Initiate ($\alpha = .73$ to .84), Sustain ($\alpha = .74$ to .88), Organize ($\alpha = .87$ to .91), Plan ($\alpha = .81$ to .90), and Working Memory ($\alpha = .84$ to .88). Alphas for the full measure (a composite of these five subscales) ranged from .96 to .97 across raters. The five selected scales consisted of 42 items (e.g., "Underestimates time needed to finish tasks") that were rated by parents as "never," "sometimes," or "often" a problem for the child. Thus, higher scores on the BRIEF indicate more parent-reported problems with EF. These five subscales were combined into a total BRIEF score, which was used in the current study. Because the mother- and father-reported total 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.

Teacher Questionnaires

Intrinsic motivation. As a measure of autonomy, intrinsic motivation was assessed using a teacher-report of Harter's (1980) 10-item 4-point scale of Intrinsic Versus Extrinsic Orientation in the Classroom – Revised. This measure assesses child motivation across five dimensions: Challenge, Curiosity, Independent Mastery, Independent Judgment, and Internal Criteria for Success/Failure. The total intrinsic motivation score was used (i.e., the average item score across all items). The alpha for the intrinsic motivation scale across SB and comparison samples was .82.

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. Studies have found the PPVT-R to correlate moderately to highly with the Wechsler Intelligence Scale for Children–III (WISC-III) Full Scale IQ and Verbal IQ scores (e.g., Slate, 1995).

Examiner-administered measures of attention and executive functioning. Participants were administered portions of a multidimensional neurocognitive measure, the Cognitive Assessment System (CAS; Naglieri & Das, 1997). Two CAS subscales, Planned Connections and Planned Codes, were used in the current analyses. The Planned Connections subtest includes five trials, three of which assess planning and sequencing ability (PCo-sequencing) and two of which assess planning and sequencing abilities, mental flexibility, and switching ability (PCo-switching). Trials assessing sequencing ability require the youth to sequentially connect letters or numbers in a random array, while trials assessing switching require the youth to switch between sequentially connecting letters and numbers in an array. Importantly, tasks assessing switching ability typically also require one to engage more basic EFs to perform well. For example, in PCo-Switching, a child also must engage sequencing abilities in addition to mental flexibility in order to execute the task. Because such switching tasks are composed of multiple EF skills, they may be a more challenging measure of executive functioning

and thus more likely to differentiate EF-impaired samples than measures requiring fewer, more basic EF skills such as sequencing. For this reason, the CAS trials assessing switching ability were of particular interest in the current study.

To directly assess switching ability through the PCo-switching trials, composites of raw trial scores were calculated. The total Planned Connections scaled score was not used because it combines scores from PCo-sequencing and PCo-switching trials, preventing the separate examination of contributions from these unique cognitive skills. Thus, the average of each adolescent's response time for the three PCo-sequencing trials was calculated, as was the average of each adolescent's response time for the two PCo-switching trials. These averages were used in subsequent analyses; because the averages represent response time, higher averages indicate more difficulty with the relevant EF skill. The Planned Codes (PCd) subtest also assesses planning ability and requires the youth to assign codes to an array of letters using a key. Youth can complete the codes in any order, allowing the use of strategy to enhance speed of completion. Scaled scores from this subtest were used in analyses; higher scaled scores indicate better performance of the relevant EF skill. PCd was weakly correlated with PCo-Switching ($r = -.25$, $p = .009$) but was highly correlated with PCo-sequencing ($r = -.75$, $p < .001$). PCo-sequencing was moderately correlated with PCo-switching ($r = .39$, $p < .001$). The CAS has been found to correlate moderately with other tests assessing similar constructs, such as Trail Making A and the Stroop task and to have adequate to high test-retest reliability (Strauss, Sherman, & Spreen, 2006).

Observational Family Measures

Observational measure of parental intrusiveness. Observational data were coded using a global coding system developed by Johnson and Holmbeck (1995) that is based on a methodology devised by Smetana et al. (1991; see Holmbeck et al., 2002 for a detailed description). Coders viewed two family interaction tasks on videotape and provided Likert-scale ratings on a variety of dimensions. This coding system taps Levy's (1943) four dimensions of overprotectiveness (OP): (a) parental prevention of independent behavior in the child (which included two codes: nonverbal prevention of exploratory behavior in the child and parental encouragement of the child's expression of individual views or opinions; reverse coded), (b) excessive physical contact with child, (c) infantilization (which included two codes: parental behavior that infantilizes the child and active catering to the child), and (d) excessive parental control (i.e., intrusiveness). All codes indexed excessive (rather than normative) levels of parental protection, given the developmental level of the child. Because of low frequencies of occurrence, two codes were dropped from further analysis (i.e., nonverbal prevention of exploratory behavior in the child and excessive physical contact with the child). For the four remaining codes, mother and father behaviors were coded separately, for a total of eight OP codes.

Items were coded on 5-point Likert scales; the manual includes behavioral descriptions for each of the points along the Likert scale. Any score greater than 1 indexed the presence of excessive parental protection. Undergraduate and graduate student coders were trained for roughly 8–10 hours until they had achieved 90% agreement with an expert graduate student coder (during training, agreement was assumed when two codes were within one Likert-scale point). All coders were blind to the specific purpose of the coding system and the hypotheses of this study, but not necessarily to the group status of the child. During training, average coder agreement was 93.6%. During actual coding, all tasks on all tapes were coded by two coders. After coding was completed, the average interrater reliabilities

(intraclass correlation coefficients; Suen & Ary, 1989) across coders, tasks, and parents were .83 and .66 for the SB and comparison samples, respectively. These values were deemed acceptable in accordance with past recommendations (Hartmann & Wood, 1990; Landis & Koch, 1977). The somewhat lower rater reliability for the comparison sample appears to have been due to the lower frequencies of intrusiveness (and lower variability) for this sample. For data analysis, the value for each coding item reflects an average of the responses of two independent coders across the two family interaction tasks. The average internal consistency *scale* alphas for the total intrusiveness scale were .80 and .77 for the SB and comparison samples, respectively.

Observational child dependency. As a measure of autonomy, observed child dependent behavior was coded using the same scheme developed by Johnson and Holmbeck (1995), as described above. Trained undergraduate and graduate-level coders viewed family interaction tasks and then provided 5-point Likert scale ratings (ranging from "almost never" to "almost always") assessing the child's independent/dependent behaviors. Any score greater than one indexed the presence of excessive child dependent behavior. The child dependent behavior scale included five codes: (a) "child engages in nonverbal exploratory behavior" (reverse scored), (b) "child expresses individual views/opinions" (reverse scored), (c) "child is needy," (d) "child seeks an excessive amount of physical contact," and (e) "child acts like a baby." As was done when coding parental intrusiveness, all items in the coding system were rated by two coders for two interaction tasks across all families. Ratings were then combined across the two raters and two tasks to create one code for each variable for each family. Scale alpha for the observed dependent behavior scale was .54 across the comparison group and the group with SB. Interrater reliability was .78 across the comparison group and the group with SB.

RESULTS

As discussed above, 61 adolescents with SB and 65 matched comparison sample adolescents were included in the current study ($N = 126$). The two samples were found to significantly differ in estimated intellectual functioning (PPVT-R score), $t(123) = -6.00$, $p < .001$. The sample with SB performed in the lower end of the average range ($M = 92.37$), while the comparison sample performed in the upper end of the average range ($M = 109.63$). As a result, the PPVT-R score was used as a covariate to ensure that any observed differences between the samples could be attributed to group status rather than overall verbal intellectual ability.

Because a number of adolescents with SB required shunts to treat hydrocephalus or reported a history of seizures, independent samples *t*-tests were conducted to determine whether adolescents with shunts and/or a history of seizures differed on any variables considered in the study. This required that 42 *t*-tests (14 Study Variables × 3 Shunt/Seizure Variables) be conducted to determine the effect of the presence of a shunt, the need for shunt revisions, and seizure activity. Bonferroni correction was used to control family-wise error rate for this large number of analyses, with a corrected value of $p = .004$ (.05/14) required for statistical significance. The presence of a shunt only produced differences in measures of executive functioning. Adolescents with SB and a shunt were rated by parents as having more problems with executive functioning on the BRIEF, $t(55) = 3.25$, $p = .002$, and were slower when completing the performance measures of planning, $t(46) = -5.37$, $p < .001$, sequencing and planning, $t(46) = 3.77$, $p < .001$, and switching, $t(46) = 3.10$,

$p < .001$, than unshunted adolescents with SB. No other variables were significantly different based on shunt status ($ps > .05$). A history of shunt revisions or replacements did not significantly impact any study variable nor did the presence of a history of seizures ($ps > .05$).

Executive Functioning in SB

Four independent-samples *t*-tests were conducted to compare adolescents in the comparison sample with those with SB on parent-reported executive functioning problems (BRIEF score), planning and sequencing ability (Planned Codes [PCd] and Planned Connections sequencing subtests [PCo-sequencing]), and switching ability (Planned Connections switching subtests [PCo-switching]). As expected, adolescents in the comparison sample performed significantly better than adolescents with SB on CAS subtests assessing planning (PCd), $t(109) = -7.93$, $p < .001$, and sequencing ability (PCo-sequencing), $t(109) = 8.39$, $p < .01$, and on those assessing mental flexibility and switching ability (PCo-switching), $t(109) = 2.45$, $p = .016$. Furthermore, the parents of adolescents with SB rated them as having significantly more executive functioning problems on the BRIEF ($M = 1.80$) than did parents of adolescents in the comparison sample ($M = 1.65$), $t(120) = 2.33$, $p = .02$.

Executive Functioning and Autonomy Development

To test whether parent-report and performance measures of executive functioning were predictive of adolescents' behavioral and emotional autonomy, 10 regression analyses were conducted with estimated verbal ability (PPVT-R) as a covariate. These analyses used parent report on the BRIEF and youth performance on the three subtests of the CAS as predictors and parent report of behavioral autonomy, teacher report of intrinsic motivation, observational child dependency, and child report of maternal and paternal emotional autonomy as dependent variables (2 EF Tests × 5 Autonomy Outcomes = 10 regressions). Regression findings were tabled only if a BRIEF or CAS scale emerged as a significant predictor.

Preliminary analyses. Prior to conducting analyses examining the relation of executive functioning to adolescents' level of autonomy, participants in the comparison sample and those with SB were compared on the five measures of autonomy using independent sample *t*-tests. Consistent with Friedman et al. (2009), adolescents with SB were rated by independent observers as displaying significantly more dependent behavior than adolescents in the comparison sample, $t(106) = 5.14$, $p < .001$. Moreover, adolescents with SB rated themselves as having less emotional autonomy from their mothers, $t(120) = -2.27$, $p = .03$, and their fathers, $t(115) = -3.11$, $p = .002$, than did adolescents in the comparison sample. Adolescents with SB were also rated by teachers as exhibiting less intrinsic motivation than their peers in the comparison sample, $t(107) = -4.66$, $p < .001$. However, there were no differences between adolescents in the comparison sample and those with SB on a parent- and child-report measures of behavioral autonomy, $t(112) = 0.11$, $p > .05$.

Observed child dependent behavior. In two separate regression analyses, a parent-report measure of executive functioning problems (BRIEF) and performance

measures of planning, sequencing, and switching abilities (PCd, PCo-sequencing, and PCo-switching, respectively) were employed as predictors of observed child dependent behavior. Group status (SB vs. comparison sample, with SB coded as 0 and comparison coded as 1) and interactions between-group status and neuropsychological predictors were added in later steps in the analyses to determine whether executive functioning measures predicted autonomy differentially across samples. The PPVT-R was used as a covariate in both analyses. Finally, common method variance was reduced by using different data sources for the predictor and outcome variables in all regression analyses in this section and in subsequent sections (e.g., parent data predicting child data or observational data predicting questionnaire data).

Total BRIEF scores were found to be significantly associated with observed child dependency, $\beta = 0.23$, $F = 7.70$, $p = .007$ (see Table 1). Specifically, adolescents with more parent-reported executive functioning problems exhibited more dependent behavior. However, the interaction between BRIEF score and group status was not significant, suggesting that the relationship between BRIEF score and observed dependency did not differ by group (Table 1).

The results for the regression analysis predicting observed child dependent behavior from performance measures of planning (PCd) and sequencing (PCo-sequencing) and switching ability (PCo-switching) are listed in Table 2. To assess the relative contribution of sequencing and switching abilities for autonomy development, raw scores (average response times) from the Planned Connections subtests were used in analyses, as standardized scores are not calculated for individual tasks in this subtest. Scaled scores from Planned Codes (PCd) were also entered in to the analyses. PCd, PCo-sequencing, and PCo-switching scores were added as main effects in a stepwise fashion to assess the relative contribution of these executive abilities for autonomy development. As in the regression analysis with BRIEF scores, interaction terms between group status and EF scores were entered in a subsequent step to determine whether EF-autonomy relationships differed by group membership.

Sequencing ability was found to be significantly associated with observed dependent behavior, $\beta = 0.41$, $F = 18.16$, $p < .001$, see Table 2. Specifically, adolescents who have difficulty with sequencing also exhibit more dependent behavior. Interactions between PCo

Table 1 Regression Results for Prediction of Autonomy and Parenting Behaviors from Parent Report on the BRIEF.

Step	Variable	β	R	ΔR^2	F
	DV = Observational Child Dependency				
1	PPVT	−0.47	.47	.22	30.66***
2	BRIEF total score (M+ F)	0.23	.53	.05	7.70**
3	SB Status	−0.21	.56	.03	5.01*
	DV = Intrinsic Motivation (Teacher Report)				
1	PPVT	0.43	.43	.18	24.14***
2	BRIEF total score (M+ F)	−0.28	.51	.08	11.40**
3	SB Status	0.19	.54	.03	4.01*

Note. The BRIEF variable is a composite of mother and father report. Beta weights are standardized and indicate the direction of the effect at the step the predictor entered the equation. SB = SB status.

*$p < .05$. **$p < .01$. ***$p < .001$.

and PCd scores and group status were nonsignificant, indicating that relations did not vary by group membership (Table 2).

Emotional autonomy from parents. The contribution of parent-reported EF problems and performance measures of planning, sequencing, and switching abilities for emotional autonomy from mothers and fathers was assessed in four separate regression analyses. Parent-rated executive functioning problems on the BRIEF were unrelated to emotional autonomy from both mother and father. The results of the regression analyses predicting emotional autonomy from the mother and father from the CAS measures are shown in Table 2. Overall, main effects were nonsignificant (see Table 2). However,

Table 2 Regression Results for Prediction of Autonomy and Parenting Behaviors from Child's CAS Scores.

Step	Variable	β	R	ΔR^2	F
	DV = Observational Child Dependent Behavior				
1	PPVT	−0.48	.48	.23	32.15***
2	PCo-Sequencing	0.41	.59	.11	18.16***
	DV = Emotional Autonomy from Mother (Child Report)				
1	PPVT	0.22	.22	.05	5.66*
2	PCo-Sequencing	−0.12	.24	.01	1.16
3	SB Status	0.07	.25	.003	0.33
4	PCd	0.003	.25	.00	0.00
5	PCo-Switching	0.00	.25	.00	0.00
6	SB × PCo-Switching	−0.72	0.38	0.08	10.08**
	DV = Emotional Autonomy from Father (Child Report)				
1	PPVT	0.11	.11	.01	1.20
2	SB Status	0.33	.30	.08	8.97**
3	PCo-Sequencing	−0.10	.31	.005	0.59
4	PCo-Switching	0.08	.32	.006	0.64
5	PCd	−0.04	.32	.001	0.07
6	SB × PCo-Switching	−0.46	.37	.03	3.83*
	DV = Intrinsic Motivation (Teacher Report)				
1	PPVT	0.46	.46	.21	25.85***
2	PCd	0.24	.51	.05	6.46*
3	SB Status	0.15	.52	.01	1.75
4	PCo-Switching	−0.11	.53	.01	1.50
5	PCo-Sequencing	−0.03	.53	.00	0.04
6	SB × PCo-Switching	−0.48	.57	.04	5.68*
	DV = Maternal Observational Intrusiveness				
1	PPVT	−0.54	.54	.29	42.06***
2	PCd	−0.23	.58	.04	6.87**
	DV = Paternal Observational Intrusiveness				
1	PPVT	−0.20	.20	.04	2.92
2	SB Status	−0.34	.36	.09	7.04**
3	PCo-Switching	−0.09	.37	.008	0.61
4	PCo-Sequencing	0.16	.38	.01	0.98
3	PCd	0.14	.39	.007	0.52
4	SB × PCo-Sequencing	0.97	0.45	0.05	4.14*
5	SB × PCd	−0.66	.53	.08	6.95**

Note. Beta weights are standardized and indicate the direction of the effect at the step the predictor entered the equation. SB = SB status; PCo = planned connections.

*$p < .05$. **$p < .01$. ***$p < .001$.

there was a significant interaction between group status and switching ability for emotional autonomy from mother ($\beta = -0.72$, $F = 10.08$, $p = .002$), and for emotional autonomy from father ($\beta = -0.46$, $F = 3.83$, $p = .05$). Post hoc regression analyses revealed that the relationship between emotional autonomy from the mother and switching ability was significant for adolescents with SB ($\beta = -0.60$, $F = 8.01$, $p = .006$) but not for those from the comparison sample ($p > .05$). More specifically, adolescents with SB who have more difficulty with mental flexibility and switching exhibited less emotional autonomy from the mother, while a similar relationship was not found for those in the comparison sample. This interaction is presented in Figure 1. Post hoc analyses revealed a similar relationship for analyses involving fathers, such that the relationship between switching ability and emotional autonomy from fathers was significant, although only marginally, for adolescents with SB ($\beta = -0.38$, $F = 3.73$, $p = .08$) but was not significant for their typically developing peers ($p = .15$).

Intrinsic motivation. The results of the regression predicting intrinsic motivation from parent-reported executive functioning problems can be found in Table 1. BRIEF scores were found to be significantly related to intrinsic motivation, $\beta = -0.28$, $F = 11.40$, $p = .001$, indicating that adolescents with more parent-reported executive functioning problems also exhibit less teacher-reported intrinsic motivation in the classroom (see Table 1).

Planning abilities, assessed with PCd, were also significantly related to intrinsic motivation, $\beta = 0.24$, $F = 6.46$, $p = .01$, such that adolescents with more difficulty in planning displayed less teacher-reported intrinsic motivation (see Table 2). Also, a significant interaction between group status and switching ability emerged, $\beta = -0.48$, $F = 5.68$, $p = .02$. Post hoc regression analyses revealed that the relationship between intrinsic motivation and switching ability was significant for adolescents with SB, $\beta = -0.65$, $F = 9.28$, $p = .003$, but not for adolescents in the comparison sample ($p > .05$). Adolescents with SB who have more difficulty with mental flexibility and switching were found to have less teacher-reported intrinsic motivation.

Behavioral autonomy. BRIEF and CAS scores were unrelated to behavioral autonomy either as main effects or in interaction effects.

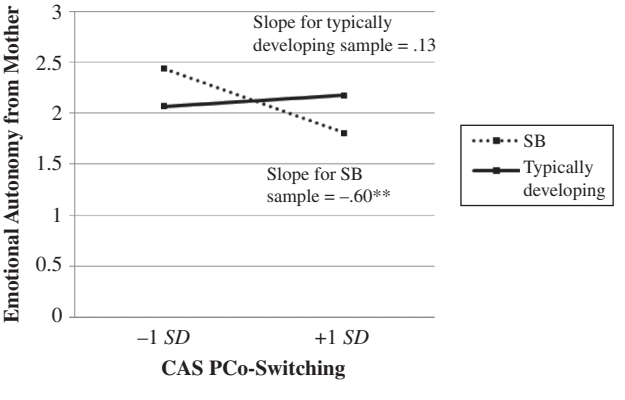

Figure 1 Emotional autonomy from mother as a function of CAS PCo-Switching performance and Group Status, with higher PCo-Switching scores indicating more EF difficulties (**$p < .01$).

Executive Functioning and Parenting Behaviors

Preliminary analyses. Prior to analyses examining the relation of executive functioning to autonomy-related parenting behaviors, participants in the comparison sample and those with SB were compared on four measures of autonomy-related parenting behaviors using independent sample *t*-tests. As expected, both mothers, $t(105) = 4.21$, $p < .001$, and fathers, $t(71) = 3.20$, $p = .002$, of adolescents with SB were rated by independent observers as exhibiting more intrusiveness than parents of adolescents in the comparison sample. However, there were no differences found between the groups on a parent-report measure of parental intrusiveness for either mother or father (*p*s > .05).

Observational parental intrusiveness. Parent-reported executive functioning problems on the BRIEF were unrelated to both maternal and paternal observed intrusiveness (*p*s > .05). Additionally, interactions between group status and BRIEF scores were not significant for either maternal or paternal observed intrusiveness, indicating that their relationship is constant across groups. There was a significant relationship between planning ability (measured with the PCd) and observed maternal intrusiveness, $\beta = -0.23$, $F = 6.87$, $p = .01$ (see Table 2). Specifically, adolescents who had more difficulty with sequencing had mothers who displayed more observed intrusiveness.

For the prediction of observed paternal intrusiveness, the interaction of group status with sequencing ability was significant for PCo-sequencing, $\beta = 1.04$, $F = .06$, $p = .01$. Similarly, the interaction between planning, measured by the PCd, and group status was also significant, $\beta = -0.66$, $F = 6.95$, $p = .01$ (see Table 2). Post hoc regression analyses revealed that the relationship between observed paternal intrusiveness and sequencing ability (PCo-sequencing) was marginally significant for adolescents with SB, $\beta = .28$, $F = 2.79$, $p = .10$, but not for those in the comparison sample ($p > .05$). More specifically, adolescents with SB who have more difficulty with sequencing had fathers who displayed more intrusiveness, a relationship not seen for adolescents without SB. Post hoc probing of the interaction between planning, measured by the PCd, and group status revealed that the relationship was not significant for either group (*p*s > .05).

Questionnaire measure of parental intrusiveness. The composite measure of self- and child-reported maternal and paternal intrusiveness was found to be unrelated to BRIEF and CAS scores (*p*s > .05).

DISCUSSION

The purpose of the current study was to examine the relationship between executive functioning, autonomy development, and parental intrusiveness in adolescents with and without spina bifida. Consistent with past research (e.g., Burmeister et al., 2005; Rose & Holmbeck, 2007), adolescents with spina bifida were found to exhibit more parent-reported EF problems and also performed at lower levels on performance EF measures than adolescents in the comparison sample. Adolescents with spina bifida also exhibited lower levels of autonomy development and had parents who exhibited higher levels of intrusiveness than their counterparts in the comparison sample. These findings parallel past research (e.g., Friedman et al., 2009; Holmbeck et al., 2002, 2003) and point to the importance of considering autonomy development and its relation to EF in this population. The current study found that both parent-reported levels of executive functioning problems

and performance-based measures of EF predicted measures of autonomy development and parental intrusiveness.

In terms of observed child dependency, both parent-reported measures of EF problems and a performance measure of sequencing ability (PCo-sequencing) were predictive. Higher levels of EF problems and more difficulty on tasks assessing sequencing were associated with more dependency. These relationships held for adolescents with SB as well as for adolescents from the comparison sample. Such findings coincide with reports of lower levels of EF being associated with less self-care abilities (Ries et al., 2003) and with lower levels of various independence outcomes in samples with SB (Sawin et al., 2003).

In contrast to results with child dependency, parent-reported levels of EF problems were unrelated to emotional autonomy from mother or from father. This was true for both adolescents with spina bifida and adolescents in the comparison sample. Mental flexibility and switching ability, on the other hand, were predictive of emotional autonomy from mother and father exclusively for adolescents with SB. Adolescents with SB who had more difficulty on tasks assessing mental flexibility and switching were more emotionally dependent on their parents. One explanation for this finding may be that an adolescent's difficulty with mental flexibility may also be manifested in an inability to transition from a child's complete reliance on their parents to an adolescent's ability to discern when they can rely on themselves instead of their parents. Alternatively, it may be that parents of adolescents with difficulties in mental flexibility discourage the adolescent's self-reliance in an effort to protect and care for them. Regardless, it appears that EFs, particularly the ability to maintain mental flexibility, are particularly important to the development of autonomy for adolescents with SB. Because adolescents with SB have additional barriers to autonomy development, having adequate EF abilities may be necessary to overcome these increased demands.

Intrinsic motivation was significantly predicted both by parent-rated EF problems and a performance measure of planning ability, regardless of group status. Adolescents in both samples whose parents reported more EF problems or who had difficulty on a measure of planning ability displayed lower levels of teacher-reported intrinsic motivation. There was also a significant interaction between switching ability and group status, so that the relationship between mental flexibility and intrinsic motivation was only significant for adolescents with SB. It appears that mental flexibility may be particularly important in fostering the development of intrinsic motivation for adolescents with SB. In general, the deficits of adolescents with difficulties in planning or mental flexibility may make them more cautious in school settings and more reliant on the teacher in order to compensate for these difficulties and to ensure optimal academic performance.

With regards to an observational measure of maternal and paternal intrusiveness, a performance measure of planning ability was significantly related to maternal observational intrusiveness for both samples. Adolescents who had more difficulty on this test of planning also had mothers who displayed more intrusiveness during videotaped family interactions. In terms of paternal observational intrusiveness, while there were no main effects of any measure of EF, there was an interaction between a performance measure of sequencing and group status, which revealed that this relationship was significant only for adolescents with SB. Adolescents with SB who exhibited difficulties in sequencing also had fathers who displayed more intrusiveness. It appears that children with EF difficulties had relatively intrusive mothers regardless of group status. It may be that having a child who exhibits EF deficits in everyday life increases maternal worry about her child and therefore increases her intrusiveness into her child's life. In contrast, when paternal

intrusiveness was examined, this relationship was only found for youth with SB. It may be that fathers of typically developing children are either not alarmed by EF deficits in their children or spend less time with their children than mothers, exposing them to such behavior less often. This would dilute the relationship between EF deficits and paternal intrusiveness in the typically developing sample. However, fathers of adolescents with SB may be hypervigilant for signs that their adolescent is having difficulty, and, when such signs are detected, fathers may then exhibit more intrusiveness.

It is intriguing that, in several analyses, performance measures rather than parent-reported measures of EF were predictive of autonomy development and parenting. This may have occurred because these measures assess different manifestations of EF (Rose & Holmbeck, 2007). The performance measures of EF used in this study assess the ability to quickly plan one's responses, to develop strategies for efficient responding, to hold information in working memory while sequencing stimuli, and to flexibly switch mental sets. While performance-based assessments are important in that they can isolate and directly measure different EFs, they do not address whether such deficits observed in a laboratory setting would also occur in the person's everyday life. In contrast, report measures like the BRIEF assess behavioral and social manifestations of problems with EF. Therefore, it may be that adolescents with SB have deficits in planning, sequencing, and mental flexibility but are able to minimize their effects in real-life situations.

An additional explanation may be that performance measures of EF also incorporate visuospatial and motor components (Rose & Holmbeck, 2007). In fact, Fletcher et al. (1996) found that significant differences in performance on neuropsychological measures in a sample with SB were reduced to nonsignificance when motor coordination was statistically controlled. However, this cannot explain the significant group differences in BRIEF scores or the relations between the BRIEF and certain measures of autonomy found in the present study. Furthermore, in the regression analyses, the main effects of performance measures were added in a separate block prior to interactions. This statistically controls for the main effects of these EF measures, including motor coordination, prior to assessing the significance of interactions. Despite this, several interactions involving performance EF measures reached significance, suggesting they were significant above and beyond differences in motor coordination.

An interesting pattern emerged in the relative predictive abilities of planning, sequencing, and mental flexibility/switching ability. In all cases, when a main effect of a performance measure of EF was found, it was for planning and sequencing instead of mental flexibility and switching ability. In contrast, when an interaction occurred, it involved mental flexibility and switching ability, with one exception. Planning may preferentially predict autonomy development and parental intrusiveness overall because it is a relatively more basic EF skill than the ability to maintain mental flexibility and to switch mindsets, which require multiple EF skills for one to perform well. As a result, planning may have more general, far-reaching effects on autonomy development, regardless of the presence of SB. However, when variables were differentially related depending on group status, it was mental flexibility that played an important role. As mentioned earlier, mental flexibility and switching ability are viewed as more difficult, higher level EF skills to master. For adolescents with SB, who have more EF deficits and barriers to autonomy development than the average adolescent, the presence of well-developed mental flexibility skills may allow adolescents to overcome these barriers in order to successfully develop autonomy.

The present study had several strengths. It is the first to examine the impact of EF on autonomy development and parental intrusiveness in adolescent samples with and without

SB. Furthermore, it used several observational measures of autonomy and parenting as well as performance measures of EF. However, this study also had certain limitations. The cross-sectional nature of the analyses precludes causal interpretations. Additionally, the study's power to detect significant results may have been attenuated by a small sample size; although this issue is typical in studies of pediatric populations. The presence of a large number of participants with shunted hydrocephalus in this sample and the relationship of shunt status with EF implies that this subpopulation of adolescents with SB may be particularly vulnerable to difficulties with autonomy development. Whether the present findings replicate with a sample of adolescents with SB who are not shunted should receive further examination in the literature. The current study's sample also included only a limited number of Latino families. It is important for future studies in this area to include more Latino participants, as SB has been found to preferentially affect this population. Future research should also employ a longitudinal design to determine whether the relationship between EF and autonomy development is prospective.

The results of this study have implications for fostering autonomy development in adolescents with spina bifida. While the correlational nature of this study's findings does not eliminate the possibility of alternate causal explanations, it does suggest several possibilities for intervention strategies to enhance autonomy development in this population. In light of their difficulties in planning and mental flexibility, educators and mental health providers can develop interventions that target these important executive functions. Furthermore, educational information can be prepared for parents of adolescents with spina bifida focusing on parenting behaviors that promote autonomy development.

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