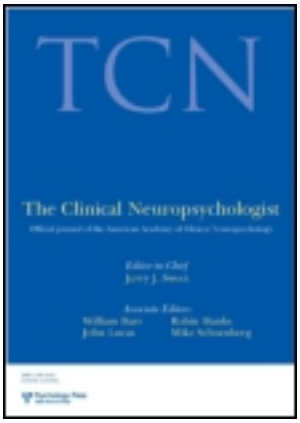

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Parent- and Self-Ratings of Executive Functions in Adolescents and Young Adults With Spina Bifida

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This study examined the agreement and consistency of parent- and self-report of executive functioning (EF) (Behavior Rating Inventory of Executive Functions; BRIEF) in an adolescent cohort of youth with myelomeningocele and shunted hydrocephalus (MMH). A total of 30 youth participants with MMH and their parents were recruited during adolescence (age 11–18, mean age 14), and a smaller sample ($n = 13$) was re-evaluated during young adulthood (age 18–26, mean age 22). Parent- and self-report T-scores were moderately correlated during adolescence (General Executive Composite, GEC, $r = .504$, $p = .007$) and adulthood (GEC, $r = .571$, $p = .041$). Compared to adolescent self-ratings, parent-ratings suggested higher levels of overall executive dysfunction and problems with metacognitive abilities during adolescence. Preliminary results from a small follow up sample, however, suggest that self- and parent-report of executive functioning may become more comparable during young adulthood. These preliminary data also suggest stability of deficit and/or possible improvement in executive presentation during the transition from adolescence to adulthood in this clinical population. Implications of these findings are discussed.

Keywords: Spina bifida; Executive function; Behavior Rating Inventory of Executive Functions; BRIEF; Adolescent; Myelomeningocele.

INTRODUCTION

Myelomeningocele is the most common neural tube defect compatible with life, and continues to occur in approximately 0.5 children per 1000 live births (Edmonds & James, 1990). Hydrocephalus is a common co-occurring condition with myelomeningocele (MMH), and is frequently treated via shunting in the days and weeks following birth (but see Bowman & McLone, 2010, for a review of changing practices in surgical management). In the past three decades medical advances have greatly increased the survival rates of individuals with MMH, and many individuals with this condition now survive into adulthood (Hunt & Oakeshott, 2003; Oakeshott & Hunt, 2003). Nevertheless these individuals remain

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at high risk for a number of co-occurring conditions, including recurrent hydrocephalus secondary to shunt failure, tethered cord, neurogenic bladder and bowel, pressure sores/decubitus ulcers, kidney failure, lower extremity paralysis, and seizures (Charney, 1992; Northrup & Volcik, 2000). These risks and associated conditions create a unique, MMH-specific constellation of atypical self-care competencies that add additional self-care burden to the functioning of these individuals (Tarazi, Mahone, & Zabel, 2007). For example, bladder-voiding complications often require clean intermittent catheterization at regular intervals to maintain bladder and kidney function. Furthermore, lower extremity sensory loss frequently necessitates regular self-inspection for pressure ulcers as well as proactive implementation of prophylactic exercises (e.g., wheelchair push-ups).

Although general intellectual functioning in individuals with MMH frequently falls broadly within the average range, deficits in executive functioning (EF) (Burmeister et al., 2005; Iddon, Morgan, Loveday, Sahakian, & Pickard, 2004; Rose & Holmbeck, 2007) as well as other cognitive deficits (for review see Dennis & Barnes, 2010) have been well documented. As such, individuals with MMH are faced with increased adaptive skill requirements, while presenting with impairments in the executive abilities thought to be necessary for consistent self-care implementation in general (Tarazi et al., 2007). Further complicating the clinical picture, demands for application of EF skills are typically thought to increase with age, as greater levels of independent problem solving and behavioral self-regulation are expected of youth in general as they approach adulthood (Tarazi, Zabel, & Mahone, 2008).

Increased attention has been given to the development of EF and other skills that might impact the acquisition of self-management capabilities of youth with MMH as they transition into older adolescence and young adulthood (Thibadeau, Alriksson-Schmidt, & Zabel, 2010). Given the profile of executive dysfunction documented in youth and young adults with MMH, concern remains regarding the extent to which deficits in executive skills such as *initiation* and *working memory* may detract from the completion of required self-care skills such as clean intermittent catheterization (Tarazi et al., 2007). Moreover, there is emerging evidence that measurement of EF in youth with spina bifida during mid-adolescence can be a useful means of predicting later functional independence and successful acquisition of adult competencies in this clinical population (Heffelfinger et al., 2008; Zukerman, Devine, & Holmbeck, 2010)

As such, there is considerable interest in early assessment of EF in adolescence, with a need for effective clinic-based screening of these skills and other salient predictors of distal outcomes in this clinical population. The Behavior Rating Inventory of Executive Functions (BRIEF; Gioia, Isquith, Guy, & Kenworthy, 2000) has become a popular behavioral checklist that can be used to quickly and efficiently screen "real world" EF in the clinic setting. In several samples of youth with MMH, parent ratings on the BRIEF have suggested a number of problematic aspects of EF, particularly those clustered on the "metacognitive" composite of the BRIEF (i.e., initiation, working memory, plan/organize) (Brown et al., 2008; Mahone, Zabel, Levey, Verda, & Kinsman, 2002).

The added value of adolescent self-report of executive functioning to clinical care of this medical population is still in question. Mahone and colleagues (2002)

provided one of the first comparisons of self- and parent-report on the BRIEF using a pre-publication version of the Behavior Rating Inventory of Executive Function-Self-Report Version (BRIEF-SR; Guy, Isquith, & Gioia, 2004). Preliminary data collected using the BRIEF-SR were published using raw score totals/means rather than norm-referenced standardized scores, as the initial publication of findings (Mahone et al., 2002) occurred in advance of the availability of complete normative information for the BRIEF-SR (Guy et al., 2004). Mahone and colleagues (2002) reported higher parent-reported raw score means (relative to self-ratings) on a scale of planning/organization, but higher self-reported raw score means (compared to parent ratings) on scales suggestive of poor behavioral regulation. The current study was designed to examine the agreement between self- and parent-report of EF using normative information for the various standardized BRIEF forms during different time periods of development (i.e., adolescence and young adulthood). Moreover, this study was intended to examine the predictive validity of adolescent BRIEF scores with regard to the functioning of individuals with MMH during young adulthood. Since cross-sectional examination of raw score averages on the BRIEF suggests persistence of executive dysfunction across the course of adolescence (i.e., ages 10 to 18) (Tarazi et al., 2008), we hypothesized that parent-report on the BRIEF would result in elevated ratings of executive dysfunction during both adolescence *and* young adulthood. Moreover, as self-appraisal is thought to be a component of EF that is deficient in those with MMH, we hypothesized that parent-report on the BRIEF would result in higher ratings of executive dysfunction when compared to self-ratings provide by adolescents and young adults with MMH.

METHOD

Participants

The initial phase of this study involved enrollment of 30 adolescents with MMH (Mahone et al., 2002). Participants were recruited from the Spina Bifida Clinic (SBC) at the Kennedy Krieger Institute and were assessed within the course of routine multidisciplinary medical care (i.e., not specifically referred for neuropsychological assessment). Individuals with MMH are typically seen in this clinic every 6 to 12 months for comprehensive care and monitoring of spina-bifida-related medical issues (e.g., continence, wound care, orthopedics, orthotics, etc.). Participants with documented histories of MMH were recruited for study inclusion if they were between the ages of 11 and 18, and if they had not been diagnosed with intellectual disability/mental retardation based on existing medical or school records. To recruit a sample representative of the population of adolescents with MMH in general, patients were recruited from this routine medical care clinic, in most cases prior to any subsequent referral for neuropsychological evaluation. Initial recruitment methods are further described in Mahone et al. (2002).

Parents of adolescents enrolled in this study completed the Behavior Rating Inventory of Executive Functions-Parent Report Version (BRIEF-PR; Gioia et al., 2000). Similarly, 27 of the 30 adolescents enrolled in the initial phase of this study completed a pre-publication version of the Behavior Rating Inventory of Executive Function-Self-Report Version (BRIEF-SR; Guy et al., 2004) provided to the

researchers by the developers of this instrument. As noted, preliminary data collected using the BRIEF-SR were published using raw score totals/means rather than norm-referenced standardized scores, as the initial publication of findings (Mahone et al., 2002) occurred in advance of the availability of complete normative information for the BRIEF-SR (Guy et al., 2004). Following the publication of the BRIEF-SR technical manual, item responses from the BRIEF-SR protocols were transferred to the commercially available BRIEF-SR forms, and standardized scores (i.e., T-scores) were subsequently calculated using the published age-based norms.

The second phase of this study involved recruitment of those young adults with MMH (now between the ages of 18 and 26) who had participated in the initial phase of this study during their adolescence (ages 11 to 18). Recruitment was conducted via mailings with ''opt-in'' response postcards as well as by research flyers circulated in the Spina Bifida Clinic. All of the initial recruitment during adolescence, as well as subsequent recruitment during young adulthood, was conducted with the permission of the Institutional Review Board of the Johns Hopkins University School of Medicine.

Of these original participants, 4 were deceased and 13 were lost to follow-up; 13 of the original participants were enrolled in the second phase of this study. All but 3 of the 13 participants had completed the BRIEF-SR at the time of the initial study visit, and BRIEF scores based on parent ratings were available for all 13. Once consent was obtained from these young adults with MMH they were asked for permission to contact and enroll their parents in the second phase of the study as well. Subsequently, one parent was enrolled for each of the 13 young adults with MMH. Each parent enrolled during the second phase of the study was the same parent that was enrolled during the first phase of the study.

Measures

The Behavior Inventory of Executive Function-Parent Report Form. The BRIEF-PR (Gioia et al., 2000) is a caregiver-report questionnaire designed to assess the behavioral manifestations of executive function in children aged 5 to 18 years. The Parent Form contains 86 items on which parents rate each behavior as occurring ''never,'' ''sometimes,'' or ''often.'' The questionnaire takes approximately 10 to 15 minutes to complete. Items are organized into eight scales and two primary indices—Metacognition (MI) and Behavioral Regulation (BRI)—with an overall composite score—Global Executive Composite (GEC). T-scores are derived for each scale and Index, with higher T-scores indicating greater impairment. The Metacognition Index is comprised of five subscales (Initiate, Working Memory, Plan/Organize, Organization of Materials, and Monitor) and represents skills that are essential to self-regulation of *cognitive* processes. The Behavioral Regulation Index is comprised of three subscales (Inhibit, Shift, Emotional Control) and represents skills essential for the self-regulation of *behavior*. The validity of the BRIEF and the described two-factor structure has been supported in various clinical groups (Gioia, Isquith, Retzlaff, & Espy, 2002;

Mahone et al., 2002; Slick, Lautzenhiser, Sherman, & Eyrl, 2006). To avoid confusion this instrument is hereafter referred to as the BRIEF-PR in this paper.

The Behavior Inventory of Executive Function-Self-Report Version. The BRIEF-SR (Guy et al., 2004) is much like the BRIEF-PR, and requires the adolescent to provide one of three possible self-report ratings (e.g., never, sometimes, often) to 80 different behavioral items assessing the application of EF skills. The BRIEF-SR was normed and validated for use with adolescent youth between the ages of 11 and 18, and was written to require late fourth-grade-level reading skills.

There are differences between the adolescent self-report and parent-report versions of the BRIEF with regard to content, item wording, and scale composition. The test manual indicates that the BRIEF-PR items and scales "served as a starting point for the development of the items" (Guy et al., 2004, p. 37) for the BRIEF-SR scales. This reportedly began by adapting the 86 items from the BRIEF-PR into "self-statements," with subsequent removal of items considered inappropriate for adolescents as well as the addition of several new items consistent with the BRIEF theoretical executive function domains. Final scale composition was based on results from item–total correlation analyses and factor analyses. Of note, these processes resulted in the elimination of the Initiate scale from the BRIEF-SR, and reconfiguration of the Monitor scale to include only items assessing social self-monitoring. The items relating to task-oriented monitoring that were previously part of the Monitor scale on the BRIEF-PR were combined with new item content to form the Task Completion scale on the BRIEF-SR. In summary, the test development of the BRIEF-SR resulted in the elimination of one executive domain (i.e., Initiate), the addition of a new executive domain (i.e., Task Completion), and the retention of the following executive domains: Inhibit, Shift, Emotional Control, Monitor, Working Memory, Plan/Organize, and Organization of Materials. The factor structure of the BRIEF-SR composite scales is presented in Table 1.

The BRIEF-SR manual (Guy et al., 2004) describes score consistency between the BRIEF-PR and BRIEF-SR within a combined clinical and normative subsample ($n = 148$). For this mixed sample, moderate correlations were reported between T-scores based on parent ratings (BRIEF-PR) and self-ratings (BRIEF-SR) on the Behavioral Regulation ($r = .52$), Metacognition ($r = .57$), and the Global Executive Composite ($r = .56$) Indices. In this subgroup it was rare (i.e., occurring in only 4–5% of the cases) for adolescents to generate Index scores on the BRIEF-SR that were one or more standard deviations above (i.e., more problematic) corresponding Index scores based on parent report on the BRIEF-PR. In contrast, BRIEF-PR T-scores frequently were one or more standard deviations higher than corresponding BRIEF-SR T-scores on the Behavioral Regulation (36.3%), Metacognition (33.4%), and the Global Executive Composite (38.8%) Indices. Similar disparity between parent- and self-report using the BRIEF-PR and BRIEF-SR has been described in other clinical populations (Hughes, Turkstra, & Wulfeck, 2009).

The Behavior Rating Inventory of Executive Function-Adult Versions. The Adult-BRIEF versions (Roth, Isquith, & Gioia, 2005) (i.e., informant report and self-report versions) are similar to the BRIEF-PR and

Table 1 Organization of BRIEF scales across various forms

BRIEF-PR	BRIEF-SR	Adult BRIEF
BRI	BRI	BRI
Inhibit	Inhibit	Inhibit
Shift	Shift	Shift
Emotional Control	Emotional Control	Emotional Control
–	Monitor	Self-Monitor
MI	MI	MI
Initiate	–	Initiate
Working Memory	Working Memory	Working Memory
Plan/Organize	Plan/Organize	Plan/Organize
Org. of Materials	Org. of Materials	Org. of Materials
Monitor	–	–
–	Task Completion	Task Monitor

BRIEF = Behavior Rating Inventory of Executive Function; PR = Parent Report; SR = Self-Report; BRI = Behavioral Regulation Index; MI = Metacognition Index; Org. = Organization.

BRIEF-SR in composition, including two pages of short behavioral statements requiring one of three possible respondent ratings. Both the self-report version (Adult BRIEF-SR) and informant-report version were developed using the same item content. Both Adult BRIEF forms were standardized and validated to assess the application of EF skills in men and women between the ages of 18 and 90. The Adult-BRIEF was written at a fourth-grade reading level. As the informant-report version of the BRIEF was completed solely by parents for the current study, it is hereafter referred to as the Adult BRIEF-PR (PR; Parent Report).

The Adult BRIEF-SR and Adult BRIEF-PR versions differ in some respects from the BRIEF-PR and BRIEF-SR with regard to content, item wording, and scale composition. The test manual reports that the Adult-BRIEF items and scales were developed according to theory, clinical practice, and research literature in executive functions, within the context of previous versions of the BRIEF, to allow consistency across measures. Development of the item pool for the Adult-BRIEF started with items contained on the BRIEF-PR. Subsequently, items considered to be inappropriate for assessment of adult functioning were eliminated, and some items were added to provide a better measure of adult function. Items were subsequently reduced based on item–total correlations and factor analysis was used to clarify scale structure. A Task Completion scale was found to correlate highly with a Monitor scale, so the shared content was reconfigured into separate Self-Monitor and Task Monitor scales. These two new theoretical scales on the Adult-BRIEF were combined with seven other clinical scales (i.e., Inhibit, Shift, Emotional Control, Initiate, Working Memory, Plan/Organize, and Organization of Materials). The factor structure of the Adult-BRIEF subscales is presented in Table 1.

The test manual describes informant-report/self-report score consistency within a combined clinical and normative subsample ($N = 180$). In this mixed sample moderate correlations were reported between Adult BRIEF-PR and Adult

Table 2 Mean age, sex, and BRIEF composite scores (GEC) of participants in adolescence

	Entire sample[a]	Retained sample	Dropouts[d]	*p*
Age (*SD*)	14.14 (2.37)	14.00 (2.41)	14.24 (2.41)	.798
Sex (% male)	53.3	61.5	47.1	.448
GEC-Parent (*SD*)	62.73 (14.20)	64.00 (14.17) [b]	61.76 (14.58)	.677
GEC-Self (*SD*)	56.11 (12.20)	60.10 (13.40) [c]	53.76 (12.91)	.236

p value reflects significance of difference between participants remaining in the study and those who did not. Sample sizes vary due to attrition; [a]$N = 30$; [b]$n = 13$, [c]$n = 10$, [d]$n = 17$; BRIEF = Behavioral Rating Inventory of Executive Functions; GEC = General Executive Composite; *SD* = standard deviation; GEC scores are reported as T-scores.

BRIEF-SR T-scores on the Behavioral Regulation ($r = .63$), Metacognition ($r = .61$), and the Global Executive Composite ($r = .63$) Indices. It was rare (i.e., occurring in only 7–9% of the sample) for Index scores from the Adult BRIEF-SR to be one or more standard deviations above (i.e., more problematic) corresponding Index scores on the Adult BRIEF-PR. In contrast, Adult BRIEF-PR T-scores frequently were one or more standard deviations higher than corresponding Adult BRIEF-SR T-scores on the Behavioral Regulation (24.6%), Metacognition (28.5%), and the Global Executive Composite (28.9%) Indices.

RESULTS

Analysis plan

Of the original 30 participants, 27 completed BRIEF-SR forms and all 30 were assessed by their parents at the initial time point (1998–2001) using the BRIEF-PR. A subsample of these participants and their parents also later completed appropriate Adult BRIEF rating forms during young adulthood (2008–2010). Sample demographics are presented in Table 2.

At the first time point the study examined comparisons of self- and parent-ratings of EF during adolescence. Composite and individual scale scores were compared to normative values (T-score = 50) and examined for agreement across raters. At the second time point the study examined self- and parent-ratings of EF in individuals with MMH during young adulthood. Ratings were examined for stability over time and agreement between raters.

Sample characteristics and attrition analyses

At time 1, 27 of the 30 adolescent participants, and all 30 of their parents, completed valid ratings on the BRIEF, with 13 of the original participants and their parents also subsequently completing valid ratings on the Adult BRIEF during the young adulthood phase of this study. There were no significant differences between the original adolescent participants and those available to complete ratings as young adults in terms of age at initial time point, $t(27) = 0.259$, $p = .798$, or gender composition, $\chi^2(1) = 0.621$, $p = .431$, of the sample. There were also no significant

Table 3 Mean parent-ratings ($N = 30$) and self-ratings ($n = 27$) in adolescence (T score, *SD*), compared with BRIEF normative means

			Paired comparisons[a]	
BRIEF Index	T score (*SD*)	*p*	*t*	*p*
GEC-Parent	62.22 (13.36)	.000	2.29	.031
GEC-Self	56.11 (13.20)	.024		
BRI-Parent	57.14 (12.10)	.001	1.18	.250
BRI-Self	54.44 (12.41)	.074		
MI-Parent	63.41 (14.19)	.000	2.14	.042
MI-Self	56.63 (12.83)	.012		

Paired comparisons represent differences on composite scores across raters. [a]$n = 27$; BRIEF = Behavioral Rating Inventory of Executive Functions; GEC = General Executive Composite; BRI = Behavioral Regulation Index; MI = Metacognition Index; *SD* = standard deviation; GEC scores are reported as T-scores.

differences among parent- and self-report BRIEF composite scores during adolescence (e.g., GEC, BRI, MI) between the original sample and those participants who remained in the study into young adulthood. Finally, there were no significant differences on parent- and self-ratings during adolescence between participants who remained in the study into young adulthood and those who did not (see Table 2), although there was a trend towards higher (i.e., more problematic) parent and self-ratings of EF in the retained sample relative to the participants lost to follow up.

Time 1: Adolescence

Mean self- and parent-report BRIEF Index scores were significantly higher than normative values (T-score = 50) during adolescence across most BRIEF composite scales (Table 3). All of the parent-rated Index scores were significantly higher than normative means, while two of the three self-rated Index means were also higher than normative means (MI and GEC). When paired BRIEF-SR and BRIEF-PR mean T-scores were compared, parent-report resulted in significantly higher T-score means on two of the three Indices—GEC: $t(26) = 2.29$, $p = .031$; MI: $t(26) = 2.14$, $p = .042$. In short, during adolescence, mean parent-report T-scores on the MI and GEC Indices were significantly higher than mean self-report T-scores, although mean self-report T-scores were higher than normative values in general.

Among comparisons of parent- and self-ratings on matched BRIEF subscales during adolescence, parent-report resulted in significantly higher T-score means on the Working Memory (BRIEF-PR T-score $M = 67.8$; $p < .001$) and Plan/Organize (BRIEF-PR T-score $M = 63.3$; $p = .002$) scales compared to self-report T-score means on these same subscales. The Initiate subscale is only included on the BRIEF-PR and is not on the BRIEF-SR. While parent- and self-report T-scores could thus not be compared directly on this scale, parent ratings of adolescents with MMH resulted in significantly higher T-scores than normative means (BRIEF-PR Initiate T-score $M = 64.6$; $p < .001$).

Table 4 Mean parent-ratings ($n = 13$) and self-ratings ($n = 13$) at young adulthood (T score, *SD*), compared with BRIEF normative means

			Paired comparisons[a]	
BRIEF Index	Adulthood	*p*	*t*	*p*
GEC-Parent	55.85 (9.62)	.049	.307	.764
GEC-Self	56.62 (9.90)	.033		
BRI-Parent	52.92 (10.56)	.338	.419	.683
BRI-Self	54.00 (10.34)	.188		
MI-Parent	57.38 (8.28)	.007	.174	.865
MI-Self	57.85 (9.80)	.014		

Paired comparisons represent differences on composite scores across raters; [a]$n = 13$; BRIEF = Behavioral Rating Inventory of Executive Functions; GEC = General Executive Composite; BRI = Behavioral Regulation Index; MI = Metacognition Index; *SD* = standard deviation; GEC scores are reported as T-scores.

Parent- and self-report composite scores were significantly correlated during adolescence (GEC, $r = .50$, $p = .007$; BRI, $r = .42$, $p = .030$; MI, $r = .41$, $p = .033$). However, there was a trend towards discrepancy between parent- and self-ratings in terms of clinical categorization. When examining clinical elevations, scores of 60 or higher (at least 1 *SD* above the mean) were considered "elevated." During adolescence, 56.7% ($n = 17$) of parent ratings of adolescents with MMH resulted in elevated GEC scores. In comparison, self-ratings during adolescence resulted in significantly fewer elevated GEC scores ($n = 9$; 33.3%; $p = .006$). T-scores from the BRIEF-PR were frequently 1 *SD* or higher than T-scores based on self-report provided on the BRIEF-SR, occurring at the following frequencies: GEC (37%), MI (25.9%), and BRI (37%). In short, parent- and self-ratings of EF in adolescents with MMH are significantly correlated, although parent-report T-scores were more likely to result in "clinically elevated" scores.

Time 2: Young adulthood

Of the original participants, 13 were examined at Time 2. These participants, now young adults, and their parents again completed the age-appropriate BRIEF rating forms (i.e., Adult BRIEF-SR and Adult BRIEF-PR). Of note, while significant differences were noted between BRIEF-SR and BRIEF-PR Index scores in the original adolescent sample, the same discrepancy was *not* identified between parent- and self-report during adolescence for the follow-up subgroup. During young adulthood, comparisons of participants' mean composite scores with BRIEF normative values (Table 4 and Figure 1) revealed T-score means that were significantly higher than normative values on the MI and GEC composite scales, but not on the BRI. Paired comparisons between parent- and self-report in young adults with MMH indicated no significant differences in severity of ratings on the composite scales.

Correlations between parent- and self-ratings during young adulthood remained strong (GEC, $r = .57$, $p = .041$; BRI, $r = .61$, $p = .028$; MI, $r = .45$, $p = .122$).

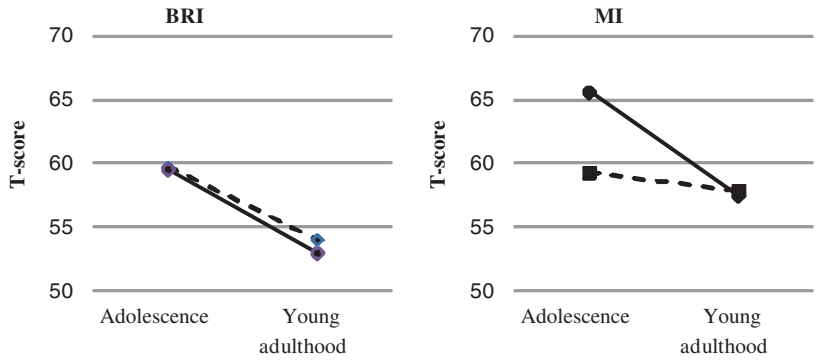

Figure 1 Longitudinal parent- ($n = 13$) and self-ratings ($n = 10$) from adolescence to young adulthood (BRIEF T-scores). Parent-report BRIEF scores are depicted as solid lines. Self-report BRIEF scores are depicted as dashed lines. BRIEF = Behavioral Rating Inventory of Executive Functions; BRI = Behavioral Regulation Index; MI = Metacognition Index.

There was general agreement between parent- and self-ratings in terms of clinical categorization. During young adulthood, 38.5% (5 of 13) of parent ratings of adults with MMH resulted in elevated GEC scores. Similarly, self-ratings during adulthood resulted in a comparable number of elevated GEC scores (7 of 13; 56.7%; $p = .196$). Of note, parent-report provided on the Adult BRIEF-PR rarely resulted in GEC T-scores that were one *SD* higher than GEC T-scores from the Adult BRIEF-SR, occurring in only one (7% of cases) individual for each composite scale.

Intra-individual comparisons of mean GEC T-scores across time points (adolescence and young adulthood) revealed significantly lower Adult BRIEF-PR T-score means in young adulthood compared with T-scores in adolescence: BRIEF-PR GEC, $t(12) = 2.303$, $p = .040$. Although not reaching the level of statistical significance, a moderate correlation was noted between parents' overall BRIEF ratings across time (i.e., BRIEF-PR and Adult BRIEF-PR; GEC, $r = .478$, $p = .098$). Examination of specific Index scores provides additional clarity of this trend: parent ratings were generally consistent on scales of behavioral regulation during adolescence and young adulthood (BRI, $r = .634$, $p = .020$), but were more discrepant on scales of cognitive self-regulatory abilities (MI, $r = .338$, $p = .258$), likely due to the pattern of lower parent-rated MI T-scores in young adulthood relative to adolescence. During adolescence, 61.5% of parent ratings resulted in elevated GEC scores, compared to 38.5% of parent ratings during young adulthood ($p = .079$).

A closer examination of the pattern of parent ratings over time for participants remaining in the sample into young adulthood revealed that, in this particular group, each of the individuals with MMH who were rated by their parents as "sub-clinical" (GEC score < 60, $n = 5$) on the BRIEF-PR during adolescence were subsequently rated as "sub-clinical" on the Adult BRIEF-PR. Similarly, about half (62.5%, $n = 5$; 4 males) of those individuals with MMH with elevated BRIEF-PR T-scores (GEC score ≥ 60) during adolescence continued to have elevated Adult BRIEF-PR T-scores. A subset of individuals with elevated

BRIEF-PR T-scores during adolescence ($n = 3$; two females) were rated as "sub-clinical" on the Adult BRIEF-PR. In short, in this small sample of individuals with MMH who had transitioned from adolescence to young adulthood, BRIEF T-scores based on parent report did not get worse over time, and longitudinal comparisons suggest a degree of possible EF "improvement" in a subgroup of this sample. Qualitatively, the participants who were "persistently elevated" based on parent report on the BRIEF over time also anecdotally reported greater levels of psychiatric concerns (e.g., emotional dysregulation, depression) both during adolescence and during young adulthood.

Comparisons of self-ratings over time, in adolescence and young adulthood, revealed non-significant correlations across corresponding BRIEF composites. No significant differences were observed when comparing GEC T-scores from self-ratings obtained during adolescence and young adulthood ($p = .959$). In the subgroup of individuals with self-ratings from both adolescence and adulthood, 70% (7 of 10) of the self ratings in young adults produced elevated GEC T-scores, as compared with 50% (5 of 10) of self-ratings acquired during adolescence. Of those whose self-ratings were elevated during adolescence (GEC score > 60, $n = 5$), 80% (4 of 5) produced elevated self-ratings during young adulthood. Three of the five individuals with "sub-clinical" (GEC score < 60) self-ratings on the BRIEF-SR during adolescence produced elevated ratings on the Adult BRIEF-SR.

DISCUSSION

Informant report of EF has become an important component in the neuropsychological assessment of medically involved youth. This study examined the pattern of self- and parent-reported EF skill development in youth with MMH across the adolescent–young adulthood transition. The findings from this study provide preliminary information regarding the development of EF over time in this medical population, as well as additional psychometric information specific to the clinical use of the family of BRIEF instruments themselves.

In terms of the development of EF, current findings are consistent with previous findings of executive dysfunction reported in *adolescents* with MMH, particularly in areas such as working memory, initiation, and organization (Brown et al., 2008). In our adolescent sample both parent- and self-reports resulted in mean T-scores that were significantly higher than normative values on the MI and GEC composite indices of the BRIEF. Moreover, in our small *adult* follow-up sample, parent- and self-reports continued to result in higher T-score means (relative to normative values), suggesting persistence of some aspects of executive dysfunction as youth with MMH transition into adulthood. These findings thus have relevance for transition-aged youth with MMH, particularly within the context of other research suggesting persisting learning and cognitive deficits into adulthood (Jenkinson et al., 2011). We have proposed that, when combined with other cognitive and psychosocial variables, executive dysfunction may further detract from the ability of young adults to become independent in managing aspects of their own self-care and medical self-management (Tarazi et al., 2007). Given the apparent persistence of EF concerns over time, as well as the potential for poor health care outcomes in this population, it will continue to be critical that health care services

maintain continuity of care, structure, and supports to accommodate for executive dysfunction as youth with MMH transition into adult roles and self-care responsibilities. While this is not the focus of this article, we encourage ongoing discussion of methods by which these transition-related EF issues could be addressed, including different case management approaches, technology applications, supported transition of medical home, and/or extension of specialized pediatric medical care to 25 years of age.

Poor accuracy of self-assessment has been described in populations at risk for executive dysfunction (Knouse, Bagwell, Barkley, & Murphy, 2005), and concerns are often raised regarding the capacity for insight of youth with MMH regarding symptoms of executive dysfunction. Self-assessment of EF has been proposed as an important component of treatment planning, and self-understanding/awareness of EF deficits has been proposed as an important factor for gauging the level of necessary external support in treatment and educational intervention (Guy et al., 2004). Several findings from the current study suggest that youth with MMH may demonstrate a degree of insight into the executive issues often associated with their medical condition. First, as noted above, mean T-scores based on self-report were higher than normative values (T-score = 50) during adolescence. Second, in this sample of adolescents with MMH, parent- and self-report resulted in moderately strong inter-rater reliability coefficients on the BRIEF composite indices. Both of these findings indicate a degree of parent–adolescent agreement regarding symptoms of executive dysfunction and suggest at least a limited degree of adolescent insight into these issues.

There was, however, significant discrepancy between parent- and self-report regarding the *degree* of executive dysfunction during adolescence, with parent ratings resulting in significantly higher mean T-scores on several BRIEF composite indices. This finding stands in marked contrast to the mixed preliminary findings based on BRIEF raw score means published by Mahone et al. (2002), which found higher ratings of behavioral disinhibition based on adolescent self-report, and higher ratings of disorganization and poor planning based on parent report. Due to the differences found in the current study, more adolescents with MMH were classified as ''clinically elevated'' on the BRIEF based on parent-report than were similarly classified based on adolescent self-report. As such, while our findings indicate some degree of parent–adolescent agreement regarding perceptions of EF competence during adolescence, there remain considerable differences with regard to the clinical implications of the resulting scores during this time period. Specifically, more youth with MMH will potentially be identified for EF-focused treatment services and/or educationally based accommodations based on parent-ratings on the BRIEF compared with self-ratings. Alternatively, relying on self-report alone may underestimate the actual level of executive dysfunction and related service need. Evidence-based practices regarding the treatment of and accommodation for executive dysfunction in adolescents are still being developed, but it has been proposed that accuracy of self-appraisal of EF may be a useful consideration in determining the extent of external structure necessary in treatment intervention. From this perspective, our findings argue for a combination of insight-based intervention *combined with* external support and behavioral intervention when

accommodating for or treating executive dysfunction in youth and young adults with MMH.

There are several possible explanations for the discrepancy between parent- and self-report of application of EF skills during the adolescence of youth with MMH. In addition to the aforementioned concerns regarding poor adolescent insight into their own executive dysfunction, there is also the possibility of parental over-endorsement of symptoms. One explanation for the high level of endorsement of symptoms by parents may involve the somewhat distinct family dynamics often associated with MMH, including atypical patterns of conflict and/or cohesion often observed during adolescence (Jandasek, Holmbeck, DeLucia, Zebracki, & Friedman, 2009). These unique trajectories of conflict/separation between parents and their adolescents with MMH may affect parent expectations for independent functioning, and therefore impact ratings of EF on the BRIEF. We have also proposed that the additional self-care and self-management expectations associated with MMH may contribute to increased report of "executive dysfunction" (Tarazi et al., 2007), such as elevated reports of initiation and working memory deficits associated with the atypical self-care requirement to initiate and "remember to remember" to perform self-catherization four to five times per day. Finally, differences in the content and composition of scales between the BRIEF-PR and BRIEF-SR may contribute to the differences noted between informants, particularly as the Initiate scale is present only on the parent-report version (and not the self-report version) of the BRIEF during adolescence.

In contrast to the differences noted between parent- and self-report of EF during adolescence, mean T-scores based on parent- and self-report were reasonably well correlated *and* well matched in terms of clinical intensity during young adulthood. It should be noted, however, that this follow-up subgroup did *not* display significant differences between self- and parent-ratings of EF during adolescence, suggesting that this group may not have been entirely representative of the original sample as a whole. For this reason there is a limited degree to which longitudinal data acquired from individuals at both time 1 and time 2 can be generalized to the population of individuals with MMH as a whole.

With this caveat in mind, however, the longitudinal portion of this study resulted in a number of promising findings. First, there is evidence to suggest increased agreement between parent- and self-ratings of EF during adulthood. There was only one instance (7%) of a parent–child dyad in which a noteworthy T-score discrepancy (>1 *SD*) was observed during the young adult years, with the rest of the parent–child dyads producing GEC T-scores within a standard deviation of each other. These preliminary results are surprising, as more pronounced disparity has been described between self- and parent-ratings of behavioral symptoms in other populations with central features of executive dysfunction, i.e., young adults with ADHD (Barkley, Fischer, Smallish & Fletcher, 2002). Second, there is evidence of stability of deficit and/or possible improvement in EF presentation in individuals with MMH over time. This, too, is somewhat unexpected, as cross-sectional analysis of raw scores from the BRIEF-PR suggests the potential for persistence of symptoms of executive dysfunction as youth with MMH move into adulthood.

There are a number of possible explanations for both the apparent increase in agreement between raters during young adulthood as well as stability and/or

possible reduction in symptoms of executive dysfunction over time, including the reintroduction of the Initiate scale/content into the self-report version of the Adult BRIEF, increased EF insight on the part of young adults with MMH, and ongoing development or potential "catch-up" of EF over time. There is also the possibility that changes in daily expectations associated with increasing age result in a lessening of EF requirements. For instance, parent report of symptoms of poor initiation, working memory, and organization may lessen in intensity when youth with MMH are no longer subjected to the executive demands of formal schooling (e.g., completion of homework, organization of large projects, retention of information for tests). Additional research will be necessary to further investigate these possibilities.

There are a number of limitations associated with this research that should be noted, including a small sample size, particularly with regard to the sub-sample that continued to participate in young adulthood. Second, it is unclear whether either the adolescent or adult sample was representative of individuals with MMH in general, as their medical home was in a specialized pediatric hospital setting and we did not include individuals seen in community clinics and practices. Finally, while the family of BRIEF forms shares much of the same content, findings may be due to differences between various versions of this measure rather than actual differences in EF in general.

In closing, from an evidence-based neuropsychological perspective, the preliminary results provided by this study suggest possible utility of screening EF with the BRIEF-PR during adolescence, particularly for the purposes of identifying necessary services in adolescence as well as anticipating EF needs of individuals with MMH as they transition into adulthood. Specifically, in this small sample of individuals, sub-clinical parent ratings of EF during adolescence were consistently associated with sub-clinical parent ratings of EF during young adulthood. Similarly, clinically elevated scores based on parent report during adolescence were often associated with elevated scores during young adulthood, although parent ratings of executive dysfunction declined from *clinical* to *subclinical* in a number of cases. As such, early parent report of executive dysfunction in adolescence may be a useful way of identifying youth at risk for persistent executive problems later in life. Qualitatively, those young adults with MMH who were rated by parents as showing persistently elevated levels of executive dysfunction also self-reported affective/emotional concerns as well. Additional inquiry will be necessary to determine if high-frequency emotional concerns in this population such as depression or anxiety (Bellin et al., 2010) or other factors mediate the expression of executive dysfunction in young adulthood.

REFERENCES

Barkley, R. A., Fischer, M., Smallish, L., & Fletcher, K. (2002). The persistence of attention-deficit/hyperactivity disorder into young adulthood as a function of reporting source and definition of disorder. *Journal of Abnormal Psychology*, *111*(2), 279–289.

Bellin, M. H., Zabel, A., Dicianno, B. E., Levey, E., Garver, K., Linroth, R., et al. (2010). Correlates of depressive and anxiety symptoms in young adults with spina bifida. *Journal of Pediatric Psychology*, *35*(7), 778–789.

Bowman, R. M., & McLone, D. G. (2010). Neurosurgical management of spina bifida: Research issues. *Developmental Disabilities Research Reviews*, *16*(1), 82–87.

Brown, T. M., Ris, M. D., Beebe, D., Ammerman, R. T., Oppenheimer, S. G., Yeates, K. O., et al. (2008). Factors of biological risk and reserve associated with executive behaviors in children and adolescents with spina bifida myelomeningocele. *Child Neuropsychology*, *14*(2), 118–134.

Burmeister, R., Hannay, H. J., Copeland, K., Fletcher, J. F., Boudousquie, A., & Dennis, M. (2005). Attention problems and executive functions in children with spina bifida and hydrocephalus. *Child Neuropsychology*, *11*, 265–283.

Charney, E. (1992). Neural tube defects: Spina bifida and myelomeningocele. In M. Batshaw & Y. Perret (Eds.), *Children with disabilities: A medical primer, third edition* (pp. 471–488). Baltimore: Brookes Publishing Co.

Dennis, M., & Barnes, M. A. (2010). The cognitive phenotype of spina bifida meningomyelocele. *Developmental Disabilities Research Reviews*, *16*, 31–39.

Edmonds, L. D., & James, L. M. (1990). Temporal trends in the prevalence of congenital malformations at birth based on the birth defects monitoring program, United States, 1979–1987. *MMWR. Surveillance Summaries: Morbidity and Mortality Weekly Report. Surveillance Summaries/CDC*, *39*(4), 19–23.

Gioia, G. A., Isquith, P. K., Guy, S. C., & Kenworthy, L. (2000). *Behavior Rating Inventory of Executive Function*. Lutz, FL: Psychological Assessment Resources, Inc.

Gioia, G. A., Isquith, P. K., Retzlaff, P. D., & Espy, K. A. (2002). Confirmatory factor analysis of the Behavior Rating Inventory of Executive Function (BRIEF) in a clinical sample. *Child Neuropsychology*, *8*(4), 249–257.

Guy, S. C., Isquith, P. K., & Gioia, G. A. (2004). *Behavior Rating Inventory of Executive Function – Self-Report Version*. Lutz, FL: Psychological Assessment Resources, Inc.

Heffelfinger, A. K., Koop, J. I., Fastenau, P. S., Brei, T. J., Conant, L., Katzenstein, J., et al. (2008). The relationship of neuropsychological functioning to adaptation outcome in adolescents with spina bifida. *Journal of the International Neuropsychological Society*, *14*(5), 793–804.

Hughes, D. M., Turkstra, L. S., & Wulfeck, B. B. (2009). Parent and self-ratings of executive function in adolescents with specific language impairment. *International Journal of Language and Communication Disorders*, *44*(6), 901–916.

Hunt, G. M., & Oakeshott, P. (2003). Outcome in people with open spina bifida at age 35: Prospective community based cohort study. *British Medical Journal*, *326*(7403), 1365–1366.

Iddon, J. L., Morgan, D. J. R., Loveday, C., Sahakian, B. J., & Pickard, J. D. (2004). Neuropsychological profile of young adults with spina bifida with or without hydrocephalus. *Journal of Neurology, Neurosurgery, and Psychiatry*, *75*, 1112–1118.

Jandasek, B., Holmbeck, G. N., DeLucia, C., Zebracki, K., & Friedman, D. (2009). Trajectories of family processes across the adolescent transition in youth with spina bifida. *Journal of Family Psychology*, *23*(5), 726–738.

Jenkinson, M. D., Campbell, S., Hayhurst, C., Clark, S., Kandasamy, J., Lee, M. K., et al. (2011). Cognitive and functional outcome in spina bifida-Chiari II malformation. *Childs Nervous System*, *27*, 967–974.

Knouse, L. E., Bagwell, C. L., Barkley, R. A., & Murphy, K. R. (2005). Accuracy of self-evaluation in adults with ADHD: Evidence from a driving study. *Journal of Attention Disorders*, *8*(4), 221–234.

Mahone, E. M., Cirino, P. T., Cutting, L. E., Cerrone, P. M., Hagelthorn, K. M., Hiemenz, J. R., et al. (2002). Validity of the behavior rating inventory of executive function in children with ADHD and/or Tourette syndrome. *Archives of Clinical Neuropsychology*, *17*(7), 643–662.

Mahone, E. M., Zabel, T. A., Levey, E., Verda, M., & Kinsman, S. (2002). Parent and self-report ratings of executive function in adolescents with myelomeningocele and hydrocephalus. *Child Neuropsychology*, *8*, 258–270.

Northrup, H., & Volcik, K. A. (2000). Spina bifida and other neural tube defects. *Current Problems in Pediatrics*, *30*, 313–332.

Oakeshott, P., & Hunt, G. M. (2003). Long-term outcome in open spina bifida. *The British Journal of General Practice*, *53*(493), 632–636.

Rose, B. M., & Holmbeck, G. N. (2007). Attention and executive functions in adolescents with spina bifida. *Journal of Pediatric Psychology*, *32*, 983–994.

Roth, R. M., Isquith, P. K., & Gioia, G. A. (2005). *Behavior Rating Inventory of Executive Function – Adult Version*. Lutz, FL: Psychological Assessment Resources, Inc.

Slick, D. J., Lautzenhiser, A., Sherman, E. M., & Eyrl, K. (2006). Frequency of scale elevations and factor structure of the Behavior Rating Inventory of Executive Function (BRIEF) in children and adolescents with intractable epilepsy. *Child Neuropsychology*, *12*(3), 181–189.

Tarazi, R., Mahone, E. M., & Zabel, T. A. (2007). Self-care independence in children with neurological disorders: An interactional model of adaptive demands and executive dysfunction. *Rehabilitation Psychology*, *52*(2), 196–205.

Tarazi, R., Zabel, T. A., & Mahone, E. M. (2008). Age-related changes in executive function among children with spina bifida/hydrocephalus based on parent behavior ratings. *The Clinical Neuropsychologist*, *22*, 585–602.

Thibadeau, J. K., Alriksson-Schmidt, A. I., & Zabel, T. A. (2010). The National Spina Bifida Program Transition Initiative: The people, the plan, and the process. *Pediatric Clinics of North America*, *57*(4), 903–910.

Zukerman, J. M., Devine, K. A., & Holmbeck, G. N. (2010). Adolescent predictors of emerging adulthood milestones in youth with spina bifida. *Journal of Pediatric Psychology*, *36*, 265–276.