ORIGINAL ARTICLE

Limiting factors in peak oxygen uptake and the relationship with functional ambulation in ambulating children with Spina Bifida

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Abstract The objective of this study is to interpret the outcomes of peak oxygen uptake (VO_{2peak}) in children with SB and explore the relationship between VO_{2peak} and functional ambulation using retrospective cross-sectional study. Twenty-three ambulating children with SB participated at Wilhelmina's Children's Hospital Utrecht, the Netherlands. VO_{2peak} was measured during a graded treadmill-test. Eschenbacher's and Maninna's algorithm was used to determine limiting factors in reaching low VO_{2peak} values. Energy expenditure during locomotion (both O_2 rate and O_2 cost) and percentage of VO_{2peak} and HR_{peak} were determined during a 6-min walking test (6MWT). Differences between community and normal ambulators were analyzed. VO_{2peak}, VO_{2peak}/kg, HR_{peak}, RER_{peak} and VE_{peak} were significantly lower compared to reference values, with significant differences between normal and community ambulators. Limiting factors

according to the algorithm were mostly "muscular and/or deconditioning" (47%) and ventilatory "gasexchange" (35%). Distance walked during 6MWT was 48.5% of predicted distance. Both O_2 rate and O_2 cost were high with significant differences between normal and community ambulators [17.6 vs. 21.9 ml/(kg min) and 0.27 vs 0.43 ml/(kg m)]. Also $\%HR_{peak}$ and $\%VO_{2peak}$ were significantly higher in community ambulators when compared to normal ambulators (resp. 97.6 vs. 75% and 90.2 vs. 55.9%). VO_{2peak} seems to be mostly limited by deconditioning and/or muscular components and possible ventilatory factors. For both peak values and functional ambulation, community ambulators were significantly more impaired than normal ambulators. High energy expenditure, $\%VO_{2peak}$ and $\%HR_{peak}$ reflect high level of strain during ambulation in the community ambulators. Future exercise testing in children with SB should include assessment of ventilatory reserve. Exercise training in ambulatory children should focus on increasing both VO_{2peak} and muscular endurance, as well as decreasing energy cost of locomotion.

Keywords Exercise test · Energy cost of locomotion · Spina Bifida · 6 min walking test · VO_{2peak}

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Introduction

Spina bifida (SB) is the most frequently seen congenital deformity of the neural tube, with an incidence ranging from 1.4 to 3.6 of every 1,000 in the Netherlands (RIVM 2006). The severity of these deficits is largely determined by both the type and level of lesion of the SB. In 80% of children with the more serious open type of SB (Spina Bifida Aperta), hydrocephalus and a Chiari II malformation—a malformation in the brainstem—are present (Hunt

and Poulton 1995). The level of lesion, classified according to the ASIA guidelines (Maynard et al. 1997) determines which muscles are being (partly) innervated. Besides this medical classification, children are functionally classified using the Hoffer classification (Hoffer et al. 1973), recently adapted by Schoenmakers et al. (2005), presented in Table 1. About 20% of the lesions occur at the sacral level, enabling them to be, in most cases, community or normal ambulators. Despite high levels of functioning, these patients still experience difficulties in performing both dynamic motor skills and activities of daily living (Schoenmakers et al. 2004). This could be an important factor in inducing a cycle of less ability resulting in less activity, further reducing physical fitness and ambulation. In adolescence a large number of children seem to become wheelchair dependent as ambulation becomes too strenuous (Findley et al. 1987). Studies indeed have shown children and young adults with SB to be less active with reduced levels of physical fitness compared to their healthy peers (Agre et al. 1987; Steele et al. 1996; van den Berg-Emons et al. 2003a, b). Based on these results, van den Berg-Emons et al. (2003a, b) concluded that programs aimed at regular physical exercise and daily physical activity should be started in childhood to prevent further decline in physical fitness and daily functioning.

At the same time, small studies have shown higher levels of energy expenditure during ambulation in patients with SB, a finding that is associated with a pathological gait pattern (Bare et al. 2001; Buffart et al. 2006; Waters and Mulroy 1999). Energy expenditure refers to both O_2 rate [ml O_2/(kg min)], an indicator of strain or effort and O_2 cost [ml O_2/(kg m)] (Schwartz 2007). Higher energy expenditure during ambulation may result in higher levels of fatigue while physically active. This combination of reduced exercise capacity and higher cost of locomotion is

referred to as "diminished physiological fitness reserve" (McArdle et al. 1996).

In our SB clinic, 23 ambulatory children with SB were seen for sports and lifestyle advice. Results showed low levels of overall muscle strength, exercise capacity and daily physical activity (Schoenmakers et al. 2008). While designing an exercise program specifically aimed at improving both endurance and ambulation in ambulatory children with SB, the following questions were raised:

1. Which factors (cardiovascular, pulmonary or muscular) are limiting VO_{2peak} in ambulatory children with SB?
2. Since components of physical fitness are associated with efficiency of movement (Bar-Or 1996), is VO_{2peak} related to oxygen expenditure during ambulation in children with SB?

Methods

Subjects

The study group consisted of a 23 ambulatory children with SB visiting the SB clinic of the University Children's Hospital in Utrecht (The Netherlands) for lifestyle and sports advice in 2004. Study procedures were approved by the University Medical Ethics Committee.

Children were included when they were (1) at least community ambulatory (see Table 1), (2) able to follow instructions regarding testing and (3) between 6 and 18 years of age. Parents and children signed informed consent prior to testing.

Exclusion criteria were medical events that might interfere with the outcomes of the testing and medical status that did not allow maximum exercise testing.

Study design

Retrospective cross-sectional study, using outcomes of incremental exercise testing and the 6-min walking test (6MWT).

Demographics

Data concerning medical history were obtained from medical records. These data included type of SB, level of lesion, use of orthotics, ambulation level, age and sex.

Body mass index (BMI)

BMI was calculated as weight (kg)/length (m)^2. This index has proven to be a reliable and valid tool to estimate

Table 1 Ambulation level by Hoffer et al. adapted by Schoenmakers et al. (2005)

Level of ambulation	Description
Normal ambulation	Independent and unrestricted ambulation without use of assisted devices
Community ambulation	Independent outdoor ambulation with or without use of braces and/or assisted devices; using wheelchair for longer distances
Household ambulation	Using braces or assisted devices for indoor ambulation; using wheelchair for outdoor locomotion
Non-functional ambulation	Walking only in therapeutic situations
Non-ambulation	Wheelchair dependent

children's nutritional status, e.g. whether they are over- or underweight (Dietz and Robinson 1998; Mei et al. 2002). Weight was measured using an electronic scale. Height was measured while standing using a wall-mounted centimeter.

Peak oxygen uptake (VO_{2peak})

In exercise testing, $VO_{2\ peak}$ is considered to be the single best indicator of aerobic exercise capacity, which is often referred to as aerobic fitness. Gas exchange analysis during an incremental ergometry test to the point of volitional termination due to exhaustion is considered the gold standard to measure VO_{2peak} (Shephard et al. 1968). Earlier studies employing exercise testing in healthy children show it is possible to test VO_{2peak} in healthy children (Armstrong et al. 1996; Eiberg et al. 2005; Gulmans et al. 1997; Reybrouck et al. 1992; Rowland 1993). In this study, VO_{2peak} was measured using a treadmill test (EnMill, Enraf, Delft, The Netherlands), since all children would be able to perform this test and reference values are available for both young children and adolescents. In previous studies, treadmill protocols have been used to test VO_{2peak} in children with disability (Hoofwijk et al. 1995; Verschuren et al. 2006), including children with Spina Bifida (Agre et al. 1987; Shermans et al. 1997). In order to accommodate children with different ambulatory abilities, two progressive exercise test protocols were used. Children considered community ambulators were tested with a starting speed of 2 km/h, which was gradually increased by 0.25 km/h every minute. Children classified as normal ambulators were started at a speed of 3 km/h, with the speed being increased 0.50 km/h every minute. The protocols were continued until the patient stopped due to exhaustion, despite verbal encouragement of the test leader. During the incremental 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 gas-exchange parameters during exercise (Brehm et al. 2004; Medbo et al. 2002).

Functional ambulation

Functional ambulation was measured during a 6-min walking test (6MWT). The test was performed on an 8-m track in a straight corridor and gas exchange parameters were measured continuously with a portable Cortex gas analysis system (Cortex Metamax B^3, Cortex Medical GmbH, Leipzig, Germany). Patients were instructed to cover the largest possible distance in 6 min at a self-selected walking speed. The test and encouragements during the test were performed in accordance with the ATS guidelines (ATS 2002).

Data analysis

Peak oxygen uptake

Peak exercise parameters were calculated as the average value over the last 30 s during the exercise test. Normalized VO_{2peak} was calculated as VO_{2peak}/kg. Predicted peak values were obtained from established values from age- and sex- matched historical Dutch controls (Binkhorst et al. 1991). For comparison with healthy children Z-scores were calculated for VO_{2peak}, VO_{2peak}/kg, HR_{peak} RER_{peak} and VE_{peak}. Standard deviation scores >2 SD below or above normal were considered to be significantly different from the norm values.

Data were evaluated using the algorithm from Eschenbacher and Maninna (Eschenbacher and Mannina 1990). This algorithm has been developed for the interpretation of outcomes of exercise testing in adults. For this purpose it uses cut-off points routinely measured during exercise testing in order to make a distinction between cardiac, pulmonary or "other" limitations (deconditioning and/or musculoskeletal factors) to explain exercise capacity. The following parameters were measured and used in the algorithm: VO_{2peak}, VCO_{2peak}, VE_{peak}, VE_{peak}/VCO_{2peak}, anaerobic threshold (AT) and heart rate response (HRR) expressed as $(HR_{peak} - HR_{rest})/(VO_{2peak} - VO_{2rest})$. Whereas the original algorithm uses adult cut-off points, in this study values were adapted to the pediatric population, based on earlier work from our laboratory regarding maximum exercise testing parameters in healthy children (van Leeuwen et al. 2004).

In this algorithm, VE_{peak}/VCO_{2peak} is a general indicator of the efficiency of the both the lungs and gas exchange; high values (VE_{peak}/VCO_{2peak} >36) suggest gas exchange difficulties. Since FEV_1 was not measured and no known pulmonary problems were present, it was assumed that ventilatory reserve in all children was normal. HRR refers to the increase in HR in relation to the increase in VO_2. An excessive increase in HR ($>[-6.25 \times age] + 150$) might reflect either cardiac disease or deconditioning. In our patients no known cardiac history was present and therefore increased HRR was considered an indicator of deconditioning rather than cardiac disease. AT occurring at less than 40% VO_{2peak} was considered an indicator of poor circulatory or "pump" limitation. When no ventilatory or cardiac limitations were present, patients were considered to be limited by "other limitations".

Functional ambulation

The following functional parameters were measured based on the 6MWT: (1) 6-min walking distance (6MWD) and percentage of predicted 6MWD. Predicted 6MWD was calculated using the formula of Li et al. (2007), based on heart rate increase and sex (see Eqs. 1 and 2);(2) O_2 rate as uptake per minute [ml/(kg min)];

$$554.16 + ((HR_{6min} - HR_{rest}) \times 1.76) + (\text{height (cm)} \times 1.23) \text{ Boys} \quad (1)$$

$$526.79 + ((HR_{6min} - HR_{rest}) \times 1.66) + (\text{height (cm)} \times 0.62) \text{ Girls} \quad (2)$$

Steady state was taken as the average value over the period during which oxygen uptake changed less than 5% (Schwartz 2007); (3) Subsequently, the following parameters were derived: Speed (m/min), calculated as 6MWD/6; O_2 cost [ml/(kg m)], calculated as O_2 rate/speed (Waters and Mulroy 1999); individual strain ($O_2 rate_{steady\ state}/VO_{2peak} \times 100\%$ and maximum $HR_{6mwt}/HR_{peak} \times 100\%$)

For all measurements, t tests were used to test differences between normal and community ambulators after testing for normal distribution and equality of means. When this was not the case (VO_{2peak}), Mann–Whitney U test were used. For the correlation between VO_{2peak} and O_2 expenditure, a Spearman Rho was calculated. Significance level was set at $p < 0.05$. Statistical analyses were performed using SPSS for Windows (version 15.0, SPSS Inc, Chicago, Ill.).

Results

Population

The study population consisted of 23 children (13 boys/11 girls, age 6–17) with either SB aperta ($n = 16$) or SB occulta ($n = 7$). Children's age, height, weight and BMI are described in Table 2. The level of lesion, classified according to the ASIA guidelines (Maynard et al. 1997) and the ambulation level are presented in Table 3.

Peak oxygen uptake and other peak parameters

Out of 23 children, 21 performed the treadmill test without any significant problems. Two did not participate due to anxiety ($n = 1$) or pain during ambulation ($n = 1$). VO_{2peak}, VO_{2peak}/kg, HR_{peak}, VE_{peak}, VCO_{2peak} and RER_{peak} and their Z scores are presented in Table 4.

One tailed t test showed a significant difference between the normal and community ambulators for Z scores of

Table 2 Subjects characteristics

	Mean (SD)	Z scores (SD) compared to reference values
Age (years)	10.4 (3.1)	
Height (m)	1.4 (0.18)	−0.09
Weight (kg)	37.5 (12.7)	0.4
BMI (kg/m^2)	18.4 (2.9)	1.4

Table 3 Level of lesion and functional ambulation level

	Number (%)
Level of lesion	
L5–S1	10 (43.5)
S1–S4	6 (26.1)
No motor loss	7 (30.4)
Ambulation level	
Normal ambulator	17 (74)
Community ambulator	6 (26)
Use of orthotics	13 (56)

VO_{2peak}, VO_{2peak}/kg, HR_{peak}, VE_{peak} scores and outcomes of VCO_{2peak} and VO_{2peak}/kg. Normal ambulators showed higher scores for both ventilation, VO_{2peak} and VCO_{2peak}. Ventilatory equivalents for both carbon dioxide and oxygen (VE_{peak}/VCO_{2peak} and VE_{peak}/VO_{2peak}) did not differ, but were high in both groups.

Limiting factors using Eschenbacher's and Mannina's algorithm

Eighty-five percent of complete test data ($n = 20$) scored below 90% of predicted VO_{2peak}, indicating lower levels of fitness than expected.

Looking at the limiting factors, 47% showed signs of "other limitation", e.g. muscular deficiency and/or deconditioning, as indicated by high heart rate response (HRR).

Thirty-five percent showed possible signs of insufficient gas exchange at the pulmonary level, as indicated by $VE_{peak}/VCO_{2peak} > 36$ Table 5.

Functional ambulation

Ambulation parameters from 22 children were analyzed. Results are shown in Table 6 and Figs. 1 and 2. A steady state was reached in 20 children after the 2 or 3 min of walking in both groups. A difference between normal and community ambulators is clear throughout the 6MWT, with significant differences starting during minute 2.

Table 4 Descriptives of exercise testing in 21 children with SB

	Mean (SD) all children	Z score (SD)	Mean (SD) NA	Z scores (SD)	Mean (SD) CA	Z scores (SD)
VO_{2peak} (l/min)	1.28 (0.57)	−2.2[a]	1.39 (0.58)	−1.9	0.85 (0.24)	−3.6[a], *
VO_{2peak}/kg [ml/(kg min)]	33.14	−2.9[a]	34.77	−2.5[a]	26.2*	−4.5[a], *
HR_{peak} (beats/min)	172.2 (21.2)	−3.4[a]	175.5 (20.8)	−3.0[a]	158.5 (19.1)	−5.0[a], *
$V'E_{peak}$ (l/min)	45.6 (19.1)	−2.4[a]	49.3 (19.3)*	−2.0	30.0 (7.0)	−3.8[a], *
VCO_{2peak} (l/min)	1.30 (0.63)	NA	1.42 (0.65)	NA	0.82 (0.24)*	NA
RER_{peak} (VCO_2/VO_2)	1.00 (0.13)	−2.1[a]	1.01 (0.14)	−2.0	0.97 (0.2)	−2.4[a]

All Z scores are in SD compared to reference values

NA normal ambulator, *CA* community ambulator

* $p < 0.05$ between normal and community ambulators

[a] Z scores > −2SD

Table 5 Cut-off points in the algorithm by Eschenbacher and Maninna as measured in 21 children with SB

	Mean	Range	Used cut off points	Indicative for	% reaching critical values
VO_{2peak} pred (%)	79.6	51–120	<90%	Low VO_{2peak}	(85%)
VE/VCO_{2peak}	36.6	27.6–52.7	>36	Ventilatory limitations	(35%)
HRR	88.5	−0.33–182.7	>(−6.25 × age) + 150	Deconditioning and/or muscular limitations	(47%)
AT%	72.3	35.8–98.86	<44	Cardiovascular limitations	(6%)

VE/VCO_2, HRR and AT% are being shown for those VO_{2peak} pred <90% ($n = 18$)

HRR heart rate reserve, *AT* anaerobic threshold

Table 6 Outcomes of 6MWT

	Mean (SD) all children	Mean (SD) normal ambulators	Mean (SD) community ambulators
6MWD (m)	391.4 (61)	408.5 (57.2)	333.4 (30.6)*
Predicted distance (%)	48.5 (8.3)	50.2 (8.3)	41.1 (2.7)*
Mean O_2 rate [ml/(kg min)]	18.5 (3.9)	17.6 (3.3)	21.9 (4.8)*
O_2 rate $_{steady state}$ [(ml/(kg min)]	20.0 (3.9)	19.3 (3.5)	22.9 (4.3)
$HR_{max6MWT}$	134 (18)	129 (15)	150 (14)*
O_2 cost [(ml/(kg m)]	0.3 (0.09)	0.27 (0.06)	0.43 (0.07)*
% VO_{2peak}[a]	63.1 (20)	55.9 (14.3)	90.2 (7.6)*
%HR_{peak}	80 (14.8)	75 (11.6)	97.6 (11.6)*
Speed (km/h)	3.9 (0.6)	4.1 (0.6)	3.3 (0.3)*

* $p < 0.05$ between normal and community ambulators

[a] measured during steady state

Average distance walked was 391.4 (±61) m, which was 48.5% of predicted distance. Significant differences were seen between the community and normal ambulators regarding distance ($p < 0.01$), %predicted distance ($p < 0.05$), speed ($p < 0.01$), oxygen rate ($p < 0.05$), oxygen cost ($p < 0.0005$), maximum HR reached during 6MWT ($p < 0.015$), %VO_{2peak} ($p < 0.0001$) and %HR_{peak} ($p < 0.04$). Figures 1 and 2 show O_2 rate and %VO_2 during the 6MWT. Percentage VO_{2peak} during the 6MWT averaged 63.1% (±20%), with significant differences between normal and community ambulators (55.9 vs. 90.2%). Similar differences were seen for %HR_{peak}, respectively 75.0 versus 97.6% for normal versus community ambulators. Both O_2

rate and O_2 cost were high with significant differences between normal and community ambulators [17.6 vs. 21.9 ml/(kg min) and 0.27 vs. 0.43 ml/(kg m)]. The community ambulators walked more slowly and thus covered less distance, while performing the task at a much higher percentage of their maximum capacity and at the same time requiring more energy during locomotion.

Correlation between VO_{2peak} and oxygen utilization during ambulation

Both O_2 rate and O_2 cost during 6MWT correlated negatively with VO_{2peak} (respectively $r_{sp} = -0.56$ ($p < 0.001$)
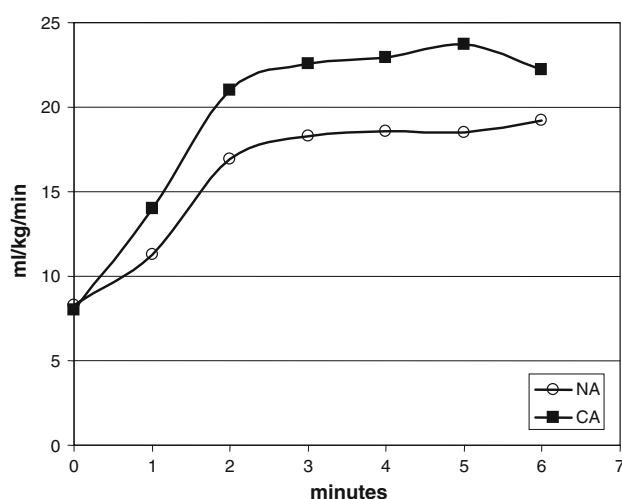
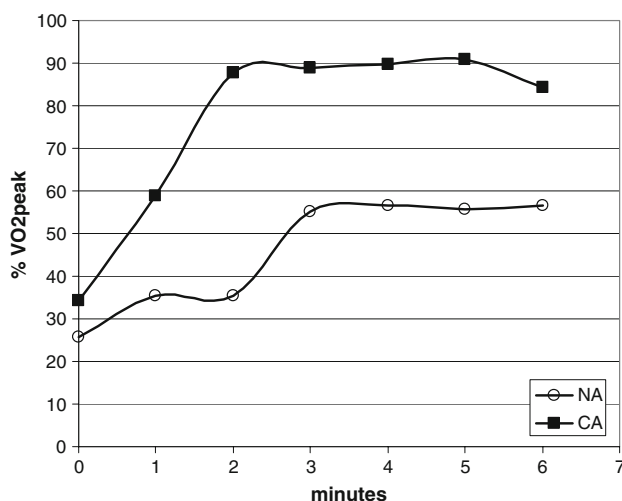**Fig. 1** O_2 rate during 6MWT comparing normal and community ambulators

Fig. 2 $\%VO_{2peak}$ during 6MWT comparing normal and community ambulators

and $r_{sp} = -0.76$ ($p < 0.001$)), indicating lower energy expenditure during locomotion in children reaching higher VO_{2peak}.

Discussion

Peak oxygen uptake

The first purpose of the study was to determine why ambulatory children with SB showed significantly reduced levels of VO_{2peak} compared to healthy peers. Despite subjective signs of peak effort, the treadmill testing resulted in both low HR_{peak} and RER_{peak} values, calling into question the true maximal character of the testing procedures.

Currently we are using a protocol of Rossiter et al. (2006) using a supra-maximal step (105–110% of last reached speed) to determine whether the regular protocol yields true maximum values or whether a different approach in exercise testing should be used in children with SB. At the same time the low HR_{peak} seems to be in line with other studies regarding exercise testing in children with SB (Agre et al. 1987; Shermans et al. 1997) with HR_{peak} ranging from 130 to 185 beats per minute. In one of these studies (Agre et al. 1987) HR_{peak} seems negatively related to level of lesion. Another explanation for low HR_{peak} could be that exercise capacity in children with SB is not limited by the cardiovascular system, but by muscular components.

Using the algorithm (Eschenbacher and Mannina 1990), limiting factors in exercise capacity in ambulatory children with SB indeed seem to be mostly "deconditioning and/or muscular" components. Muscular limitations can partly be explained by the disease itself. Lower muscle mass in children with SB results in lower VO_{2peak}. VO_{2peak}/kg was even more reduced. This is likely a result of the different body composition in these children with less active muscle mass and increased fat mass (Liusuwan et al. 2007b). In contrast with the muscular consequences of the disease, atrophy and muscular inefficiency as a result of disuse and sedentary lifestyle can be expected to improve through a training program. In the studied population, not only was exercise capacity reduced, they also showed lower strength above the lesion level and a sedentary lifestyle (Schoen-makers et al. 2008). In combination with a high HRR, the muscular limitations indentified in this study seem partly due to deconditioning. Exercise programs aimed at improving physical fitness in ambulatory children with SB are scarce, but they do show significant improvements in both strength and endurance (Andrade et al. 1991; Liusu-wan et al. 2007a; Widman et al. 2006).

In this study high ventilatory equivalents were found for both VCO_2 and VO_2, which could be an indicator of ventilatory limitations. These findings are consistent with one other study reporting on pulmonary dysfunction due to restrictive lung disease and respiratory muscle weakness in children with SB during exercise (Shermans et al. 1997).

Another possible explanation for these high ventilatory equivalents might be the presence of a Chiari II malformation, often present in children with SB. This malformation affects the brain stem and is known to influence both O_2 and CO_2 peripheral chemoreceptor function in children with SB (Gozal et al. 1995; Petersen et al. 1995). Future studies should include assessment of ventilatory reserve during exercise, so that a distinction can be been made between the different causes of pulmonary limitations, e.g. diffusion type versus gas exchange versus mechanical limitations.

Functional ambulation

Our second purpose was to determine the relationship between VO_{2peak} and functional ambulation. The 6MWT is a submaximal test of functional exercise capacity (Li et al. 2007). One of the main outcomes is the distance walked. In our study only half of the predicted distance was reached. A likely reason for reduced 6MWD is reduced muscle strength resulting in decreased motor control and an altered gait pattern leading to high energy expenditure (Gutierrez et al. 2005; Waters and Mulroy 1999). In this study, both O_2 rate and O_2 cost were indeed high compared to values for healthy children, which range in the literature from 12 to 18 ml/(kg min) and 0.18 to 0.27 ml/(kg m). In the literature values for O_2 rate and O_2 cost during ambulation in children with SB range from 17.5 to 19 ml/(kg min) and from 0.28 to 0.47 ml/(kg m) (Bartonek et al. 2002; Gutierrez et al. 2005). The O_2 rate in our study was higher, while the cost ranged from 0.27 ml/(kg min) in normal ambulators to 0.43 ml/(kg min) in community ambulators. These other studies used different protocols regarding both mode of testing (treadmill vs. level ground), walking speed (self-selected vs. imposed) and preparation of the subjects, factors hinderomg comparison between the studies. Therefore we are using new protocols, proposed in recent literature (Brehm et al. 2007; Schwartz 2007) in our current study.

Another interesting result from this study is the individual level of strain during locomotion. Community ambulators showed a much higher HR during the 6MWT than normal ambulators. Studies in healthy children show HR reached during the 6MWT to be around 134 beats per minute, which is similar to the normal ambulators (Lammers et al. 2007; Li et al. 2005). Looking at the intensity of ambulation, community ambulators performed this task at a very high level of individual strain (91% VO_{2peak} and 97.6% HR_{peak}). Studies in healthy children reach 65–70% of HR_{peak} during the 6MWT (Paap et al. 2005). One explanation for this could be the low HR_{peak} reached during the exercise test. But as mentioned above the community ambulators reached a high HR during the 6MWT. In combination with high oxygen utilization, the effort of walking remains high.

Correlation between VO_{2peak} and oxygen utilization during ambulation

VO_{2peak} values were related to oxygen utilization during locomotion. This indicates lower levels of peak oxygen uptake are associated with both higher oxygen rate and oxygen cost during locomotion. Rate is indicator of strain or effort, which might explain the level of fatigue during locomotion. Reybrouck et al observed similar trends in treadmill testing when comparing children with chronic fatigue with healthy peers, concluding that high individual strain was associated with early fatigue (Reybrouck et al. 2007).

Energy cost is an indicator of efficiency (Schwartz 2007). From the literature it is known that energy cost of locomotion in people with disabilities can be improved by training (Felici et al. 1997; Protas et al. 2001), so future research should also look at the effects of training to reduce the energy cost during locomotion in ambulatory children with SB.

Limitations of the study

Questions could be raised with regards to use of a treadmill protocol. Other studies (Bruinings et al. 2007; van den Berg-Emons et al. 2003a, b; Widman et al. 2007) have used upper extremity ergometry. An advantage of arm ergometry in this population could be that the muscles tested are less involved in the disease process. In this way the outcomes of the test might more closely reflect cardiorespiratory limitations in exercise testing. On the other hand, upper extremity ergometry has been known to result in lower VO_{2peak} values, due to the smaller muscle mass being involved in testing (Franklin 1985). In this study a treadmill protocol was chosen for several reasons. First, for all children, ambulation was the main mode of transportation. In this case it is recommended to use a treadmill for maximum exercise testing, due to the specificity of testing (Stromme et al. 1977). Secondly, we were interested in comparing outcomes from the peak exercise test to other ambulation parameters. If we had used arm ergometry, these comparisons would be hard to interpret due to the differences in physiological responses between arm ergometry and treadmill testing. In this study the 6MWT was performed using an 8-m track. Looking at the studies establishing reference values (Geiger et al. 2007; Lammers et al. 2007; Li et al. 2007) for the 6MWD in children, a 20 m track seems to result in a longer 6MWD. In this study however, the children were walking with a significantly lower speed as compared to healthy children reducing the importance of the shorter distance between the turning points. Despite this shorter track a steady state of O_2 utilization was reached by all, but two children. Currently we are working with the more commonly used 20 m track.

Conclusion

Lower levels of VO_{2peak} in ambulatory children with SB seem to be related to muscular and/or deconditioning components rather than cardiopulmonary deficiencies. Future exercise testing in ambulatory children with SB should include evaluation of the ventilatory reserve to better determine possible ventilatory limitations.

Both O_2 rate and O_2 cost during locomotion are high in ambulatory children with SB, even in those considered to be normal ambulators. Oxygen utilization correlated negatively with VO_{2peak}. Overall, community ambulators showed significantly worse outcomes than normal ambulators. At the same time, the normal ambulators only walked half of the predicted distance at high O_2 rate and O_2 cost. Future training programs for ambulatory children with SB should focus on improving VO_{2peak} and muscular endurance, as well as decreasing energy expenditure during locomotion to increase physiological reserve.

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