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ORIGINAL PAPER

Upper limb cerebellar motor function in children with spina bifida

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Abstract
Purpose To investigate upper limb cerebellar motor function in children with spina bifida myelomeningocele (SBM) and in typically developing controls.
Methods Participants with SBM, who had either upper level spinal lesions (n=23) or lower level spinal lesions (n=65), and controls (n=37) completed four upper limb motor function tasks (posture, rebound, limb dysmetria, and diadochokinesis) under four different physical and cognitive challenge conditions. Functional independence was assessed by parental questionnaire.
Results Fewer SBM participants were able to complete the posture task, and they were less likely than controls to obtain a perfect rebound score. Participants with SBM showed impaired performance in either time, accuracy, or both, on the limb dysmetria and diadochokinesis tasks but responded like controls to physical and cognitive challenges.
Conclusions Because upper limb motor performance predicted aspects of functional independence, we conclude that upper limb impairments in children with SBM are significant and have direct implications for the level of independent functioning in children with SBM.

Keywords Spina bifida · Myelomeningocele · Upper limb motor function · Cerebellum · Functional independence

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Introduction

Spina bifida myelomeningocele (SBM) is the most common and severe form of spina bifida, a congenital disorder that occurs following failure of neural tube closure around 3–4 weeks gestation. Individuals with SBM have deficits in upper as well as lower limb control [1, 2]. In individuals with SBM, motor deficits lead to urinary and bowel dysfunctions and oftentimes limited unaided mobility [3].

Upper limb function is affected in the majority of children with SBM. Hand function is impaired on everyday tasks such as simulating eating, turning pages, stacking checker pieces, writing, card turning, lifting objects, and opening a jar [1, 4]. Hand coordination, dexterity, dotting speed, and muscle strength are all performed more poorly by individuals with SBM than by same age peers [5–7]. Children with SBM present with clumsiness, parasthesia, weakness, and numbness [8].

Severe impairment in children with SBM is also evident from studies of broader upper limb function. On a clinical neurological exam, which included a finger-nose-finger task and diadochokinesis, and muscle and hand function tasks, children with SBM demonstrated slow performance and unsystematic variability. Poor hand function was strongly correlated with hydrocephalus [9]. Hetherington and Dennis [2] tested motor function in children with hydrocephalus, many with SBM, and found impairments in fine motor skills and persistent motor control, as well as in balance, gait, posture, and upper and lower extremity strength. More recently, Dennis and collaborators [10] described upper limb function in young adults with SBM. Tests administered included rebound, posture, limb dysmetria, and diadochokinesis in each of four conditions: no challenge, physical challenge (closed eyes), cognitive challenge (counting backward from 50 concurrently), and combined physical and cognitive challenge (closed eyes and count backward from 50). Deficits in the group with SBM were apparent on all tasks, with fewer participants with SBM having perfect posture and rebound scores. The physical challenge had an adverse effect on limb dysmetria scores for both groups, whereas participants with SBM were more affected by the cognitive challenge. The cognitive challenge worsened performance on diadochokinesis in both groups [10].

The motor deficits described are related in part to the characteristic congenital dysmorphologies of the cerebellum in Chiari II malformation. The posterior fossa contents are crowded in Chiari II, with the cerebellar vermis shifted upwards and the inferior vermis herniated below the foramen magnum [11]. Hypoplasia of the cerebellar hemispheres has been identified along with reduced cerebellar volume [11–13]. Stretching of the brainstem inferiorly occurs with the mesencephalic tectum assuming a beak-like appearance [11]. The cerebellum is involved in motor control, visual feedback processing, and predicting the sensory consequences of motor acts [14], and as a result, many motor tasks including the maintenance of balance, gait, and posture are therefore impaired by its abnormalities.

Although dysmorphology of the cerebellum is characteristic in individuals with SBM and Chiari II, there is variation in the extent of the malformation that has been shown to be linked to eye movement function [15]. Specifically, Salman et al. [15] found an expanded midsagittal cerebellar vermis to be associated with normal eye movements, whereas a nonexpanded midsagittal vermis and smaller cerebellar volume were associated with abnormal eye movements. Performance on tasks that rely to a large degree on lateral parts of the cerebellum, by contrast, were proposed to be more impaired than on tasks relying on the vermis because the cerebellar hemispheres do not have the same opportunity for expansion within the confines of a small posterior fossa as the vermis does [15, 16].

Although several studies have described upper extremity function of individuals with SBM, previous studies with children have lacked a consistent coding scheme applied across a number of tasks, have used very specific tasks, have used broad scoring criteria, or have not included quantitative measures. The current study seeks to create a comprehensive quantitative overview of the upper limb motor deficits of children and adolescents with SBM, with a specific focus on tasks relying largely on the cerebellum [17], especially the cerebellar hemispheres [18], areas of the brain that seem to be significantly affected by the Chiari II malformation [11–13]. A consistent coding scheme was used across all tasks and was comprised of both qualitative and quantitative measures. Scoring was highly specific, with detailed criteria for how each level of performance should be scored. Our study attempts to quantify performance on tests of upper limb function in children and adolescents with SBM and includes physical and cognitive challenges much like those encountered by individuals with SBM in their daily lives. We also relate motor ability to medical history variables and current level of independent functioning.

Given the purported reliance on the lateral cerebellum for performance of the kinds of tasks included in this study [18] and the known reduction of the cerebellar hemispheres' volume in participants with SBM [11–13], we hypothesized that individuals with SBM would be significantly impaired on upper limb motor tasks and would be more affected than controls by physical and cognitive challenges. We further hypothesized that upper limb performance would be related to lesion level and that it would be a significant predictor of functional independence.

Method

Participants

In this study, participants were 88 children and adolescents with SBM, and 37 normal controls children recruited from clinics at two sites: The Hospital for Sick Children in Toronto (n=86) and the University of Texas Health Science Center-Houston (n=39). The study was approved by the ethics boards at each site, and the data were obtained in compliance with institutional regulations for human research. Participants ranged in age from 7.58 to 18.83 years old and had a Verbal or Performance IQ greater than 70. Table 1 presents IQ and demographic data for each group.

Medical information on the SBM participants was obtained from hospital charts or by parent report and included age, sex, handedness, type and level of spinal lesion, and number of shunt revisions. All children with SBM had been shunted for hydrocephalus shortly after birth. Of those children, 25 had no shunt revisions, 30 had one revision, 21 had two to four revisions, 12 had five to nine revisions, and no children had more than ten shunt revisions.

Participants were divided into upper spinal lesion (T12 and higher, n=23) and lower spinal lesion (L1 and lower; n=65) groups, according to current taxonomies [19, 20]. In terms of ambulatory status, 57 SBM participants were nonambulatory, 24 required assistive devices such as crutches, and 7 walked independently.

Tests

Participants completed a motor examination of upper limb function that was videotaped and subsequently analyzed. Each motor task was conducted under four different physical and cognitive challenge conditions: eyes open (no counting), eyes closed (no counting), eyes open and counting backward from 50, and eyes closed and counting backward from 50. Two observers independently watched the videotapes and scored the tasks, and any discrepancies between the first and second scoring were discussed and resolved. Time to perform tasks was recorded by the original examiner and was confirmed from the videotapes by the first and second scorers.

Posture Ambulatory participants were asked to stand next to a grid wall marked with 15-cm squares while holding their arms and hands outstretched and parallel, palms facing down for 30 s (full score) or until the hands dropped more than one square. Time taken to perform the task correctly was recorded, coded by duration on a scale of one to four, and the total posture score was the time sum over the four challenge conditions (minimum score 0, maximum score 16).

Rebound Participants were tapped on both arms, just above the wrist, at a 30^{o} angle to their outstretched, stationary, and parallel arms so as to slightly displace the arms. The task was performed twice under each of the four challenge conditions, and best performance was scored. Quality of arm return (e.g., normal, slow rebound, arms remain down), tone (e.g., normal, loose, floppy), presence of rebound, and overshooting were graded out of four for each arm, and the total rebound score was the sum of the four challenge conditions (minimum score 0, maximum score 128).

Limb dysmetria For this standardized finger-nose-finger test [21], participants held one arm straight out with the index finger pointed perpendicular to the arm. With the index finger of the other hand, they alternated between touching the tip of their nose and the tip of the extended finger for ten repetitions. The task was timed and scored

Table 1 Demographic information for control and upper and lower lesion SBM participants

Group		Control (n=37)	Lower lesion (n=65)	Upper lesion (n=23)
Age (years; mean±standard deviation)		12.47±2.60	12.65±2.98	12.72±3.00
Gender	Male	18 (48.65)	35 (53.85)	10 (43.48)
	Female	19 (51.35)	30 (46.15)	13 (56.52)
Ethnicity	Caucasian	26 (70.27)	51 (78.46)	20 (86.96)
	Hispanic	2 (5.41)	8 (12.31)	1 (4.35)
	Asian	5 (13.51)	2 (3.08)	1 (4.35)
	African-American	2 (5.41)	2 (3.08)	0 (0)
	Other	2 (5.41)	2 (3.08)	1 (4.35)
Socioeconomic status[a]		46.12±11.96	41.98±12.40	44.70±12.91
Stanford-Binet Composite IQ		108.6±9.91	91.0±13.90	82.0±9.84

Percentage is listed in brackets after each frequency

[a] Socioeconomic status was determined by the Hollingshead (1975) four-factor scale

separately under the four challenge conditions, and response measures were accuracy and time to complete the iterations. Accuracy was graded out of four for each arm in terms of the quality of arm movement (e.g., smooth, irregular, confused/jerky), steadiness of extended finger, accuracy in touching contact points, extent of elbow bend, and amount of indenting (minimum score 0, maximum score 40).

Diadochokinesis In this rapidly alternating hand movement test, participants placed one open hand palm down on a table and then turned it over to have the palm up for ten iterations per hand. The task was timed and scored separately under the four challenge conditions, and response measures were accuracy and time to complete the iterations. Accuracy was graded out of four for each arm in terms of the quality of arm movement (e.g., smooth, irregular, confused/jerky), tone (e.g., normal, loose, floppy), quality of rotation, rhythmicity, and completion of alternation (minimum score 0, maximum score 40).

Functional independence Independent functioning was assessed using the Scales of Independent Behavior-Revised (SIB-R), a structured interview that is normed by age and measures functional independence in four domains: motor, social/communication, personal living, and community living [22]. Parents of participants completed this interview.

Correlations among medical, demographic, and outcome variables were initially explored. Composite scores were obtained by summing performance over all conditions for all relevant tasks for both accuracy and reaction time. For the posture and rebound tasks, individuals were grouped according to whether they demonstrated perfect performance on the task, and group differences were analyzed using chi-square tests. For the limb dysmetria and diadochokinesis tasks, scores for each of the four challenge conditions were entered into analyses of variance to examine group differences. Regression analyses were used to identify whether upper limb motor skills were significant predictors of functional independence for nonambulatory participants over what is expected based on medical history variables.

Results

Intelligence

As expected, children with SBM had lower IQ than controls, $F(1, 123)=65.75$, $p<0.0001$, and the groups did not differ in age, socioeconomic status, gender, or ethnicity. More children with SBM ($n=22$) than controls ($n=2$) were left-hand dominant, $X^2(1)=6.45$, $p<0.05$. Children with upper level lesions had a lower IQ than those with lower level lesions, $F(1, 86)=8.17$, $p<0.01$, but there was no correlation between IQ and number of shunt revisions. In children with SBM, overall IQ was positively correlated with all four SIB-R domains, rs ranging from 0.37 to 0.47, ps<0.0005.

Posture

Of the 88 participants with SBM, only six ambulatory participants completed the posture task successfully. Statistical analyses are thus not appropriate, but descriptively, the participants with SBM who were able to complete the task did well with only one participant (as well as three controls) scoring slightly under perfect performance.

Rebound

Eighty participants with SBM completed the rebound task. There was a significant group difference with fewer participants with SBM than controls obtaining perfect scores, $X^2(1)=9.32$, $p<0.005$. Rebound scores in the SBM group were unrelated to the number of shunt revisions and did not differ as a function of lesion level.

Limb dysmetria

Sixty-nine participants with SBM completed the limb dysmetria task. Participants in the SBM group performed as accurately as controls on this task (see Table 2). The groups responded similarly to the physical and cognitive challenges, with best performance occurring with no challenge and worst performance occurring with the combined challenges of counting backwards with eyes closed, $F(1, 103)=17.34$, $p<0.0001$. Participants with SBM were slower than controls to complete this task in all challenge conditions, $F(1, 103)=9.13$, $p<0.005$. Counting backwards slowed performance for both groups, $F(1, 103)=9.82$, $p<0.005$, whereas closing the eyes had no effect on completion time.

Children with upper level lesions were as accurate as those with lower level lesions, but they did take longer to complete the task, $F(1, 67)=4.32$, $p<0.05$. Number of shunt revisions was not related to performance on this task.

Diadochokinesis

Seventy participants with SBM completed the diadochokinesis task. Participants with SBM were less accurate than controls on this task, $F(1, 102)=21.57$, $p<0.0001$ (see Table 2). Both groups were affected by the cognitive challenge of counting backwards, $F(1, 102)=72.47$, $p<0.0001$, and the physical challenge of closing the eyes, $F(1, 102)=12.15$, $p<0.001$, but these challenges did not have an interactive effect (i.e., there was no physical by cognitive

Table 2 Accuracy and time scores on two motor tasks (mean, (standard deviation))

Eyes	Accuracy				Time			
	Open		Closed		Open		Closed	
Counting	No	Yes	No	Yes	No	Yes	No	Yes
Limb dysmetria								
SBM	31.64 (4.56)	27.64 (4.63)	27.07 (4.45)	24.29 (5.06)	29.64* (9.68)	33.81* (13.92)	30.07* (9.53)	34.88* (15.33)
Controls	33.69 (4.99)	28.92 (5.63)	28.14 (4.79)	26.33 (4.74)	24.19 (8.34)	27.08 (10.34)	25.19 (9.89)	27.14 (10.66)
Diadochokinesis								
SBM	34.39* (3.24)	31.46* (3.84)	33.19* (4.04)	30.74* (4.06)	24.06* (6.76)	30.74* (13.47)	23.87* (7.88)	31.29* (13.02)
Controls	37.18 (2.66)	33.88 (3.45)	36.18 (3.22)	33.88 (2.94)	21.09 (5.41)	25.71 (8.86)	21.15 (5.35)	26.65 (9.61)

*$p<0.05$ (SBM participants significantly worse than controls)

challenge interaction). Participants with SBM took longer than controls to complete this task in all challenge conditions, $F(1, 102)=4.91$, $p<0.05$. Counting backwards slowed performance for both groups, $F(1, 102)=34.70$, $p<0.0001$, whereas closing the eyes did not increase completion time. There was no effect of lesion level on performance in terms of both accuracy and completion time, and the number of shunt revisions also was not related to performance.

Functional independence

SIB-R scores were available for 86 of the participants with SBM. All four SIB-R domains were positively correlated with rebound performance, $r(76)=0.36$, $p<0.005$; $r(76)=0.34$, $p<0.005$; $r(76)=0.45$, $p<0.0001$; and $r(76)=0.26$, $p<0.05$, for the motor, social/communication, personal living, and community living domains, respectively. Additionally, the motor score and the personal living score were associated with the limb dysmetria and diadochokinesis accuracy composite, $r(57)=0.33$, $p<0.05$ and $r(57)=0.31$, $p<0.05$, respectively. SIB-R scores were unrelated to number of shunt revisions, but the motor, personal living, and community living scores were lower in individuals with upper level spinal lesions, $F(1, 84)=34.89$, $p<0.0001$; $F(1, 84)=14.80$, $p<0.0002$; and $F(1, 84)=3.98$, $p<0.05$, respectively.

Regression analyses were performed on data from the nonambulatory participants with SBM to determine whether actual upper limb function was a more significant predictor of SIB-R scores than lesion level and number of shunt revisions. Only nonambulatory participants were included to increase homogeneity in functional independence scores. A performance composite score including data from the rebound, limb dysmetria, and diadochokinesis tasks was entered into the regression model along with lesion level and number of shunt revisions as predictors of each of the four SIB-R measures. This upper limb performance composite was found to be a better predictor than the medical history variables for the motor and personal living domains, accounting for 21% and 15% of the dependent variable variance, respectively, both ps<0.05. No other predictors were significant for any of the four domains.

Because rebound performance was significantly correlated with all four SIB-R domains, scores on this task also were entered into a regression model along with the medical history variables as predictors of functional independence. For the motor domain, lesion level was found to be the only significant predictor accounting for 21% of the variance, $p<0.001$. However, performance on the rebound task was the only significant predictor for the social/communication and personal living domains,

accounting for 8% and 21% of the variance, respectively, both $ps<0.05$.

Discussion

Children with SBM have impaired upper limb function relative to typically developing controls on tasks of posture, rebound, limb dysmetria, and diadochokinesis. The presence of upper limb deficits in children with SBM is consistent with recent reports of these deficits in young adults [10] and indicates that individuals with SBM experience upper limb motor challenges across the life span.

Children with SBM performed more poorly than controls on all four upper limb motor tasks. For limb dysmetria, children with SBM were as accurate as controls but at the cost of the time taken to perform the task. For the diadochokinesis task, children with SBM were both less accurate and slower in performing the task.

Real or ecological functionality involves being able to complete motor acts under conditions of challenge, such as one would typically encounter in everyday life. Although it is hard to simulate ecological challenges perfectly in a laboratory setting, the current study tested participants under conditions of physical (closed eyes) and cognitive (backwards counting) challenge in an attempt to investigate performance under conditions that approximate those in the real world. Although both the physical and cognitive challenges enhanced the difficulty of the limb dysmetria and diadochokinesis tasks and affected both time and accuracy of task performance, participants with SBM were not hindered disproportionately. In contrast, young adults with SBM are more affected than controls by the cognitive challenge on the limb dysmetria task [10]. Considered together, the data suggest that increased age in SBM involves disproportionate difficulty instantiating some basic physical tasks under the ecological demands of everyday life.

Number of previous shunt revisions did not affect upper limb motor performance. An interesting dichotomy is emerging in the literature in which various cognitive abilities, such as IQ and attention, do not appear to be related to the number of shunt revisions in children with SBM [23, 24] but cognitive abilities, such as IQ and memory, are negatively related to the number of shunt revisions in adults with SBM [25, 26]. However, motor skills including motor learning and performance are not related to the number of shunt revisions in either children or adults with SBM [10, 13] and may be more affected by the basic brain dysmorphology in SBM.

The effects of spinal lesion level on motor function remain somewhat unclear. Grimm [1] reported that a higher lesion level was associated with more impaired hand function in children with SBM, but Turner found no relation of hand function and lesion level [7, 8]. Dennis et al. [10] reported no effect of lesion level on upper limb motor performance in adults with SBM, but did find that individuals with a higher lesion level scored lower on a test of motor independence. In the current study, the only effect of lesion level on motor performance was that children with higher lesion levels took longer to perform the limb dysmetria task, despite attaining similar levels of accuracy to those with lower lesion levels. Further, children with higher lesion levels had lower scores on several of the functional independence measures. Although a higher spinal lesion level does seem related to problems in functional independence, these individuals were not overly disadvantaged on the upper limb motor tasks.

The lower limb motor difficulties that children with SBM experience are likely related to the presenting condition and surgical intervention that is usually performed shortly after birth to repair the myelomeningocele. It is now clear that upper limb cerebellar functions are compromised in these children as well, likely as a result of the brain dysmorphology characteristic of the Chiari II malformation. Furthermore, the amalgamated upper limb function score was a significant predictor of motor and personal living independence, and the rebound score specifically was a significant predictor of social/communication and personal living independence. Thus, upper limb function is an important contributor to functional independence in general.

A subgroup of children with SBM demonstrate abnormal eye movements [15], again indicating general motor difficulties in effectors other than the lower limbs. However, because processing of several classes of eye movements rely heavily on the cerebellar vermis, they may be normal in some children with SBM because of its ability to expand when compressed, even in the presence of reduced lateral cerebellar volume and smaller posterior fossa size. Children with an expanded midsagittal vermis typically have normal saccades, smooth ocular pursuit (in the absence of nystagmus), vestibulo-ocular reflex, and visual fixation capability [15]. By contrast, the upper limb motor tasks tested in the current study likely rely on the lateral cerebellar hemispheres, which cannot expand as the vermis can. The fact that children with SBM have impaired upper limb function is consistent with our expectations given what we know about cerebellar development in these individuals. However, further research is needed to clarify the specific relation between the upper limb tasks and brain development and whether variability in cerebellar hemisphere volume translates into differences in upper limb motor function.

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