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Original Article

Psychosocial problems and cognitive functions in children with spina bifida

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ABSTRACT

Objectives: This study aims to assess psychosocial functioning in relation to lesion level and ambulatory status in children with spina bifida (SB) and compare them to their peers.

Patients and methods: Between March 2013 and May 2013, a total of 31 patients with SB (11 males, 20 females; mean age: 9.4 years; range, 6 to 14.7 years) and 36 typically developing peers (16 males, 20 females; mean age: 9.8 years; range, 6.5 to 14.8 years) were included in the study. All participants were assessed using a semi-structured psychiatric diagnostic interview via the Kiddie Schedule for Affective Disorders and Schizophrenia Present and Lifetime Version (K-SADS-PL), Wechsler Intelligence Scale for Children-Revised (WISC-R), Behavioral Rating Inventory of Executive Functions (BRIEF) parent form, Social Responsiveness Scale (SRS), and Aberrant Behavior Checklist (ABC).

Results: In the SB group, the rate of psychiatric disorders was significantly higher (p=0.001) and the SRS scores and the planning and organizational components of the executive function were higher than their peers (p=0.02 and p=0.007, respectively). The psychiatric diagnosis rate, BRIEF, and SRS total scores did not significantly differ according to lesion level and ambulatory status. The BRIEF initiate and organization of materials subtest scores and ABC scores were significantly lower at high lesion levels (p=0.02, p=0.02, and p=0.02, respectively) and non-community walkers (p=0.002, p=0.03, and p=0.003, respectively).

Conclusion: Psychiatric disorders, impairment in social responsiveness, and planning and organization components of the executive function are prevalent in children with SB with no intellectual disabilities, compared to their peers. Therefore, psychosocial counseling and multidisciplinary follow-up for SB patients seem to be beneficial.

Keywords: Children, cognitive impairment, psychiatric symptoms, psychosocial status, spina bifida.

Spina bifida (SB) is among the most common congenital anomalies which results in various types of physical impairments and complications related to neuroanatomical abnormalities.[1] The severity of SB varies in accordance with the spinal lesion level and neurological complications. The surgical interventions, early onset of therapies which preserve renal functions, and protective rehabilitative management have led to longer life expectancy. However, attention needs to be paid to this condition due to the significant impairment in psychological, behavioral, and social areas of life.[1,2]

Children with neurocognitive impairments are at an increased risk for emotional and behavior problems and psychiatric disorders.[3] Cognitive development in children with SB may be deprived not only by the disorder itself, but also by its neurological complications. Accompanying brain anomalies (e.g., Chiari malformation, agenesis or

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dysgenesis of the corpus callosum), hydrocephalus and its associated complications have been blamed to cause neurocognitive deficits such as attentional problems, memory difficulties, and poor executive function.[3] Executive functioning is an umbrella term which is usually used to describe cognitive abilities including goal-directed problem solving and adequate cognitive functions to adapt to the needs and changes in the environment. Executive functions represent the capability of processing data obtained from the observations of outer world, resulting in showing appropriate responses, when needed.[4,5] Despite their physical problems, their IQs fall within the average range.[6] Associated health complications include weakened or paralyzed lower extremities, urinary and bowel incontinence, hydrocephalus and orthopedic problems. Problems resulting from complications such as assistance need in mobilization, neurogenic bladder and bowel problems, precocious puberty and short stature related to endocrine dysfunctions may result in internalizing disorders, limited participation in activities with their peers and social skill deficits. Therefore, internalizing problems such as anxiety and depression are common among children with SB.[3,7]

Recently, there has been a surge in the literature investigating psychosocial, behavioral, and social adjustment in patients with SB. However, a limited number of studies has examined impairments in this area in relation to the lesion level and ambulatory status. To date, psychiatric diagnoses have been assessed via caregiver-reported outcomes, and no previous research has evaluated psychiatric symptomatology using a gold-standard psychiatric diagnostic interview. In the present study, therefore, we aimed to evaluate psychiatric symptomatology in children with SB using a gold-standard psychiatric diagnostic interview, to compare children with SB and their age peers in terms of psychiatric symptomatology, cognitive functioning, behavioral problems, executive functions and social responsiveness, and to assess whether psychiatric symptomatology, behavioral problems, executive functions, and social responsiveness varied in relation to the lesion level and ambulatory status.

PATIENTS AND METHODS

This cross-sectional, case-control study was conducted at Physical Medicine and Rehabilitation and Pediatric Nephrology outpatient clinics of Marmara University School of Medicine between March 2013 and May 2013. A total of 31 patients with SB (11 males, 20 females; mean age: 9.4 years; range, 6 to 14.7 years) and 36 typically developing peers as the control group (16 males, 20 females; mean age: 9.8 years; range, 6.5 to 14.8 years) were included in the study. Exclusion criteria for both groups were as follows: mental retardation according to clinical evaluation; chronic medical illness for the control group; having a history of long-lasting shunt dysfunction or existing shunt dysfunction; primary sensory loss and hand function deficits which could render assessments impossible; and having a history of psychosis or child neglect and abuse. Demographic and clinical data of the participants, including clinical history, musculoskeletal and neurological examinations were recorded. Clinical examinations were conducted by two experienced rehabilitation physicians. The International Myelodysplasia Study Group Criteria assigning motor levels was used for the evaluation of motor status of children with SB.[8] The Mid-lumbar and thoracic level lesions were classified as the upper lesion level, whilst the sacral and lumbosacral lesions were classified as the lower lesion level.[9] All participants and parents were informed about the nature of the study and a written informed consent was obtained. The study protocol was approved by the Marmara University School of Medicine Ethics Committee (No. 09.2013.0057). The study was conducted in accordance with the principles of the Declaration of Helsinki.

Based on the Hoffer criteria, children with SB were classified as community walkers and non-community walkers.[10] Typically developing children were recruited after a routine examination. The Wechsler Intelligence Scale for Children-Revised (WISC-R) and Kiddie Schedule for Affective Disorders and Schizophrenia Present and Lifetime Version (K-SADS-PL), a semi-structured evaluation procedure for clinical psychiatric diagnoses, were utilized by the child psychiatrist. The parents were asked to complete the Behavioral Rating Inventory of Executive Functions (BRIEF),[11] Social Responsiveness Scale (SRS),[12] and Aberrant Behavior Checklist (ABC).[13]

Kiddie Schedule for Affective Disorders and Schizophrenia Present and Lifetime Version (K-SADS-PL)

The K-SADS-PL is a semi-structured interview form for collecting signs and symptoms of psychiatric disorders in children and adolescents.[14] Validity and reliability studies of the Turkish version were carried out for diagnosing childhood psychiatric disorders.[15] The K-SASD-PL is considered the gold standard among diagnostic interview instruments

for clinical psychiatric diagnoses in children and adolescents.

The Wechsler Intelligence Scale for Children-Revised (WISC-R)

The WISC-R is used to measure the intellectual capacity in children with adequate speech and language skills. It has been adapted to Turkish culture and was standardized in Turkish.[16] In our study, we used the subtests of information, vocabulary, picture completion, and picture arrangement for comparison of the groups.

The Behavioral Rating Inventory of Executive Functions (BRIEF)

The BRIEF is a caregiver report rating scale developed to assess the behaviors related to executive functions of children ages 5 to 18 years.[11] The Parent Form contains 86 items of behavior as occurring "Never (1)," "Sometimes (2)," or "Often (3)." The BRIEF consists of eight clinical scales (Inhibit, Shift, Emotional Control, Initiate, Working Memory, Plan/Organize, Organization of Materials, Monitor), and two indexes (Behavioral Regulation Index [BRI] and the Metacognition Index [MI]). The BRI is comprised of three subscales (Inhibit, Shift, Emotional Control). The MI is comprised of five subscales (Initiate, Working Memory, Plan/Organize, Organization of Materials, and Monitor), and The Global Executive Composite is a summary score which incorporates all eight clinical scales. Higher scores indicate a greater impairment for each scale and index. Clinical validity has been supported with myelomeningocele study groups.[17] The scale was shown to be valid and reliable in Turkish children and adolescents.[18]

Social Responsiveness Scale (SRS)

The SRS is a parent-completed questionnaire exploring emotionally appropriate reciprocal social interaction and communication in children.[12] It consists of 65 items rated on a four-point scale from "Not true" to "Almost always true" by the parent based on the child's behavior over the past six months and generating one total score. Higher total scores indicate a greater severity of social impairment. The reliability and validity of the Turkish version were conducted and presented in the 19th National Child and Adolescent Psychiatry Congress in 2009.[19]

Aberrant behavior checklist (ABC)

The ABC has 58 items which are rated on a four-point scale ranging from 0 to 3.[13] The items are scored into five subscales as follows: *(i)* Irritability, Agitation, Crying, *(ii)* Lethargy, Social Withdrawal, *(iii)* Stereotypic Behavior, *(iv)* Hyperactivity, Noncompliance, and *(v)* Inappropriate Speech. The Turkish version was developed and the congruent and criterion validity of the ABC was investigated in a Turkish clinical sample.[20]

Childhood Autism Rating Scale (CARS)

The CARS has 15 items that are rated on a seven-point scale of behavioral observation ranging from 1 to 4 including intermediate values. The scale was developed as a tool to aid the diagnosis of Autism Spectrum Disorder (ASD) by Schopler.[21] It was adapted to Turkish language and the cut-off of the CARS was calculated as 29.5 for Turkish children.[22]

Statistical analysis

The sample size was calculated based on a previous study (the effect size [Cohen's d value] was found to 1.25 for MI score).[23] We calculated the sample size using the G*Power version 3.1 software (Heinrich-Heine-Universität Düsseldorf, Düsseldorf, Germany) and, accordingly, at least 25 patients per group were needed with an alpha coefficient of 0.05 and a power of 99% based on the t-test. Statistical analysis was performed using the PASW for Windows version 17.1 software (SPSS Inc., Chicago, IL, USA). The groups were compared in terms of demographic characteristics and psychiatric diagnosis using the chi-square tests for categorical variables and Mann-Whitney U test for continuous variables. Continuous variables were expressed in mean ± standard deviation (SD) or median (min-max), while categorical variables were expressed in number and frequency. A p value of <0.05 was considered statistically significant.

RESULTS

Baseline demographic and clinical characteristics of the children with SB and typically developing children are shown in Table 1. Both groups were similar with respect to age and sex distribution.

Table 2 shows the distribution of psychiatric diagnoses. Accordingly, 51.6% of the children with SB had at least one psychiatric diagnosis and 6.5% of them had two or more diagnosis. The most common psychiatric disorders were internalizing disorders and attention deficit hyperactivity disorder (ADHD). The rate of psychiatric disorders was significantly higher in

TABLE 1
Demographic and clinical characteristics of children with SB and TD children

	SB (n=31)					TD (n=36)					
	n	%	Mean±SD	Median	Min-Max	n	%	Mean±SD	Median	Min-Max	*p*
Age (year)			9.4±2.8	8.7	6-14.7			9.8±2.4	9	6.5-14.8	0.49
Sex											0.46
Male	11	35.5				16	44.4				
Female	20	64.5				20	55.6				
Lesion level											-
Sacral	10	14.9									
Lumbosacral	7	10.4									
Midlumbar	11	16.4									
Thoracic	3	4.5									
Hoffer ambulatory criteria											-
Community walker	12	17.9									
Household walker	8	11.9									
Exercise walker	7	10.4									
Nonwalker (nonambulator)	4	6									
Psychiatric diagnosis	16	51.6				5	13.9				**0.001**
WISC-R											
Verbal IQ											
Information			7.5±3.1	7.5	2-16			10.3±3.1	10.5	1-16	**0.001**
Vocabulary			10.4±2.8	10	6-19			11.9±2.0	12	7-16	**0.015**
Performance IQ											
Picture completion			9.5±3.1	10	1-15			9.0±3.2	11	6-16	**0.042**
Picture arrangement			10.3±3.0	9	2-17			10.3±3.0	11	5-18	0.11
ABC total			15.5±15.7	10	0-67			14±13.7	10	0-55	0.87
Hyperactivity			5.0±5.8	3.5	0-24			4.6±5.4	2.5	0-21	0.84
Irritability			5.6±6.3	3	0-28			5.5±6.0	3	0-23	0.91
Lethargy			2.7±2.9	1	0-9			2.5±2.9	2	0-13	0.82
Stereotypic behavior			2.3±3.0	1.5	0-11			1.3±2.2	0	0-9	0.13
BRIEF											
Behavioral Regulation Index			59.8±13.9	58	37-94			57.3±12.7	56	36-91	0.44
Inhibit			22.0±6.2	21	15-39			20.8±5.8	19.5	15-43	0.41
Shift			19.2±4.8	19	11-27			17.9±4.3	18	11-27	0.23
Emotional control			18.6±4.6	18	11-29			18.7±4.2	19	10-28	0.98
Metacognition Index			84.7±18.5	86	52-127			77.9±14.8	75.5	53-123	0.11
Initiate			14.5±3.5	15	8-21			13.6±2.7	13.5	8-19	0.26
Working memory			19.3±4.8	18	12-32			17.3±3.6	17	11-29	0.06
Plan/organize			25.3±6.7	25	14-39			21.4±4.8	21	14-37	**0.007**
Organization of materials			11.9±3.7	11	8-20			12.7±3.0	12	8-22	0.32
Monitor			13.7±3.3	14	8-21			12.9±3.2	12	8-20	0.3
Total			144.5±30.5	142	92-216			135.2±24.6	130	88-200	0.17
SRS			45.8±29.6	45	5-124			30.2±24.3	22	6-93	0.02

SB: Spina bifida; TD: Typically developing; SD: Standard deviation; Min: Minimum; Max: Maximum; WISC-R: Wechsler Intelligence Scale for Children-Revised; ABC: Aberrant Behavior Checklist; BRIEF: Child Behavioral Rating Inventory of Executive Functions; SRS: Social Responsiveness Scale; P-values in bold indicate statistical significance.

Table 2
Psychiatric diagnosis according to K-SADS-PL

	SB		TD		
	n	%	n	%	*p*
Attention-deficit/hyperactivity disorder	8	25.8	3	8.3	0.054
Internalizing disorders	9	29	1	2.8	**0.004**
Generalized anxiety disorder	3	9.7	1	2.8	0.23
Separation anxiety disorder	3	9.7	0	0	0.056
Social anxiety disorder	2	6.5	0	0	0.12
Specific phobia	2	6.5	1	2.8	0.46
Tic disorders	1	3.2	0	0	0.27

K-SADS-PL: Kiddie Schedule for Affective Disorders and Schizophrenia Present and Lifetime Version; SB: Spina bifida; TD: Typically developing children; P-values in bold indicate statistical significance.

TABLE 3
Comparison of outcomes according to spinal lesion level

	Low level (n=17)			High level (n=14)			
	Mean±SD	Median	Min-Max	Mean±SD	Median	Min-Max	*p*
WISC-R							
Verbal IQ							0.72
Information	7.5±3.0	7	2-16	7.6±3.4	8	3-14	0.8
Vocabulary	10.3±2.6	10	7-15	10.6±3.1	10	6-19	
Performance IQ							0.24
Picture completion	10.4±2.9	10.5	5-15	8.6±3.3	10	1-13	0.24
Picture arrangement	9.8±2.8	9.5	6-17	8.2±3.6	8.5	2-15	
ABC total	21.7±17.5	15.5	0-67	9.5±11.4	5	0-40	**0.02**
Hyperactivity	6.5±6	5	0-24	3.6±5.4	2	0-20	0.07
Irritability	8.5±7.6	5.5	0-28	2.7±2.9	2.5	0-9	**0.02**
Lethargy	4±3.4	3.5	0-9	1.2±1.5	1	0-4	**0.01**
Stereotypic behavior	2.7±3.3	2	0-11	2±2.9	1	0-9	0.42
BRIEF							
Behavioral Regulation Index	63.3±12.9	62	39-94	57.6±15.1	53.5	37-89	0.26
Inhibit	23.3±5.8	22	15-39	21.1±6.8	19.5	15-37	0.24
Shift	21±4.6	21	13-27	17.6±4.7	17.5	11-26	0.09
Emotional control	19.1±4.6	18	11-29	18.9±4.6	18.5	11-28	0.71
Metacognition Index	88±17.5	92	57-121	79.9±20.3	76.5	52-127	0.13
Initiate	15.6±3.3	16	10-21	12.9±3.4	12	8-21	**0.02**
Working memory	18.9±4.2	19	13-28	19.4±5.8	18	12-32	0.83
Plan/organize	26.7±7	26	17-39	23.9±6.8	23.5	14-37	0.19
Organization of materials	13.1±3.9	13	8-20	10.4±3.1	9	8-16	**0.02**
Monitor	13.7±3.6	15	8-19	13.4±3.2	13.5	8-21	0.5
Total	151.3±27.8	151	96-193	137.6±34.8	131.5	92-216	0.14
SRS	50.3±31.6	50	11-124	39.6±9.1	30.5	5-103	0.21

SD: Standard deviation; Min: Minimum; Max: Maximum; WISC-R: Wechsler Intelligence Scale for Children-Revised; ABC: Aberrant Behavior Checklist; BRIEF: Child Behavioral Rating Inventory of Executive Functions; SRS: Social Responsiveness Scale; P-values in bold indicate statistical significance.

the children with SB (p=0.001). The percentage of internalizing disorders rate was also significantly higher in SB group (p=0.004). The ADHD rate was higher in the SB group (25.8%; p=0.054). Information, vocabulary, picture completion subtests of WISC-R were lower in the SB group than the control group (p=0.001, p=0.015, and p=0.042, respectively). The WISC-R subtests did not significantly differ according to lesion level or ambulatory status (p>0.05) (Tables 3 and 4). There were no statistically significant differences between the children with SB and typically developing children in terms of the ABC scores (p>0.05). Stereotypic behavior subtest scores of the ABC did not significantly differ between the groups (p>0.05). Irritability and lethargy subtest scores of the ABC were significantly lower in the children with high level (p=0.02 and p=0.01, respectively), while hyperactivity, irritability, and lethargy subtest scores of the ABC were significantly lower in the non-community walkers, compared to the community walkers (p=0.03, p=0.003, and p=0.001, respectively) (Tables 3 and 4).

In addition, planning and organization subscale scores of the BRIEF were significantly higher in the SB group than the control group (p=0.007) (Table 1). The initiate and organization of materials subtest scores were significantly lower in the children with a high lesion level than those with a low lesion level (p=0.02) (Table 3). Also, the MI, initiate and organization of materials were significantly higher in children who were community walkers than the non-community walkers (p=0.02, p=0.002, and p=0.03, respectively) (Table 4).

All children scored below the CARS cut-off of 30 for ASD. The SRS total score was significantly higher in the children with SB than typically developing children (p=0.02) (Table 1). However, there was no statistically significant difference between either lower lesion level group and upper lesion level group, or the community walkers and non-community walkers (p>0.05) (Tables 3 and 4).

TABLE 4
Comparison of outcomes according to ambulatory status

	Non-community walker (n=19)			Community walker (n=12)			
	Mean±SD	Median	Min-Max	Mean±SD	Median	Min-Max	*p*
WISC-R							
Verbal IQ							
Information	8±3.5	8	3-16	6.7±2.3	7	2-10	0.36
Vocabulary	10.7±2.9	10	6-19	10.1±2.8	9	7-15	0.53
Performance IQ							
Picture completion	9.1±3.4	10	1-15	10.5±2.4	11	7-13	0.37
Picture arrangement	8.6±3.4	9	2-15	9.8±3.0	9	7-17	0.56
ABC total	9.6±10.8	6	0-40	27.6±7.7	25	7-67	**0.003**
Hyperactivity	3.6±5.0	2	0-20	8±6.5	6	0-24	**0.03**
Irritability	2.8±2.9	2	0-9	11.1±7.9	10	2-28	**0.003**
Lethargy	1.3±1.5	1	0-4	5.3±3.3	6	1-9	**0.001**
Stereotypic behavior	2.0±3.8	1	0-9	3.2±3.8	2	0-11	0.41
BRIEF							
Behavioral Regulation Index	57.7±1	55	37-89	66±13.1	62.5	45-94	0.25
Inhibit	21.5±6.4	21	15-37	23.6±6.2	22	15-39	0.45
Shift	18±4.9	18	11-27	21.9±3.9	21	14-27	0.08
Emotional control	18.2±4.4	18	11-28	20.5±4.7	19	13-29	0.56
Metacognition Index	78.9±19.0	80	52-127	93.9±15.6	93	66-121	**0.02**
Initiate	12.9±3.2	12	8-21	16.8±2.7	16.5	12-21	**0.002**
Working memory	18.4±5.4	18	12-32	20.7±3.7	21	16-28	0.07
Plan/organize	23.7±6.3	24	14-37	28.4±7.3	26.5	19-39	0.07
Organization of materials	11±3.7	10	8-19	13.3±3.7	12.5	9-20	**0.03**
Monitor	12.9±3.2	13	8-21	14.7±3.4	16	9-19	0.06
Total	139.6±32.0	134	92-216	159.9±25.7	155	111-193	**0.05**
SRS	41.7±28.9	32	5-103	51.7±33.6	49	12-124	0.32

SD: Standard deviation; Min: Minimum; Max: Maximum; WISC-R: Wechsler Intelligence Scale for Children-Revised; ABC: Aberrant Behavior Checklist; BRIEF: Child Behavioral Rating Inventory of Executive Functions; SRS: Social Responsiveness Scale; P-values in bold indicate statistical significance.

DISCUSSION

In the present study, children with SB and typically developing children were compared in terms of psychiatric symptomatology, cognitive functioning, behavioral problems, executive functions, and social responsiveness. Also, we investigated whether psychiatric symptomatology, cognitive functioning, behavioral problems, executive functions, and social responsiveness varied as a function of two variables in children with myelomeningocele: lesion level or community ambulatory status. Our study results showed that psychiatric pathology was more common among children with SB than typically developing children.

Similar to the results of the present study, children with SB were found to be at risk for exhibiting higher levels of internalizing symptoms (e.g., anxiety and depression), compared to their peers in previous studies.[1,3] In the present study, ADHD and internalizing disorders were detected as the prominent psychiatric diagnoses consistent with the previous literature.[3,4,24] Ammerman et al.[3] detected ADHD and internalizing disorders as the two most prevalent diagnostic categories using the Child Symptom Inventory (CSI). Burmeister et al.[25] also reported the ADHD prevalence as 31% in children with SB using the Swanson Nolan Achenbach Pelham-IV (SNAP IV). Fletcher et al.[24] reported impaired behavioral adjustment in children with SB using the Child Behavior Checklist (CBCL). However, the CSI, SNAP IV, and CBCL are all parent report screening tools with a lower diagnostic value. Different from the previous studies, in the present study, we examined the children using a semi-structured interview to reach a final diagnosis. Although the literature reports that children with SB have an elevated risk of depression,[26-29] interestingly none of the participants in the present study reached the diagnostic cut-off for major depressive disorder (MDD). A possible explanation can be that reaching the diagnostic cut-off for MDD is more unlikely using a semi-structural interview compared to a self- or parent-report rating scale. Therefore, it is likely that over-diagnosis is an issue with rating scales, in general.

Psychiatric evaluation is necessary for identifying psychopathology in children with SB.

In the current study, we also evaluated the intelligence levels and behavioral parameters related to executive functions. The three subtests of WISC-R including information, vocabulary, and picture completion were within the average range, but lower than the control group. Although two groups had no significant differences in most subtests of BRIEF, planning and organizing components of executive functions were scored higher in children with SB. These findings are in line with the previous literature.[4,7,30] Children with SB demonstrated typical patterns of cognitive deficits, such as difficulties in perceptual, visuomotor, mnemonic areas and impairments in executive functioning, including working memory and ability to inhibit response.[7,31-33] On the other hand, most of BRIEF subtest scores, MI, BRI and the total scores did not significantly differ between the two groups. This finding can be explained by the sample characteristics, as the children with SB were clinically followed by a multidisciplinary team in our study and, therefore, their care were most likely managed better than the general SB population. Also, it is important to note that presence of hydrocephalus is likely to have influenced the intellectual and neuropsychological functioning of children. Previously, it has been concluded that hydrocephalus rather than SB itself causes cognitive deficits and lower IQ in children with SB.[33] Similarly, children with hydrocephalus showed a poorer performance IQ than verbal IQ. However, this discrepancy was not observed in children with only SB (without hydrocephalus). Following the shunt operations, the prevalence of mental retardation in SB population has decreased.[30] In our study, we included only children with no intellectual disabilities without any history or presence of shunt dysfunction to compare the children with SB to their peers.

Furthermore, we assessed social responsiveness which involves elements of social cognition in this study. Previous reports have suggested that children with SB tend to have fewer social contacts and difficulties in unstructured social situations, more problems with social relationship and are more socially immature.[7,8] Attention and executive function are necessary for social interactions in children with SB.[7,8] In our study, the SRS total score was significantly higher in children with SB, compared to typically developing children, supporting the previous findings on social functioning of children with SB. Neurocognitive deficits may be associated with the social cognition deficits. In addition, posterior attention system plays a critical role in joint attention development, which is vital for social competence,[34] and it has been hypothesized that executive dysfunction in children with SB is related to posterior attention system;[35,36] therefore, problems related to posterior attention system may lead to social cognitive impairments in children with SB. Although none of the children were diagnosed as having ASD according to the CARS or clinical evaluation, as previously reported, social cognition abilities may be impaired. Even if children do not have autism, they should be supported in terms of social skills.

In the present study, outcomes in relation to lesion level and ambulatory status were also investigated. The results showed that the lesion level had no significant association with psychopathology or intellectual capacity as measured by the WISC-R. Similar to the findings, Holmbeck et al.[6] demonstrated that child psychosocial adjustment did not significantly differ in relation to lesion level. Similarly, Ammerman et al.[3] found no statistically significant difference between the lower and higher lesion groups and related to the ambulation status (i.e., no assistance, assistance, wheelchair) in terms of psychiatric symptomatology. Different from the previous studies, in the present study, the children were categorized according to their ambulatory status with the Hoffer criteria which evaluates ambulatory status in the context of the International Classification of Functioning. Children with a lower lesion level and children who are community ambulators demonstrated statistically more important impairments in the initiation and organization of materials subtests of BRIEF and showed more maladaptive behaviors in ABC.

In the study by Rose and Holmbeck.,[7] the spinal lesion level was not found to be a predictor of BRIEF performance. Ammerman et al.[3] found that ambulation status or lesion level had no relationship with the presence of ADHD. Supporting the study of Ammerman et al.,[3] Tarazi et al.[23] reported that the BRIEF MI and BRI scores were not significant related to the lesion level. In contrast, Burmeister et al.[25] noted significant differences in the lesion level and ambulation among the ADHD subtypes (I= inattentive type; hyperactive/impulsive type [ADHD-HI]; C= combined type). They detected that children with SB and ADHD-C were more likely

to have spinal lesions below the thoracic level. As a possible explanation of this result, the researchers suggested that, since children with sub-thoracic spinal lesions were more independent and ambulatory than others, combined type ADHD was more prevalent in this group. In the present study, the initiate and organization of materials subtests of BRIEF and hyperactivity, irritability and lethargy subtests of ABC were found to be lower in the non-community walkers. The common aspect of the aforementioned subtests is being related to movement. Despite being used in many of the previous studies, none of the psychiatric measurement tools have been validated in children with SB and they are not specifically developed for this population. From this point of view, they might have been not very useful in reflecting the exact status of children with SB. Since children with SB are at a high risk for poor neuropsychological and psychological outcomes, development of new SB specific tools is needed for appropriate treatment and diagnosis.

Nonetheless, the current study has several limitations. First, the cross-sectional nature of the study limits to learn how neuropsychological and behavioral problems may change over time. A longitudinal study would provide more useful information about psychosocial functioning. Second, the participants of the present study were selected from a group of children who were long followed by a multidisciplinary team. Therefore, it limits the generalization of results. Third, environmental factors such as socioeconomic status, family environment, and resilience factors such as coping skills could impact mental health; however, such factors were unable to be assessed in the present study.

On the other hand, to the best of our knowledge, this study is the first to assess psychiatric symptomatology using a gold-standard semi-structural interview, which appears as the main strength of the present study.

In conclusion, psychiatric disorders are more prevalent in children with SB and impairment in certain aspects of executive functioning appears to be associated with the lesion level and ambulatory status of children with SB. However, there is still a need for developing assessment tools which are sensitive for assessing neuropsychological functioning in this patient population.

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