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③ **SUPERTITLE**

Muscle strength, aerobic capacity and physical activity in independent ambulating children with lumbosacral spina bifida

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Abstract

Purpose. This cross-sectional study investigates deficits and associations in muscle strength, 6-minute walking distance (6MWD), aerobic capacity (VO_{2peak}), and physical activity (PA) in independent ambulatory children with lumbosacral spina bifida.

Method. Twenty-tree children participated (13 boys, 10 girls). Mean age (SD): 10.4 (± 3.1) years. Muscle strength (manual muscle testing and hand-held dynamometry), 6MWD, VO_{2peak} (maximal exercise test on a treadmill), and PA (quantity and energy expenditure [EE]), were measured and compared with aged-matched reference values.

Results. Strength of upper and lower extremity muscles, and VO_{2peak} were significantly lower compared to reference values. Mean Z-scores ranged from -1.2 to -2.9 for muscle strength, and from -1.7 to -4.1 for VO2peak. EE ranged from $73-84\%$ of predicted EE. 6MWD was significantly associated with muscle strength of hip abductors and foot dorsal flexors. VO_{2peak} was significantly associated with strength of hip flexors, hip abductors, knee extensors, foot dorsal flexors, and calf muscles.

Conclusions. These children have significantly reduced muscle strength, 6MWD, VO_{2peak} and lower levels of PA, compared to reference values. VO_{2peak} and 6MWD were significantly associated with muscle strength, especially with hip abductor and ankle muscles. Therefore, even in independent ambulating children training on endurance and muscle strength seems indicated.

Keywords: *Spina bifida, child, exercise test, physical fitness, muscle strength, performance*

Introduction

Over the last decade, few studies have reported on problems in young children with lumbosacral spina bifida (SB) [1–4]. There is increasing evidence that physical fitness in children with SB is impaired, even in those with lumbosacral lesions [2–4]. Several studies on aerobic capacity showed that oxygen cost of walking is significantly higher compared to healthy peers [2,3]. This is partly attributable to their altered gait pattern. Many compensatory movements are observed in the frontal and sagittal plane, particularly caused by muscle weakness in lower extremities [2,4].

There is very weak evidence that exercise training can improve muscle strength and physical fitness in children with SB [5]. At the same time, studies on determinants of physical fitness are scarce. This information is important for designing proper exercise programs to improve physical fitness in children who will benefit most. The purposes of the present study were two-fold. First, we investigated muscle strength, six-minute walking distance (6MWD), aerobic capacity, and physical activity (PA) in two groups of ambulatory children with lumbosacral SB. We compared the outcome of children with myelomeningocele (MMC), who mostly have hydrocephalus and Chiairi II malformation, to

those with lipomyelomeningocele (LMMC), where there is only involvement of the spinal cord. In addition, each group was compared with reference values of the normal population. Second, we investigated associations between muscle strength on the one hand and aerobic parameters and PA on the other.

Methods

Participants

The study group consisted of 23 children diagnosed with MMC or LMMC, from the SB clinic of the Wilhelmina University Children's Hospital, Utrecht, The Netherlands. Participants included those with paralysis level L5 or below, Intelligence Quotient (IQ) > 80, aged 6–18 years and being able to ambulate 500 m or more without crutches or para-walkers. Participants who had surgery less than six months prior to inclusion, and those with monoparesis or cerebral movement impairments were excluded, as were Non-Dutch speaking participants.

Thirty-three subjects met the inclusion criteria, 23 of them were willing to participate (13 boys and 10 girls). Their general characteristics are presented in Table I. The parents of 10 patients refused to participate. There were no differences in terms of gender, mean age, or ambulation level between the MMC and LMMC group.

All study procedures were approved by the University Medical Ethics Committee. Informed consent was obtained from the parents and the participants themselves when they were ≥12 years of age.

Measurements

All measurements were performed by the same experienced researchers for all participants.

Data concerning the presence of shunted hydrocephalus, mental status (IQ), and lesion level,

Table I. Patient characteristics.

	MMC $n = 16$	LMMC $n = 7$
Mean age (years) ($\pm$SD)	9.9 ($\pm$3.2)	11.6 ($\pm$2.7)
Ambulation		
• normal	10	7
• community	6	
Hydrocephalus		
• shunted	9	
• non-shunted	3	
• no hydrocephalus	4	7
Lesion level		
• L5-S1	7	3
• S1-S4	5	1
• No motor deficit	4	3

MMC, myelomeningocele; LMMC, lipomyelomeningocele; SD, standard deviation.

according to the ASIA criteria [6], were obtained from the medical records.

Ambulation level was defined according to Hoffer et al. [7]. The scoring was adapted for children with normal ambulatory skills. In Hoffer et al.'s description, participants with normal ambulatory skills are not distinguished from community walkers (walking outdoors with/without braces and possibly using a wheelchair for longer distance). This difference was considered clinically important, and we described participants who did not need a wheelchair at all, as 'normal ambulant' (community ambulator = + Ankle-Foot-Orthosis (AFO), + wheelchair (WC); normal ambulator = +/− AFO, − WC).

Anthropometrics

Body weight and height, were measured to calculate Body Mass Index (BMI), and compared with reference values for healthy subjects matched for age and sex. Z-scores were calculated [8]. Body composition was assessed using the sum of seven skin folds according to Pollack et al. [9]. The measurements were taken at the biceps, triceps, supra-iliacal, mid-abdominal, subscapular, medial thigh, and calf, at the right side of the body.

Muscle strength

Muscle strength in upper and lower extremities was graded 0–5 according to the standard Manual Muscle Testing (MMT) as described by Hislop [10]. As intertester reliability of MMT is poor (range in coefficients: 0.11–0.58) [11], all tests were performed by the same examiner. As MMT appears to be less specific for grade ≥4 [11], in addition for muscles ≥ 4, Hand-Held Dynamometer (HHD) (Citec, type CT 3001, CIT Techniques, Groningen, The Netherlands) was used to quantify isometric maximal muscle strength (in Newton). The tests were performed according to Beenakker et al. [12]. We tested: Shoulder abductors, wrist extensors, hip and knee flexors, hip abductors, hip and knee extensors, ankle dorsal flexors, and grip strength. The plantar flexors were not measured using HHD, as reference values were not available. Muscle strength was compared with Dutch reference values for healthy subjects matched for age and sex, obtained from Beenakker et al. [12]. HHD data were used for studying correlations, with the exception of plantar flexors. In these muscles only MMT data were available.

Six-minute walking distance (6MWD)

The 6-minute walking test was performed on an 8-meter track in a straight corridor as described

previously [13]. Participants were instructed to cover the largest possible distance in 6 minutes at a self-chosen walking speed. Encouragements were provided according to the ATS Guideline [14]. 6MWD and average walking speed were measured. 6MWD was compared with reference values of the normal population [13,15].

Aerobic capacity

After the 6-minute walking test, subjects performed a maximal exercise test using a treadmill (Enmill, Enraf, Delft, The Netherlands). Between the 6-minute walking test and the maximal exercise test, there was a recovery period of at least 15 min. After this period, all participants were able to start walking without tiredness. It is known from the literature that children recover significantly faster than adults from an exercise bout [16]. For both tests, participants were allowed to use their orthosis or orthopaedic shoes. In order to accommodate children of varying abilities, two progressive exercise test protocols were used. Children, who were community ambulant, were tested with a starting speed of 2 km/h that was gradually increased with a speed of 0.25 km/h every minute. Children who were normal ambulant, were tested with a starting speed of 3 km/hr that was increased with 0.50 km/h every minute. The protocols were continued until the participant voluntarily stopped due to exhaustion, despite verbal encouragement of the test leader. Maximal walk/run speed was recorded. During the maximal exercise test, physiologic responses were measured using a heart rate (HR) monitor (Polar) and calibrated mobile gas analysis system (Cortex Metamax B^3, Cortex Medical GmbH, Leipzig, Germany). The Cortex Metamax is a valid and reliable system for measuring ventilatory parameters during exercise [17].

Peak oxygen uptake (VO_{2peak}) was taken as the average value over the last 30 seconds during the maximal exercise test. Relative VO_{2peak} ($VO_{2peak/kg}$) was calculated as VO_{2peak} divided by body mass. Peak oxygen pulse was calculated as VO_{2peak}/HR_{peak}. Predicted VO_{2peak} values were obtained from established values from age- and sex-matched Dutch reference values [18].

Physical activity

A diary was used to estimate daily PA, both the quantity of activities, and energy expenditure (EE) according to Bouchard [19]. The diary included static activities (sleeping, lying, or sitting) as well as dynamic activities (transfers, walking, running, sporting, etc.). The quantity of PA was determined by mean daily hours of dynamic activities from the diary.

The term EE was used for the calculation of MET's according to Bouchard [19]. In the record, every 15-min period over three days, including a Saturday, was qualified in terms of energy costs on a 1 (=sleeping or resting in bed) to 9 scale (=high intensity sport activities or sport competition), corresponding to a range of 1.0 metabolic equivalent transformation (MET) to 7.8 METs and higher [19]. Approximate median energy cost for each of the nine categories in kcal/kg/15 min was used to compute the EE for each individual. The measured total EE was compared with predicted values of healthy peers adjusted for height, sex and age [20].

An estimation of daily EE for any of the days of the activity record appears quite reproducible [19]; the intraclass reliability correlations range from 0.86–0.95. Mean Kcal energy expenditure for the three days was highly reliable with a correlation coefficient of 0.96 ($p < 0.01$). It appears to be a reliable method in children ($r = 0.91$; $p < 0.01$ [18]) and has been used previously in children with MMC [20].

The diaries were filled out at the end of the day by the participants themselves, assisted by their parents.

Statistical analysis

Associations between muscle strength on the one hand, and 6MWD, aerobic parameters and PA on the other, were tested with Pearson's correlations. Alpha level was set at $p < 0.05$ for all analyses. T-tests were used to test differences between both groups (MMC vs. LMMC) and the normal reference values. If scores were skewed, nonparametric tests Mann-Whitney U-test were used (skin folds measurements, VO_{2peak}, grip strength, knee flexor and dorsal flexor strength). Statistical analyses were performed using SPSS for Windows (version 12.0, SPSS Inc, Chicago, Ill, USA).

Results

Anthropometrics

Results are shown in Table II. In both groups, body height was significantly lower compared to reference values (MMC $p = 0.03$; LMMC $p = 0.008$). BMI was only significantly higher in MMC group ($p = 0.03$) compared to healthy peers.

Muscle strength

We found no significant differences between participants in the MMC vs. LMMC group. Therefore, data are presented for the total group. As can be appreciated from Table III, the MMT indicated that in >25% of participants, muscle strength in the hip abductors and the

Table II. Anthropometric values.

	n	Mean (SD) SB patients	Mean (SD) controls	Z-score (SD)	p-value
Height (m)					
• MMC	16	1.37 (0.19)	1.45 (6.59)	−0.01 (0.01)	0.03*
• LMMC	7	1.47 (0.18)		−0.01 (0.01)	0.008**
Weight (kg)					
• MMC	16	35.7 (12.4)	36.2 (5.1)	0.5 (2.1)	0.32
• LMMC	7	41.7 (13.5)		0.1 (0.9)	0.74
BMI (kg/m^2)					
• MMC	16	18.1 (2.93)	16.6 (1.54)	1.5 (2.9)	0.03*
• LMMC	7	18.7 (3.1)		0.9 (1.4)	0.15
Sum of 7 skin folds					
• MMC	12	89.8 (38.5)	81.7 (31.2)	0.1 (1.6)	0.78
• LMMC	6	96.3 (57.0)		0.3 (1.8)	0.67

SD, standard deviation; m, meters; MMC, myelomeningocele; LMMC, lipomyelomeningocele; * = p value < 0.05, ** = p value < 0.01; kg, kilogram. Five patients refused measuring skin folds.

Table III. Muscle strength measured with manual muscle testing. The values indicates the number of participants (%) with a certain grade (0–5).

	Muscle grade					
Muscle group	0	1	2	3	4	5
Hip flexors	–	–	–	–	2 (8.7)	21 (91.3)
Hip abductors	–	–	1 (4.3)	5 (21.7)	1 (4.3)	16 (69.6)
Hip extensors	–	–	3 (13)	1 (4.3)	8 (34.8)	11 (47.8)
Knee extensors	–	–	–	–	1 (4.3)	22 (95.7)
Knee flexors	–	–	–	–	4 (17.4)	19 (82.6)
Dorsal flexors	–	–	1 (4.3)	4 (17.4)	3 (13)	15 (65.2)
Plantar flexors	3 (13)	1 (4.3)	1 (4.3)	1 (4.3)	3 (13)	14 (60.9)

plantar flexors were <3. Moreover, <50% of the participants had fully functioning hip extensors (grade 5).

Z-scores of muscle strength measured with HHD (Figure 1) were significantly reduced compared to reference values (all P values < 0.01) in lower as well as in upper extremities (mean range: −1.2 to −2.9). Even in the participants without paralysis when measured with MMT (grade 5), strength was significantly reduced in all muscle groups compared to healthy peers (mean range: −1.4 to −3.0).

Six-minute walking distance

The mean 6MWD was 353 m (SD ± 108) in the MMC group, compared to 424 m (SD ± 65) in the LMMC group (p = 0.07), and was significantly lower compared to reference values of healthy peers (664 metre SD ± 65.3; p = 0.03).

Aerobic capacity

VO$_{2peak}$, VO$_{2peak/kg}$ and HR$_{peak}$ in children with MMC and LMMC were significantly reduced compared to healthy peers (all p values < 0.05) (Figure 2), but not different between the MMC vs. LMMC group.

Maximal treadmill walk/run speed was significantly lower in the MMC group compared to the LMMC group, 6.3 (SD ± 2.1) vs. 8.4 (SD ± 1.7) respectively (p = 0.03).

Even in the seven participants without paralysis (4MMC, 3LMMC), aerobic capacity was impaired. Mean Z-score for VO$_{2peak}$ was: −2.0 (SD ± 0.8), Z-score for VO$_{2peak/kg}$: −2.7 (SD ± 1.4), and their HR$_{peak}$ was reduced as well (Z-score −3.5 [SD ± 2.3]).

Physical activity

There were no significant differences between the LMMC and MMC group in terms of PA. The mean daily hours of dynamic activities was 2.8 for the total group. A total of 57% used an active mode of transportation to school (35% bicycle, 22% walked). All others used an inactive mode of transportation (car or taxi).

Mean EE was significant lower in both groups compared to predicted normal EE; 5953 KJoule/day
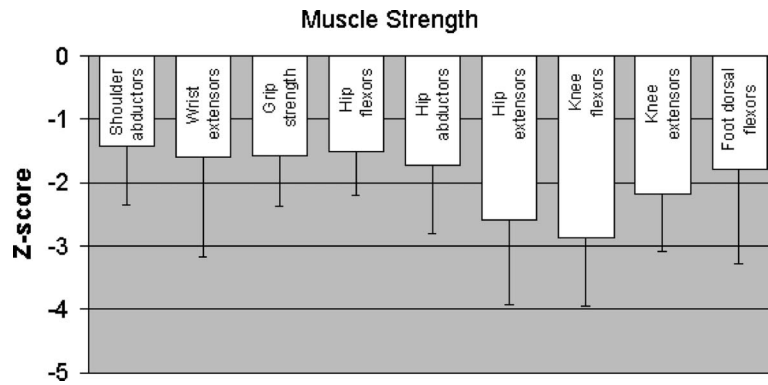
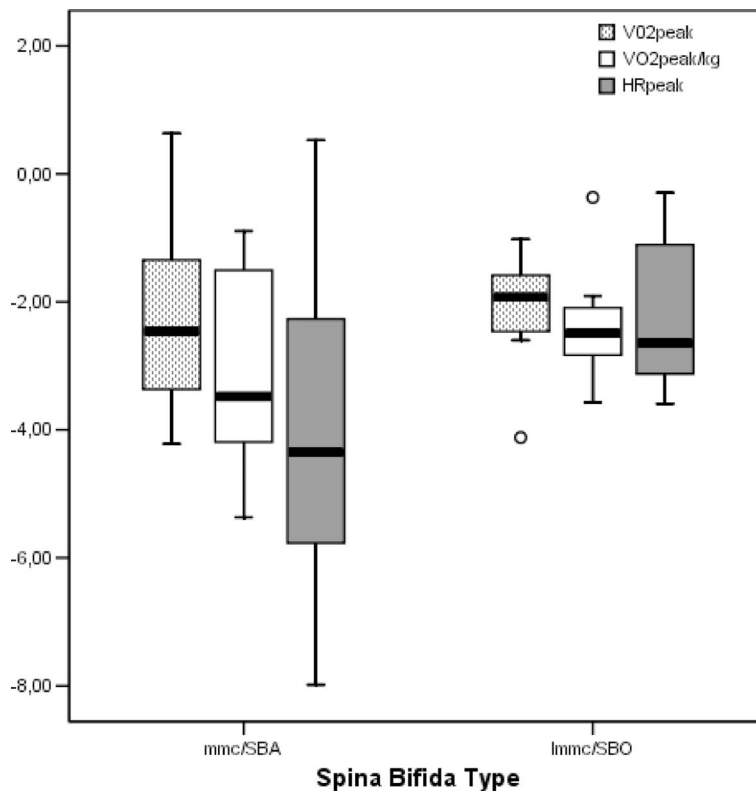Figure 1. Mean Z-scores (SD) of upper and lower extremity muscle strength.

Figure 2. Z-scores for maximal exercise capacity (VO$_{2peak}$, VO$_{2peak}$/kg and HR$_{peak}$) in subjects with MMC and LMMC.MMC, myelomeningocele; SBA, spina bifida aperta; LMMC, lipomyelomeningocele; SBO, spina bifida occulta; o, outlier.

(SD $\pm$ 1626) in the MMC group ($p = 0.009$) (is 73% $\pm$ SD 10 of predicted EE), and 7226 KJoule/day (SD $\pm$ 2260) in the LMMC group ($p = 0.03$) (is 84% $\pm$ SD 13% of predicted EE).

Correlations between muscle strength, 6MWD, aerobic capacity and PA

The correlations are presented in Tables IV and V. The 6MWD was significantly associated with muscle strength of hip abductors (0.47, $p = 0.04$) and foot dorsal flexors (0.55, $p = 0.02$), whereas peak tread-mill walking/running speed (V$_{peak}$) showed significant associations with the strength of plantar flexor muscles (0.59, $p = 0.01$). VO$_{2peak}$ was significantly associated with muscle strength of hip flexors ($r = 0.50$, $p = 0.02$), hip abductors ($r = 0.51$, $p = 0.03$), knee extensors ($r = 0.48$, $p = 0.03$), foot dorsal flexors ($r = 0.48$, $p = 0.048$), as well as with calf muscles ($r = 0.52$, $p = 0.049$). Vo$_{2peak/kg}$ was significantly associated with strength of plantar flexor muscles ($r = 0.44$, $p = 0.045$). Neither aerobic capacity, nor muscle strength were significantly correlated with PA.

Table IV. Correlations (*p* value) between muscle strength (hand-held dynamometry), 6-minute walking distance, and aerobic capacity.

	Muscle strength						
	Z-score Hip flexors	Z-score Hip abductors	Z-score Hip extensors	Z-score Knee extensors	Z-score Knee flexors	Z-score Dorsal flexors	Plantar flexors (MMT)
6MWD	0.36 (0.10)	0.47* (0.04)	0.05 (0.92)	0.15 (0.52)	0.37 (0.09)	0.55* (0.02)	0.40 (0.09)
Aerobic capacity							
• V_{peak}	0.24 (0.34)	0.16 (0.57)	0.04 (0.86)	0.09 (0.71)	0.34 (0.17)	0.38 (0.19)	0.59** (0.01)
• Z-score VO_{2peak}	0.50* (0.02)	0.51* (0.03)	0.24 (0.24)	0.48* (0.03)	0.42 (0.06)	0.48* (0.048)	0.52* (0.049)
• Z-score VO_{2peak}/kg	0.25 (0.27)	0.33 (0.17)	0.02 (0.95)	0.7 (0.77)	0.34 (0.14)	0.31 (0.22)	0.44** (0.045)

MMT, Manual Muscle Testing; 6MWD, 6-minute walking distance; Vpeak, maximum treadmill walk/running speed; * = *p* value < 0.05, ** = *p* value < 0.01. All correlations are expressed in Pearson correlation coefficients.

Table V. Correlations (*p* value) between muscle strength (hand-held dynamometry) and physical activity.

	Muscle strength						
	Z-score Hip flexors	Z-score Hip abductors	Z-score Hip extensors	Z-score Knee extensors	Z-score Knee flexors	Z-score Dorsal flexors	Plantar flexors (MMT)
Physical activity							
• Quantity	0.31 (0.90)	−0.13 (0.59)	−0.40 (0.10)	−0.03 (0.92)	−0.25 (0.31)	−0.15 (0.56)	−0.001 (0.99)
• Energy expenditure	−0.09 (0.74)	−0.02 (0.94)	−0.34 (0.20)	−0.02 (0.92)	−0.25 (0.31)	−0.15 (0.60)	−0.06 (0.80)

② MMT, Manual Muscle Testing; * = *p* value < 0.05, ** = *p* value < 0.01. All correlations are expressed in Pearson correlation coefficients.

Discussion

Our study shows that independent ambulating children with SB have significantly reduced muscle strength, 6MWD, aerobic capacity and lower levels of PA, compared to reference values. 6MWD and aerobic capacity were significantly associated with muscle strength, especially with hip abductor and ankle muscles.

In 26% of the participants the strength of hip abductor and calf muscles was below grade 4, resulting in a waddling or crouched gait, that might be energy consuming [3,4]. The reduced muscle strength in the lower extremities is in line with previous research [1,2,4]. Compared to healthy peers, we found significantly reduced strength in all lower extremity muscles, even in MMT grade 5. We observed significantly reduced muscle strength in the upper extremities. It is known that, in children with SB, the loss of muscle function below the actual level of the lesion directly affects motor performance [21]. This may impair daily functioning and influence general activity levels. The strength of intact, functional muscle groups may, according to our

data, is indirectly affected in this population as well. It might be due to deconditioning or to a certain defect in the neural drive of upper extremity muscles.

In all our participants maximal aerobic capacity was impaired, even in participants without paralysis. Since its relationship with muscle strength, aerobic capacity might be related to functional muscle mass of the patient. This has been previously suggested by Agre et al. [22]. They emphasized the importance of HHD measurements and recommended further studies to investigate the role of muscle mass in ambulatory children with SB [22].

Van den Berg-Emons et al. [23] found that adolescents and young adults with MMC (particularly non-ambulatory) were considerably hypoactive when compared to healthy peers. This was also observed in 13 to 20-year-old patients using the doubly-labelled water method [23]. Moreover, levels of objectively measured PA seemed to be significantly related to ambulation level and fitness [23]. In contrast to Van den Berg-Emons et al. [23], we found no significant associations between PA with any of the measured physical fitness parameters. This might be explained by the fact that the ambulation

level and the quantity of PA were homogenous within our participants.

Questions can be raised regarding measurement of VO_{2peak} and PA (quantity and EE) in this study, as protocols to measure VO_{2peak} and PA are not available for children with SB. Our results regarding PA (quantity and EE) should be interpreted with caution. Although an estimation of daily EE, from the activity record according to Bouchard et al. [19], appears to be a reliable method in children [19] and has been used previously in children with MMC [20], we might have underestimated the EE as the 'waddling' gait, as well as other PA which might be more energy consuming compared to healthy subjects.

There are no valid and reliable instruments for measuring PA and EE in children with SB, except for the very expensive doubly-labelled water method as previously used [24]. In future studies, accelerometers [25,26] in combination with heart-rate monitoring [27] and indirect calorimetry should be used, to estimate the intensity of daily activities more precisely.

We found aerobic capacity to be strongly related with muscle strength. Nonetheless, PA, quantity as well as EE, showed no correlations with physical fitness parameters. There is weak evidence that exercise training can improve fitness and strength in children with SB [5]. To our knowledge only one study in a heterogeneous group of eight participants exercising for 1 hour/week for 10 weeks showed improvements in muscle strength, exercise capacity, and self-concept [5]. Moreover, a low initial fitness level might provide a 'large room for improvement' for children with a chronic condition. Since there is a lack of clinical evidence, a rigorous designed exercise intervention trial in children with SB seems indicated.

It is our recommendation that prior to doing future research on improving physical fitness valid and reliable protocols for these children should be developed to measure cardio-respiratory fitness and PA.

① References

1. Schoenmakers MA, Gulmans VA, Gooskens RH, Helders PJ. Spina bifida at the sacral level: More than minor gait disturbances. Clin Rehabil 2004;18:178–185.
2. Bare A, Vankoski SJ, Dias L, Danduran M, Boas S. Independent ambulators with high sacral myelomeningocele: The relation between walking kinematics and energy consumption. Dev Med Child Neurol 2001;43:16–21.
3. Moore CA, Nejad B, Novak RA, Dias LS. Energy cost of walking in low lumbar myelomeningocele. J Pediat Ortho 2001;21:388–391.
4. Gutierrez EM, Bartonek A, Haglund-Akerlind Y, Saraste H. Characteristic gait kinematics in persons with lumbosacral myelomeningocele. Gait Posture 2003;18:170–177.
5. Andrade CK, Kramer J, Garber M, Longmuir P. Changes in self-concept, cardiovascular endurance and muscular strength of children with spina bifida aged 8 to 13 years in response to a 10-week physical-activity programme: A pilot study. Child Care Healt Dev 1991;17:183–196.
6. Manyard FM, Bracken MB, Creasy G, Ditunno JF, et al. International standards for neurological and functional classification of spinal cord injury. Spinal Cord 1997;35:266–274.
7. Hoffer MM, Feiwell E, Perry R, Perry J, Bonnett C. Functional ambulation in participants with myelomeningocele. J Bone Joint Surg Br 1973;55:137–148.
8. Gerver WJ, De Bruin R. Peadiatric Morphometrics. A Reference Manual. 2nd ed. Maastricht: Universitaire Pers Maastricht; 2001.
9. Pollack ML, Schmidt DH, Jackson AS. Measurement of cardio-respiratory fitness and body composition in the clinical setting. Compr Ther 1980;6:12–27.
10. Hislop HJ. Daniel's and Worthingham's muscle testing; techniques of manual examination. 6th ed. Toronto: WB Saunder Co; 1995.
11. Schwartz S, Cohen ME, Herbison GJ, Shah A. Relationship between two measures of upper extremity strength: Manual muscle test compared to hand-held myometry. Arch Phys Med Rehabil 1992;73:1063–1068.
12. Beenakker EA, Van der Hoeven JH, Fock JM, Maurits NM. Reference values of maximum isometric muscle force obtained in 270 children aged 4–16 years by hand-held dynamometry. Neuromuscul Disord 2001;11:441–446.
13. Paap E, Van der Net J, Helders PJ, Takken T. Physiologic response of the six-minute walk test in children with juvenile idiopathic arthritis. Arthritis Rheum 2005;53:351–356.
14. ATS. ATS statement: Guidelines for the six-minute walk test. Am J Respir Crit Care Med 2002;166:111–117.
15. Li AM, Yin J, Au JT, So HK, et al. Standard reference for the 6 minute walk test in healthy children aged to 16 yrs. Am J Respir Crit Care Med 2007;176:174–180.
16. Falk B, Dotan R. Child-adult differences in the recovery from high-intensity exercise. Exerc Sport Sci Rev 2006;34:107–112.
17. Brehm MA, Harlaar J, Groepenhof H. Validation of the portable VmaxST system for oxygen-uptake measurement. Gait Posture 2004;20:67–73.
18. Binkhorst RA, Van't Hof AM, Saris WH. Maximale inspanning door kinderen; referentiewaarden voor 6–18 jarige meisjes en jongens [Maximal exercise in children; reference values girls and boys, 6–18 year of age]. Den Haag: Nederlandse Hartstichting; 1991.
19. Bouchard C, Tremblay A, Leblanc C, Lortie G, Savard R, Theriault G. A method to assess energy expenditure in children and adults. Am J Clin Nutr 1983;37:461–467.
20. Littlewood RA, Trocki O, Shepherd RW, Shepherd K, Davies PS. Resting energy expenditure and body composition in children with myelomeningocele. Pediatr Rehabil 2003;6:31–37.
21. Coutts K, McKenzie D, Loock C, Beauchamp R, Armstrong R. Upper body exercise capacity in youth with spina bifida. Adapt Phys Activ Q 1993;10:22–28.
22. Agre JC, Findley TW, McNally MC, Habeck R, Leon AS, Stradel L, et al. Physical activity capacity in children with myelomeningocele. Arch Phys Med Rehabil 1987;68:372–377.
23. Van den Berg-Emons HJ, Bussmann JB, Meyerink HJ, Roebroeck ME, Stam HJ. Body fat, fitness and level of everyday physical activity in adolescents and young adults with meningomyelocele. J Rehabil Med 2003;35:271–275.

24. Bandini LG, Schoeller DA, Fukagawa NK, Wykes LJ, Dietz WH. Body composition and energy expenditure in adolescents with cerebral palsy or myelodysplasia. Pediatr Res 1991;29:70–77.

25. Van den Berg-Emons HJ, Bussmann JB, Brobbel AS, Roebroeck ME, Van Meeteren J, Stam HJ. Everyday physical activity in adolescents and young adults with meningomyelocele as measured with a novel activity monitor. J Pediatr 2001;139:880–886.

26. Bjornsen KF, Belza B. Ambulatory activity monitoring in youth: State of the art science. Pediatr Phys Ther 2004; 16:82–89.

27. Trost SG. Objective measurement of physical activity in youth: Current issues, future directions. Exerc Sport Sci Rev 2001;29:32–36.