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Randomized Controlled Study of Home-Based Treadmill Training for Ambulatory Children With Spina Bifida

Janke F. de Groot, MSc[1,2], Tim Takken, PhD[2], Marco van Brussel, PhD[2], Rob Gooskens, PhD[2,3], Marja Schoenmakers, PhD[2], Chantal Versteeg[1], Luc Vanhees, PhD[1,4], and Paul Helders, PhD[2]

Abstract

Background. Many ambulatory children with spina bifida (SB) decline in their walking despite stable or even improved motor function. *Objective*. The authors evaluated the effects of a home-based treadmill training program on both ambulatory function and aerobic fitness. *Methods*. This randomized clinical trial of 34 ambulatory children with SB allocated 18 to supervised treadmill training for 12 weeks at home and 14 to usual care. Patients in home training exercised twice a week at an intensity of 66% of HR_{peak} (peak heart rate) and gradually progressed from 70% to 140% of their individual walking speed. Main outcome measures were obtained at baseline, after intervention, and 3 months postintervention. Ambulation was measured using the 6-minute walk test (6MWT), during which gross energy consumption (ECS_{gross}) and energy cost were calculated. Maximal exercise capacity was measured using an incremental treadmill test. Both VO_{2peak} and $speed_{peak}$ were recorded as outcome parameters. *Results*. After training, significant changes were seen between the groups for 6MWT (P = .002; d = 1.08), $speed_{peak}$ (P = .001; d = 1.14), VO_{2peak} (P = .034; d = 0.78), and ECS_{gross} (P = .004; d = 1.01). Long-term effects were recorded for 6MWT (P = .003; η = 0.34), $speed_{peak}$ (P = .003; η = 0.35), and ECS_{gross} (P = .014; η = 0.29) but not for VO_{2peak}. *Conclusion*. A home-based, progressive treadmill training program for ambulatory children with SB has a large long-term effect on ambulation, with a moderate short-term effect on VO_{2peak}.

Keywords

treadmill training, ambulation, Spina Bifida

Introduction

Spina bifida (SB) is the most frequently seen congenital deformity of the neural tube, with an incidence ranging from 2 to 8 per 1000 live births worldwide.[1] As a result of the neural tube deformity, patients experience a variety of disturbances in cognition, motor function, sensory function, and bowel and bladder function.[2] The severity of these disturbances is largely determined by both the type and level of lesion of the SB. As a result of advances in the medical approach, mortality rates have decreased, and 60% to 80% of children with SB live to be adults.[3-6]

Children with lower lumbar lesions may experience difficulties in performing both dynamic motor skills and activities of daily living.[7] In the adolescent years, a large number of children seem to become wheelchair dependent as ambulation becomes too strenuous.[8] Looking at the prognosis and development of SB, a 25-year cohort study[3] found a decline in ambulation frequency as the main mode of locomotion from 95% at age 0 to 5 years to 46% at age 20 to 25 years, despite stable or even improving motor lesions. Ambulation in adulthood correlated with lesion level, with 93% of patients with a sacral level ambulating, and 91% with L5 and 57% with L4 motor levels ambulating. Ambulation level during teenage years predicted that of young adults. This group of children could greatly benefit from a new approach to maintain walking. Earlier studies have shown higher levels of energy expenditure during ambulation

[1]University of Applied Sciences, Utrecht, the Netherlands
[2]Wilhelmina Children's Hospital, University Medical Center, Utrecht, the Netherlands
[3]Rudolf Magnus Institute of Neuroscience, University Medical Center, Utrecht, the Netherlands
[4]Catholic University of Leuven, Leuven, Belgium

Corresponding Author:
Janke F. de Groot, Child Development and Exercise Center, Wilhelmina Children's Hospital, University Medical Center Utrecht, Room kb.02.056.0. P.O. Box 85090, 3508 AB Utrecht, The Netherlands
Email: Janke.degroot@hu.nl

in patients with SB, which are associated with a pathological gait pattern because of muscle weakness in the lower extremities.[9,10] Higher energy expenditure may result in fatigue and reduced levels of aerobic fitness.[11-13]

Reduced exercise capacity and higher energy cost of locomotion is referred to as diminished physiological fitness reserve.[14] We have shown the diminished reserve relationship of VO_{2peak} and oxygen cost of locomotion for those with SB.[15] Both VO_{2peak} and energy cost or efficiency of movement can be improved by training,[16,17] which is the focus of this interventional study.

For ambulatory children with SB, only 1 small exercise study has been conducted with positive results on muscle strength and ambulation.[18] In children with other chronic disorders, such programs have been implemented on a larger scale with positive effects on fitness and daily participation.[19-21] Looking more specifically at programs aimed to improve ambulation, treadmill training has shown promising results in other pediatric populations,[22] but there are some reservations because of methodological questions regarding study design and description of intensity and frequency of the intervention.[23] From a motor learning point of view and specificity of training effects, treadmill training is a task-specific training that allows repetitive practice of gait cycles, important for motor learning and neural plasticity.[24,25] Therefore, the purpose of this study was to evaluate the effects of an individualized treadmill training program aimed at improving both aerobic fitness and ambulation, compared with usual care, in ambulatory children and adolescents with SB.

Methods

Study Design

A randomized clinical trial (RCT) was performed with 1 arm receiving a training intervention together with their regular care and a control group without training intervention but still receiving regular care. The primary researcher and data entry assistants were blinded to group allocation. Participants were randomly allocated to the intervention or control group. Randomization was stratified according to age (older and younger than 12 years of age) and sex. For each stratum, random numbers 1 to 41 were assigned to the participants, which were put into envelopes; it was determined randomly whether the even or odd number would enter the intervention group. Participants were assigned to the study by opening the envelopes. The different steps in this process were administered by different research assistants who were blinded to the other processes. Because of the logistics involved in transportation of the treadmill and assignment of the therapists, the randomization happened prior to the first measurement. To prevent interfering with the children's motivation and performance during testing, participants were informed of their assignment after the first measurement.

Participants

The study group consisted of 41 ambulatory children with SB, recruited both through the SB outpatient clinic of the Wilhelmina Children's Hospital, University Medical Center in Utrecht, the Netherlands, and the parent organization for families with neurological diseases (the BOSK). For inclusion and exclusion criteria see Figure 1.

Measurements were taken at the Child Development and Exercise Center in the hospital and at BIO Kinderrevalidatie research center, whereas supervised training took place at home. All study procedures were approved by the University Medical Ethics Committee. Children were included when they were (1) at least community ambulatory classified by Hoffer[26,27] (see Table 1), (2) able to follow instructions regarding testing and training, and (3) between 6 and 18 years of age.

Parents and children signed informed-consent forms prior to testing. Exclusion criteria were medical events that might interfere with the outcomes of the testing and/or medical status that did not allow maximum exercise testing.

Measurements

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

Measures of Physical Fitness. According to Bouchard and Stephard,[28] physical fitness consists of a combination of morphological, muscular, and cardiorespiratory components rather than 1 single measure. Although aerobic fitness or peak oxygen uptake (VO_{2peak}) was the primary outcome measure in this domain, the following components were measured as well.

Weight, height, and BMI. Weight was measured using an electronic scale. Height was measured while standing using a wall-mounted centimeter scale. Body mass index (BMI in kg/m^2) was derived from weight and height. *Z* scores were calculated from Dutch growth charts.

Subcutaneous fat assessment. In addition to BMI, fat percentage was assessed as the sum of the following 7 skin folds, according to Pollock et al[29]: triceps, biceps, subscapular, suprailiac, midabdominal, quadriceps, and calf. Skinfold measurement provides an estimate of the total amount of subcutaneous fat. In adults, several regression equations have been used to calculate fat percentage, but the validity of these equations in children (with SB) has not yet been established. Therefore, in line with other research,

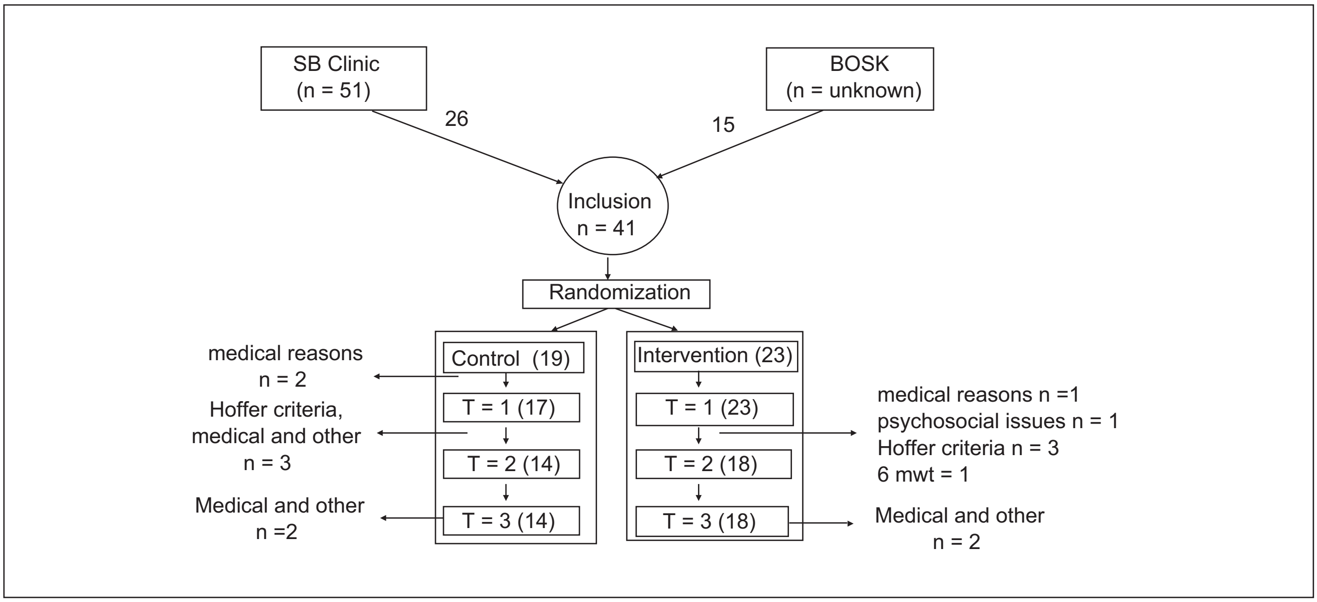

Figure 1. Inclusion, exclusion, and randomization of the USAGE study. Abbreviations: SB, spina bifida; BOSK, Parent organization for families with neurological diseases; USAGE, Utrecht Spina Bifida and Graded Exercise.

Table 1. Ambulation Level Classification by Hoffer,[26] Adapted by Schoenmakers[27]

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
Nonfunctional ambulation	Walking only in therapeutic situations
Nonambulation	Wheelchair dependent

the sum of the skinfolds was used as an index for subcutaneous fat.[30]

Muscular strength. Muscle strength was tested through the use of a handheld dynamometer as described by Beenhakker et al.[31] Handheld grip was recorded as an index for upper muscle strength. Because of the recommendation to use a handheld dynamometer only in muscle groups scoring >3.5 in manual testing, many missing values were recorded in the lower extremity. For this reason, only the quadriceps muscle strength was reported in this study for lower extremity strength.

Peak oxygen uptake (VO_{2peak}). In this study, maximal exercise testing was measured using a graded treadmill test (EnMill, Enraf, Delft, The Netherlands), using a protocol previously analyzed for validity and reproducibility in children with SB.[32] To accommodate children with different ambulatory abilities, 2 progressive exercise test protocols were used. Children ambulating <400 m during a 6-minute walk test (6MWT) were tested with a starting speed of 2 km/h, which was gradually increased by 0.25 km/h every minute, with a set grade of 2%. Children ambulating >400 m during the 6MWT were started at a speed of 3 km/h, with the speed being increased 0.50 km/h every minute, with a set grade of 2%. The cutoff point of 400 m was chosen based on earlier testing in our laboratory.[15,32,33] Children were allowed to minimally use the handrails for maintenance of balance only. The protocols were continued until the patient stopped as a result of exhaustion, despite verbal encouragement from the test leader. During the incremental exercise test, physiological responses, which included breath-by-breath gas analysis, were measured using a heart rate (HR) monitor (Polar accurex, Polar-Nederland BV, Almere, the Netherlands) and a calibrated mobile gas analysis system (Cortex Metamax B^3, Cortex Medical GmbH,

Table 2. Questions Regarding Self-Perceived Improvement

(My) Physical Fitness/Ambulation (of My Child) Is
1. The same compared to the first measurement
2. Deteriorated since the first measurement: I notice this because
3. Improved since the first measurement: I notice this because
4. I don't know

Table 3. Example of Training Diary (Week 3)[a]

Training Session 1	Date: 10-31-2009	Duration: 28 Minutes
Step: 3		
Heart rate:	OMNI Score:	Below target heart rate?[b]
122	5-6	
141		no
117		
Training Session 2	Date: 11-05-2009	Duration: 28 Minutes
Step: 3		
Heart rate:	OMNI Score:	Below target heart rate?[b]
120	3-4	
120		No/yes
120		

[a]General remarks: Took a 5-minute break during session 2 between second and third intervals.
[b]In this example, the minimum heart rate was 120.

Table 4. Example of Physical Activity Diary

Day	What Did You Do?	For How Long?
Monday	Treadmill training	30 Minutes
Tuesday	Gym at school	45 Minutes
	Played outside	1 Hour
Wednesday	Played outside	2 Hours
Thursday	No activities	
Friday	Sports at school	2 Hours
	Treadmill training	30 Minutes
Saturday	Soccer	30 Minutes
	Played outside	1 Hours
Sunday	Treadmill training	30 Minutes

Leipzig, Germany). The Cortex Metamax is a valid and reliable system for measuring gas-exchange parameters during exercise.[34,35] Besides physiological responses, maximal walking or running speed was recorded. Reliability of maximal exercise parameters in children with SB is good to excellent, with agreement for VO_{2peak} being 4.5%.[36]

Measures of Ambulation. Six-minute walk test. The 6MWT was used as a functional measure of ambulation and was performed on a 13-m track in a straight corridor. Patients were instructed to cover the largest possible distance in 6 minutes at a self-selected walking speed. The test and encouragements during the test were performed in accordance with the American Thoracic Society guidelines.[37] Next to HR in the last minute, the 6-minute walking distance (6MWD) was recorded as the primary outcome measure. Reproducibility of the 6MWT in children with SB is excellent.[36]

Energy cost of locomotion. Steady state (SS) normalized oxygen uptake (VO_2/kg/min) was calculated during the 6MWT as the average value over the period during which oxygen consumption changed 5% or less, following the same methods as used in our reproducibility study.[38] Within the period of least differences, a SS of 2 minutes was determined. The respiratory exchange ratio (RER) was calculated as VCO_2/VO_2 during steady state. Speed (m/min) was calculated as distance (m)/6 (min). Subsequently, the following parameters were derived: gross energy consumption (ECS_{gross}) and gross energy cost (EC_{gross}). ECS was expressed in J/kg/min, using SS VO_2, and RER in the following equation: J/kg/min = $(4.960 \times RER + 16.040) \times VO_2$/kg.[39] Furthermore, EC_{gross} expressed in J/kg/min was calculated, dividing ECS_{gross} by speed. Reproducibility of these energy measures was previously reported,[38] with best outcomes for gross measures.

Measures of Self-Reported Change and Adherence to the Program. After the intervention period, children and parents were asked to answer questions regarding change in both aerobic fitness and ambulation (see Table 2).

During the intervention period, a diary of daily physical activity was completed, including a diary of adherence to the exercise program for those in the intervention group (see Table 3 and Table 4). After the postintervention follow-up, children and parents from the intervention group were asked whether they had continued the treadmill program or any other type of physical activity.

Intervention. The intervention consisted of a home-based, individualized, and supervised program taking into account HR_{peak} as well as the child's own speed during the 6MWT to ensure an adequate intensity of training for both endurance and functional gait. Based on recommendations from the literature,[40] the training consisted of intervals of different speeds, increasing throughout the 12-week period based on fatigue, measured by the OMNI rating of perceived exertion scale and HR during exercise. In line with guidelines for exercise in children with chronic disease and/or disability, the child moved up to the next level[41] when fatigue reached 5 or below (on the 0-10 OMNI Scale) and/or when HR was below 66% of HR_{peak}. For a full description of the program, see Table 5. Children in the intervention group were instructed to exercise twice a week, after their regular care. Children in the control group were instructed to maintain regular care and regular patterns of physical activity.

Table 5. Intervention Protocol

Step	Intervals	Repetitions	Total
Step 1	2 Minutes at 70% $speed_{6MWT}$ + 4 minutes at 100% $speed_{6MWT}$	3	18 Minutes
Step 2	3 Minutes at 70% $speed_{6MWT}$ + 4 minutes at 100% $speed_{6MWT}$	3	21 Minutes
Step 3	3 Minutes at 70% $speed_{6MWT}$ + 4 minutes at 110% $speed_{6MWT}$	4	28 Minutes
Step 4	3 Minutes at 80% $speed_{6MWT}$ + 4 minutes at 120% $speed_{6MWT}$	4	28 Minutes
Step 5	3 Minutes at 80% $speed_{6MWT}$ + 5 minutes at 120% $speed_{6MWT}$	4	32 Minutes
Step 6	4 Minutes at 80% $speed_{6MWT}$ + 6 minutes at 120% $speed_{6MWT}$	3	30 Minutes
Step 7	4 Minutes at 90% $speed_{6MWT}$ + 6 minutes at 130% $speed_{6MWT}$	3	30 Minutes
Step 8	3 Minutes at 90% $speed_{6MWT}$ + 7 minutes at 130% $speed_{6MWT}$	3	30 Minutes
Step 9	2 Minutes at 90% $speed_{6MWT}$ + 8 minutes at 130% $speed_{6MWT}$	3	30 Minutes
Step 10	3 Minutes at 100% $speed_{6MWT}$ + 7 minutes at 140% $speed_{6MWT}$	3	30 Minutes
Step 11	2 Minutes at 100% $speed_{6MWT}$ + 8 minutes at 140% $speed_{6MWT}$	3	30 Minutes
Step 12	1 Minute at 100% $speed_{6MWT}$ + 9 minutes at 140% $speed_{6MWT}$	3	30 Minutes

Abbreviation: 6MWT, 6-minute walk test.

Postintervention Period. After the intervention period, patients were followed up to see if they were able to maintain the effects of training on their own, by either continuing the treadmill program or increasing daily physical activity. The treadmill remained at home during this postintervention period. Children in both groups did not receive specific instructions during this period with regard to their daily physical activity.

Data Analysis

Data were first checked to determine whether they met assumptions for further statistical testing, including normality of data and normality of residuals and outliers. Missing values (n = 2 in both control and intervention groups) in the follow-up measurements were filled in using the feed forward method.

Baseline data. Differences in baseline data between the intervention and control groups were analyzed using unpaired *t* tests and Mann-Whitney tests for nonnormally distributed data. Frequencies were compared, using the χ^2 test. To gain more insight into relations within baseline measures, Pearson or Spearman correlations were computed, depending on the type of data.

Efficacy of intervention. Changes between baseline (T0) and postintervention (T1) measurements were calculated for both groups. Differences in change were compared between the groups using unpaired *t* tests or Mann-Whitney tests. For nominal data, the χ^2 test was used. Effect sizes were calculated using Cohen *d* for significant differences. Values of Cohen *d* less than 0.20 were considered small, between 0.20 and 0.50 were considered medium, and effect sizes greater than 0.80 were considered large.

Maintenance of treatment effects. To test whether gains were maintained after treatment, paired *t* tests were used for parameters that had shown improvement after training. Frequencies were used to describe the continuation of the program and self-reported changes. Mixed between- and within-subjects analysis of variance was used to analyze the long-term effects of intervention. Effect sizes were calculated as partial η^2,[42] with $\eta < 0.01$ being considered small, $\eta = 0.06$ moderate, and $\eta > 0.14$ as large effects. Significance for all tests was set at $P = .05$. Statistical analyses were performed using statistical package SPSS version 17.

Results

Baseline characteristics of both the control and intervention groups are given in Table 6. No significant differences were found at baseline between them. Presence of hydrocephalus and shunting were equally distributed between the groups as well as motor level and Chairi II malformation, with significance ranging from $P = .14$ to .95. Despite the inclusion of normal and community ambulatory children only, the data do reflect a heterogeneous functional group as depicted by large SDs (standard deviations) for the 6MWD, EC_{gross}, and self-reported physical activity. Secondary analysis for the group as a whole showed that *Z* values for BMI increased with age ($r = 0.039$, with $P = .02$), and the level of lesion was correlated with outcomes of 6MWT ($r = 0.51$ with $P = .03$) and normalized VO_{2peak} ($r = 0.50$, with $P = .04$).

Efficacy of Intervention

Children in the intervention group reported a mean (±SD) of 22.6 (±6.2) completed training sessions, which resulted in 49.4 minutes (±15) of treadmill ambulation per week, not including warming up or cooling down. The average training step reached was 9.5 (±3.2).

Table 6. Baseline Characteristics

	Control	Intervention	*P* Value
Anthropometrics			
Age	11.1 (2.6)	10.3 (2.9)	.715
Boys:Girls	9:5	9:9	.42[a]
Height	141.6 (16.1)	138.0 (19.7)	.319
Weight	43.4 (18.6)	42.7 (22.5)	.255
BMI	20.2 (5.1)	20.9 (5.7)	.713
Sum of skinfolds	101.9 (56.6)	108.1 (58.2)	.775
Ambulation			
Hoffer NA:CA	4:10	5:13	.96[a]
6MWD	372.1 (116.5)	344.8 (125.3)	.643
EC_{gross}	9.5 (6.9)	10.5 (6.4)	.831
ECS_{gross}	487.4 (84.7)	460.2 (96.3)	.427
Maximum test			
VO_{2peak}	33.4 (11.0)	32.3 (7.1)	.185
HR_{peak}	174 (26)	169 (45)	.669
RER_{peak}	0.97 (0.1)	0.96 (0.1)	.769
$Speed_{peak}$	5.7 (1.9)	5.2 (2.0)	.406
Muscle strength			
Handgrip	86.4 (55.4)	73.9 (46.9)	.495
Quadriceps	157.9 (51.0)	155.2 (62.8)	.897
Fatigue and physical activity			
Total fatigue PEDS QL (%)	74.2 (16.2)	70.5 (16.4)	.550
Self-reported physical activity (min/wk)	335.2 (176.2)	415.4 (171)	.226

Abbreviations: NA, normal ambulatory; CA, community ambulatory; 6MWD, 6-minute walking distance; EC, energy cost; ECS, energy consumption; HR, heart rate; RER, respiratory exchange ratio; PEDS QL = Pediatric Scale for Quality of Life.
[a]χ^2 Test.

No significant differences were seen between groups with regard to changes in anthropometric parameters and muscle strength (Table 7). Positive values can be interpreted as growth or improvement, with the exception of EC, in which negative values reflect improvement. With respect to ambulation, there were significant differences between the 2 groups. After the intervention period, the children in the intervention group showed an improvement of 38.7 m during the 6MWT, whereas the children in the control group walked 2 m less compared with the first measurement.

The larger distance in the intervention group did not change EC_{gross} of locomotion, whereas it did raise energy consumption per minute. Energy cost was lowered significantly within the control group, but this difference falls within the measurement variability and was not significant compared with that for the intervention group.[38] The maximum graded exercise test showed significant differences between the groups in both VO_{2peak} and maximum speed. Effect sizes showed large effects for ambulation during both the 6MWT and treadmill test. It showed a moderate effect for VO_{2peak}. Because of the large SDs in the baseline measurements, secondary analyses were performed to analyze correlations between differences and baseline measurements. No correlations between baseline measures of ambulation or maximal exercise testing and differences after intervention were present. Differences after intervention for maximal

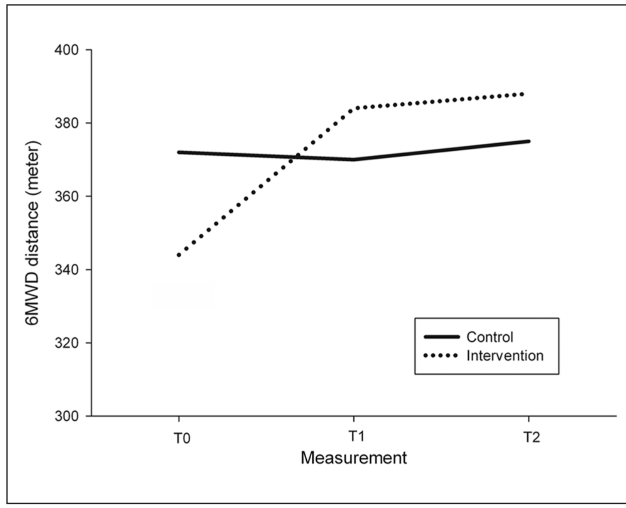

Figure 2. Maintenance of 6-minute walking distance (6MWD) at baseline, after intervention, and 3 months after last training session.

Table 7. Difference After Intervention Period (T1 − T0)

	Control	Intervention	*P* Value	Effect size
Anthropometrics	Mean (SD)	Mean (SD)		
Height	0.92 (1.1)[a]	1.2 (1.6)[a]	0.8[b]	
Weight	1.0 (1.9)	0.2 (1.6)	0.2	
BMI	−0.3 (0.9)	−0.1 (0.9)	0.1	
Sum of skinfolds	−2.4 (8.2)	−1.7 (17.5)	0.9	
Ambulation				
6MWD	−2.1 (27.8)	38.7 (34.6)[a]	0.002[c]	1.08
Percentage change 6MWD	−1.5 (10.1)	13.0 (12.4)[a]		
EC_{gross}	−0.8 (1.3)[a]	−0.3 (1.1)	0.2	
ECS_{gross}	−41.3 (63.7)[a]	49.1 (88.5)[a]	0.004[c]	1.01
Maximum test				
VO_{2peak}	−3.0 (7.5)	1.4 (3.7)	0.034[c]	0.78
Speed	−0.06 (0.6)	0.9 (0.8)[a]	0.001[c]	1.14
Muscle strength				
Handgrip (N)	−3.0 (7.6)	1.6 (9.9)	0.2	
Quadriceps (N)	−27.2 (27.2)[a]	−8.7 (71.7)	0.7[b]	
Fatigue and physical activity				
Total fatigue PEDS QL (%)	−0.4 (9.9)	7.8 (9.8)[a]	0.06	
Self-reported physical activity (min/wk)	−22.4 (145.3)	−8.3 (273.1)	0.66[b]	

Abbreviations: SD, standard deviation; 6MWD, 6-minute walking distance; EC, energy cost; ECS, energy consumption; PEDS QL = Pediatric Scale for Quality of Life
[a]Significant differences within groups.
[b]Nonparametric Mann-Whitney test.
[c]Significant difference between groups.

speed, VO_{2peak}, and 6MWD all correlated significantly with each other (*P* values between .03 and .04).

Self-perceived change showed significant differences between the groups, with 72% and 50% of parents of children in the intervention group reporting positive changes in endurance and ambulation, respectively, versus 5% ($P = .001$) and 0% ($P = .006$) in the control group. Qualitative changes were reported in terms like “now being able to join regular physical education classes at school,” “nicer ambulation,” “not wanting to use the wheelchair anymore for longer distances,” “more initiative,” and “easier to walk up and down stairs.”

Maintenance of Treatment Effects

t Tests showed no significant changes between the post-intervention measurements and those taken 3 months after the last training for ambulation and aerobic fitness measures. This indicates that the gains from the intervention were not lost (see Figure 2). Mixed between- and within-subjects analysis of variance showed a large long-term interaction effect of treatment for 6MWD ($P = .003$, $\eta = 0.34$), maximal speed ($P = .003$, $\eta = 0.35$), and ECS_{gross} ($P = .014$, $\eta = 0.29$).

When asked whether the children in the intervention group had continued their program, 30% had continued using the treadmill, 27% had started some other type of activity (walking, biking, horseback riding, or joining regular physical education classes at school), and 39% reported no continuation or start up of new activities. Reasons for continuation were “just for fun,” “to further improve,” or “my parents told me so”; reasons for not continuing were mostly “no time” or “no interest”; 1 child was adapted with new orthotics making it impossible to continue activities.

Discussion

The aim of this study was to evaluate the effects of an individualized treadmill training program to improve aerobic fitness and ambulation in ambulatory children and adolescents with SB. We found large effect sizes for both over ground ambulation during the 6MWT and treadmill walking. These functional changes were not related to our initial hypothesis of “diminished physiologic reserve” because we found moderate increases in VO_{2peak} and increases in energy consumption during locomotion. The changes in ambulation, however, are in line with those for treadmill training in children with cerebral palsy or spinal cord injury.[43]

Although energy cost per meter did not change significantly, energy consumption did increase after intervention. At first, this seems like an unwanted change as it could indicate less efficiency during gait. Without greater energy cost, the finding suggests that more muscle mass was involved, resulting in faster ambulation. Unfortunately, without kinematic and electromyographic evaluation, this is hard to prove. Studies of gait changes after treadmill training concluded that improvements in speed strongly correlated to kinematic changes and adaptations of muscle activation.[44] What was interesting was that maximal strength did not change after training, similar to our results. Changes at the spinal cord level and other levels for learning may have contributed.[45-47]

With regard to VO_{2peak}, there was a moderate effect size. Some studies in other pediatric populations have reported larger increases in aerobic fitness,[20,21] and this was seen to vary with the population.[48] Some studies report a washout effect, starting 6 weeks after cessation of the training. Furthermore, the total intervention time (12 weeks) was relatively short. The study by Verschuren et al[21] has shown that improvements continue during an 8-month trial in children with cerebral palsy. One of the principles of training is the principle of overload of the target system.[49] In the case of VO_{2peak}, the cardiorespiratory system needs to be trained at a sufficient intensity for a sufficient period of time. Looking at the adherence diaries, children did comply with twice-a-week training for an average of 25 minutes per session, which could be considered a minimal time and frequency for training. As for intensity though, Baquet et al[50] concluded in a review that a target intensity of at least 80% of HR_{peak} is required when aiming to improve VO_{2peak} in children. In our study, to prevent overtraining and injuries to the musculoskeletal system, we adhered to guidelines for children with chronic disease, in which training intensities at 60% to 70% of HR_{peak} are recommended.[41] This may explain the relatively smaller improvements in VO_{2peak}. During our training session, no injuries were reported, so it should be feasible to increase training intensity in children with SB.

Yet another principle of training is “specificity,” which refers to training the activity that needs improvement[49]—in this case, ambulation. Although it would be tempting to increase the speed on the treadmill to running speed so as to increase the cardiovascular load, it would decrease the specificity of training by altering the movement pattern from walking to running. When implementing exercise training in ambulatory children with SB, one needs to choose which system will be the target of training. This should be done based on the limiting factor in daily life (lack of aerobic fitness or walking pattern) or based on which system still shows room for improvement. Looking at the self-reported patient outcome measures, the majority of parents and children in the intervention group did notice positive effects in activities of daily life, compared with none in the control group. These changes could easily be described as a “placebo effect” because the participants were not blinded. In this study, though, these positive results are in line with the gains in ambulation and, at the same time, illustrate nicely how positive laboratory changes are being perceived in daily life.

Looking at the possible washout effects, it is interesting to note that there were no significant differences between T1 (directly postintervention) and T2 (3 months after intervention), indicating that children were able to maintain their functional gains for 3 months after training. This could be explained by the fact that close to 30% of the children had continued the treadmill training and another 30% had started some other type of sport. For some children, the training had made it possible to function at a higher level during sports: for example, joining regular physical education classes at school or starting to play soccer. These findings do feed the theory of Bar-Or and Rowland[49] and Durstine et al,[51] in that reversing the downward spiral of detraining and deconditioning by improving function may indirectly lead to increased levels of activity and, in the long run, possible increased levels of physical fitness (Table 7).

Limitations of the Study

The limitations of the study mainly involve the size of the study and the heterogeneity of the group. Although heterogeneity is not ideal for research purposes, anyone working with this group of patients knows the many medical procedures these children undergo. We did not find significant differences between those taking medication and those who did not. Looking at baseline measures, measures of ambulation, maximal testing, and muscle strength all show large variation as well. For this reason, we did look at correlations between baseline measures and outcomes of training and but did not find any.

Because of the small sample size, we decided to use the feed-forward technique to be able to include all postintervention data into the mixed between- and within-subjects analysis of variance for long-term effects. To make sure that this statistical intervention did not influence the outcomes, we first analyzed the available postintervention data using paired *t* tests. Results showed no changes between T1 and T2, making the feed-forward technique appropriate to use because it assumes no changes compared with the last measurement.[52] We also experienced many missing values because of software problems.

Finally, subjective recall of daily physical activity when using questionnaires could be misleading. Accelerometry use in future trials may yield more accurate results.[53]

Conclusion

For ambulatory children with SB, progressive treadmill training has large long-term effects on ambulation and a moderate short-term effect on VO_{2peak}.

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