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Research

Physical activity stimulation program for children with cerebral palsy did not improve physical activity: a randomised trial

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KEY WORDS

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ABSTRACT

Question: In children with cerebral palsy, does a 6-month physical activity stimulation program improve physical activity, mobility capacity, fitness, fatigue and attitude towards sports more than usual paediatric physiotherapy? **Design:** Multicentre randomised controlled trial with concealed allocation, blinded assessments and intention-to-treat analysis. **Participants:** Forty-nine walking children (28 males) aged 7–13 years with spastic cerebral palsy and severity of the disability classified as Gross Motor Function Classification System level I–III. **Intervention:** The intervention group followed a 6-month physical activity stimulation program involving counselling through motivational interviewing, home-based physiotherapy, and 4 months of fitness training. The control group continued their usual paediatric physiotherapy. **Outcome measures:** Primary outcomes were walking activity (assessed objectively with an activity monitor) and parent-reported physical activity (Activity Questionnaire for Adults and Adolescents). Secondary outcomes were: mobility capacity, consisting of Gross Motor Function Measure-66 (GMFM-66), walking capacity and functional strength, fitness (aerobic and anaerobic capacity, muscle strength), self-reported fatigue, and attitude towards sport (child and parent). Assessments were performed at baseline, 4 months, 6 months and 12 months. **Results:** There were no significant intervention effects for physical activity or secondary outcomes at any assessment time. Positive trends were found for parent-reported time at moderate-to-vigorous intensity (between-group change ratio = 2.2, 95% CI 1.1 to 4.4) and GMFM-66 (mean between-group difference = 2.8 points, 95% CI 0.2 to 5.4) at 6 months, but not at 12 months. There was a trend for a small, but clinically irrelevant, improvement in the children's attitudes towards the disadvantages of sports at 6 months, and towards the advantages of sports at 12 months. **Conclusions:** This physical activity stimulation program, that combined fitness training, counselling and home-based therapy, was not effective in children with cerebral palsy. Further research should examine the potential of each component of the intervention for improving physical activity in this population. **Trial registration:** NTR2099. **[Van Wely L, Balemans ACJ, Becher JG, Dallmeijer AJ (2014) Physical activity stimulation program for children with cerebral palsy did not improve physical activity: a randomised trial. *Journal of Physiotherapy* 60: 40–49]**

Introduction

Maintaining physical activity is especially important for children with physical disabilities such as cerebral palsy because their impairments can interfere with daily activities and participation in sport.[1] Children with cerebral palsy have lower levels of fitness[2,3] and physical activity[4] than children with typical development, and show a decrease in physical activity with increasing mobility problems.[5] Low levels of physical activity might lead to reduced levels of fitness and further deterioration of mobility, resulting in a vicious cycle of deconditioning and decreasing physical activity. Because physical activity behaviour may track into adolescence and adulthood,[6] it is important to intervene at an early stage to prevent school-age children with cerebral palsy from becoming even less active during adolescence.

'What a child can do' is not directly associated with 'what a child does do' in daily life.[7] Therefore, treatment programs in paediatric physiotherapy should include physical activity counselling and fitness promotion.[8] Exercise programs can improve the fitness levels of children with cerebral palsy,[9,10] but only limited information is available on the effectiveness of interventions for children with cerebral palsy on physical activity. A 2-month internet-based physical-activity-counselling program[11] and a 9-month fitness-training program[9] each reported non-significant but

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favourable trends in physical activity. A combination of fitness training and physical activity counselling may interrupt the vicious cycle of deconditioning in people with disabilities.[1] Additionally, recent work has addressed the need for home-based programs to improve the transfer of mobility-related skills practised in the therapy setting to the daily life situation.[12] This evidence motivated the development of the LEARN 2 MOVE 7-12 physical activity stimulation program, involving a lifestyle intervention with counselling and home-based physiotherapy, and a fitness training program.[13]

It was hypothesised that counselling focused on opportunities for increasing physical activity rather than on restrictions, in combination with practice of mobility-related skills in the home situation and fitness training, would work synergistically to break the vicious cycle of deconditioning. In addition, it was hypothesised that participation in the fitness-training component with other children with a disability would positively influence the children's and parents' attitudes towards sport, which is supposed to be a mediating factor for physical activity. Therefore the research question for this study was:

> In children with cerebral palsy, does the 6-month LEARN 2 MOVE 7-12 physical activity stimulation program improve physical activity, mobility capacity, fitness, fatigue, and attitude towards sports more than usual paediatric physiotherapy?

Method

Design

This multi-centre, parallel-group randomised controlled trial with concealed allocation and blinded assessments was conducted in paediatric physiotherapy practices and special schools for children with disabilities in the Netherlands between September 2009 and February 2011. In a previous publication we described the study design extensively.[13] The effects of the physical activity stimulation program on social participation, quality of life and self-perception will be reported in a separate paper. Participants were randomised 1:1 to the experimental or control intervention, with stratification by Gross Motor Function Classification System (GMFCS) level I versus level II/III. The GMFCS level I is walking without limitations, level II is walking with limitations and level III is walking with a hand-held mobility device.[14] Sealed envelopes were used to conceal group allocation. Participants were informed of group allocation following the baseline assessments. The intervention group followed a 6-month physical activity stimulation program, involving a lifestyle intervention and 4 months of fitness training. The control group continued their usual paediatric physiotherapy. Outcomes were assessed in the hospital: at baseline; at 4 months (ie, at the end of fitness training, when only walking capacity, functional strength and fitness were assessed); at 6 months (that is, at the end of the intervention); and at 12 months. The assessor (AB) was blinded to group allocation throughout the study. The parents' attitudes towards sport were only assessed at baseline and 12 months.

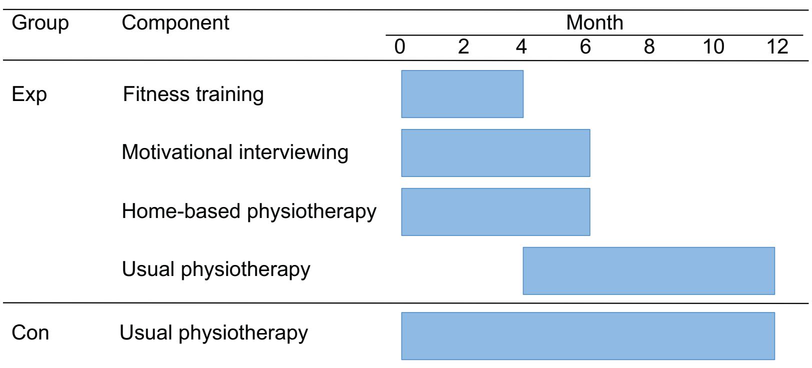
Participants, therapists and centres

Children with spastic cerebral palsy, aged 7–13 years who could walk were recruited via paediatric physiotherapy practices and special schools for children with disabilities. Inclusion criteria were: classification in GMFCS level I–III, understanding of the Dutch language and fulfilling at least one of the following criteria as determined in a telephone interview: less active than the international physical activity norm of less than 1 hour daily at >5 metabolic equivalents (METs), which is moderate or vigorous intensity;[15] no regular participation in sports or (physiotherapeutic) fitness program (ie, less than three times a week for at least 20 minutes); and experience of problems related to mobility in daily life or sports. Exclusion criteria were: surgery in the previous 6 months, botulinum toxin treatment or serial casting in the previous 3 months (or planned), unstable seizures, contra-indications for physical training, severe behavioural problems, severe intellectual disability and a predominantly dyskinetic or ataxic movement disorder.

Intervention

The intervention group followed the physical activity stimulation program, which involved a lifestyle intervention and fitness training followed by usual physiotherapy. The control group undertook only usual physiotherapy. The components of the interventions are presented in Figure 1 and described in more detail elsewhere.[13]

The lifestyle intervention included counselling to motivate and coach the children and the parents to adopt more active lifestyles, as well as home-based physiotherapy. Parents and children received counselling at home by the researcher (LW) using the motivational interviewing technique.[16] This client-centred interview style is aimed at eliciting behavioural change and offers strategies to deal with resistance to change. The key principle of this interview technique is that the client indicates which goals are feasible to achieve and what help is needed to achieve them. As a minimum, the coordinating researcher initiated three counselling sessions. The client could receive more counselling upon request. Home-based physiotherapy, aimed at increasing the capacity for daily activities in a situation relevant for the children, was tailored



Exp = experimental group, Con = control group.

Figure 1. Design of the experimental (physical activity stimulation program) and control group interventions.

individually in response to the inventory of mobility-related problems experienced by children and parents. The children's regular physiotherapists provided the home-based physiotherapy. The fitness training program was aimed at increasing lower-extremity muscle strength and anaerobic fitness, and was based on existing training protocols for children with cerebral palsy that have been proven to be effective for increasing muscle strength[17] and anaerobic capacity.[10] Children trained for 4 months, in groups of 2 to five, under the supervision of their physiotherapists. During the first 2 months, children trained for 1 hour, twice a week. In the following 2 months, training frequency was reduced to once a week, allowing children to start participating in other physical activities during the intervention, as a result of the counselling. Each training session consisted of a warm-up, two lower-extremity muscle strength exercises with a weight vest (sit-to-stand and frontal/lateral step-up or half-knee raise), three anaerobic game-like exercises (for example, running or slaloms), and a cool-down. Training load was progressively increased during the training period. To ensure standardisation of the intervention, all physiotherapists in the intervention groups received two workshops, a training manual and two visits by the coordinating researcher during the training period. For each training session physiotherapists recorded the training load, the number of sets and repetitions of the exercises, and any adverse events.

The control group continued their usual paediatric physiotherapy at the physiotherapy practice and did not receive counselling.

Outcome measures

Primary outcome

The primary outcome was physical activity measured in two ways: an objective assessment of walking activity, and a subjective assessment of physical activity by parental report. Walking activity was assessed for 1 week using an ankle-worn bi-axial accelerometer,[a] which registered accelerations in the frontal and sagittal plane at regular time intervals. By sensitivity-adjusted calibration, as previously described,[18] the accelerometer can accurately record strides (ie, complete gait cycles) for children with cerebral palsy by measuring the steps of one leg.[5] The number of strides per day, time spent at medium-to-high stride rate (>15 strides/minute), time spent at high stride rate (>30 strides/minute), and time spent inactive (0 strides/minute), were calculated in minutes. All representative days were used to calculate averages for schooldays, and weighted total values reflecting an average weekday, based on schooldays and weekend days. Parent-reported physical activity was assessed using the child-adapted Activity Questionnaire for Adults and Adolescents (AQuAA), which includes questions about the frequency, duration and intensity of the child's physical activities and sedentary behaviour in the previous 7 days.[19] Based on this information and the corresponding METs of the reported activities, the following outcome measures were calculated: weekly time spent at moderate-to-vigorous intensity (>5 METs), whether children met the physical activity guideline (one hour daily at >5 METs), and weekly time spent inactive (<2 METs). Parents also indicated whether their child was being physically active as part of sports club participation (yes/no).

Secondary outcomes

The secondary outcomes included: mobility capacity (gross motor capacity, walking capacity and functional muscle strength); fitness (isometric muscle strength, aerobic capacity and anaerobic capacity); self-reported fatigue; and attitude towards sports. Gross motor capacity was evaluated with the Gross Motor Function Measure-66 (GMFM-66) item sets.[20] Walking capacity was determined with the 1-minute walk test, which measures the completed distance in 1 minute of walking as fast as possible without running.[21] Functional muscle strength encompassed the number of lateral step-ups (left and right leg) and sit-to-stands achieved during 30 seconds.[22] Isometric muscle strength of the knee extensors and hip abductors was determined with a hand-held dynamometer[b] as the peak moment in Nm.[23] Aerobic capacity was assessed with a continuous progressive exercise test on a cycle ergometer.[2,c] To determine peak oxygen uptake (ml/minute) pulmonary gas-exchange was measured with the Quark CPET system.[d] Peak power output (W) was defined as the highest power output during the test. On the same cycle ergometer, children performed the 20-second Wingate sprint test to determine mean power output, as a measure of anaerobic capacity.[24] The children cycled as fast as possible for 20 seconds against a constant braking force. Fatigue was assessed with the PedsQL Multidimensional Fatigue Scale,[25] which provides domain scores for general fatigue, sleep/rest fatigue and cognitive fatigue, and a total score. Attitude towards sports was assessed using four different viewpoints: the children's attitudes towards the advantages of sport (seven statements), their attitudes towards the disadvantages of sport (six statements), the parents' attitudes about the accessibility of sport clubs (seven statements), and the attitudes of the parents and immediate family members towards the importance of sports ('father/mother/brother/sister thinks sport is important'). All statements were scored on a five-point ordinal scale ('totally disagree' to 'totally agree') and average domain scores were used for analyses.[26] More information about the psychometric validity of the outcome measures, as well as detailed assessment procedures have been described elsewhere.[13,18]

The assessment procedure was as follows: at home, the parents and children completed the AQuAA, the Multidimensional Fatigue Scale, and the attitude questionnaires. At the hospital body height and weight were measured, and several family characteristics were determined (siblings, parental marital status, parental educational level and sports frequency of the immediate family). Selective motor control was assessed with the modified Trost test, during which the ability of children to dorsiflex the ankle and extend the knee in an isolated movement was scored in four categories: N/A = not able to make the movement, 0 = completely synergistic, 1 = partly synergistic, 2 = no synergy.[27] Scores for each joint and leg were added to obtain a total score for selective motor control. Parents also indicated the sports frequency of immediate family members in five categories (from 1 = never to 5 = daily), from which a mean score was calculated. Children then completed mobility capacity assessments and fitness tests, after which the calibrated accelerometer was provided to register walking activity for one week. Additionally, children and parents received a diary to record their daily activities and accelerometer registration time. Information on data processing and controlling data quality of the accelerometer has been described elsewhere.[18]

Data analysis

A priori sample size calculation indicated that 22 children were needed in each group to detect a clinically relevant difference of 1000 strides per day between groups.[28] Power was set at 80%, significance level at 5% and the standard deviation of the difference was set at 1175 strides (unpublished pilot data of Dutch children with cerebral palsy). To allow for 10% loss to follow-up, 25 children were included in each group.

To determine the intervention effect, intention-to-treat analyses were performed using linear regression, or logistic regression for dichotomous outcomes ($p < 0.05$). Outcomes at 4 months, 6 months, and 12 months were the dependent variables, and group allocation and the measured outcome at baseline were the independent variables in the analyses. To correct for performing statistical tests over multiple time points, the critical *p*-value was divided by the number of tests performed, resulting in an alpha < 0.025 for outcomes measured three times, and an alpha < 0.017 for outcomes measured four times. Variables with non-normally distributed residuals were logarithmically transformed prior to performing linear regression analyses, after which the results were transformed back, providing a between-group change ratio. If residuals remained skewed after transformation, a non-parametric Mann–Whitney *U* test was used. Age, gender, selective motor control and sport frequency of the immediate family were included as covariates in the analyses when they changed the intervention effect by more than 10%.

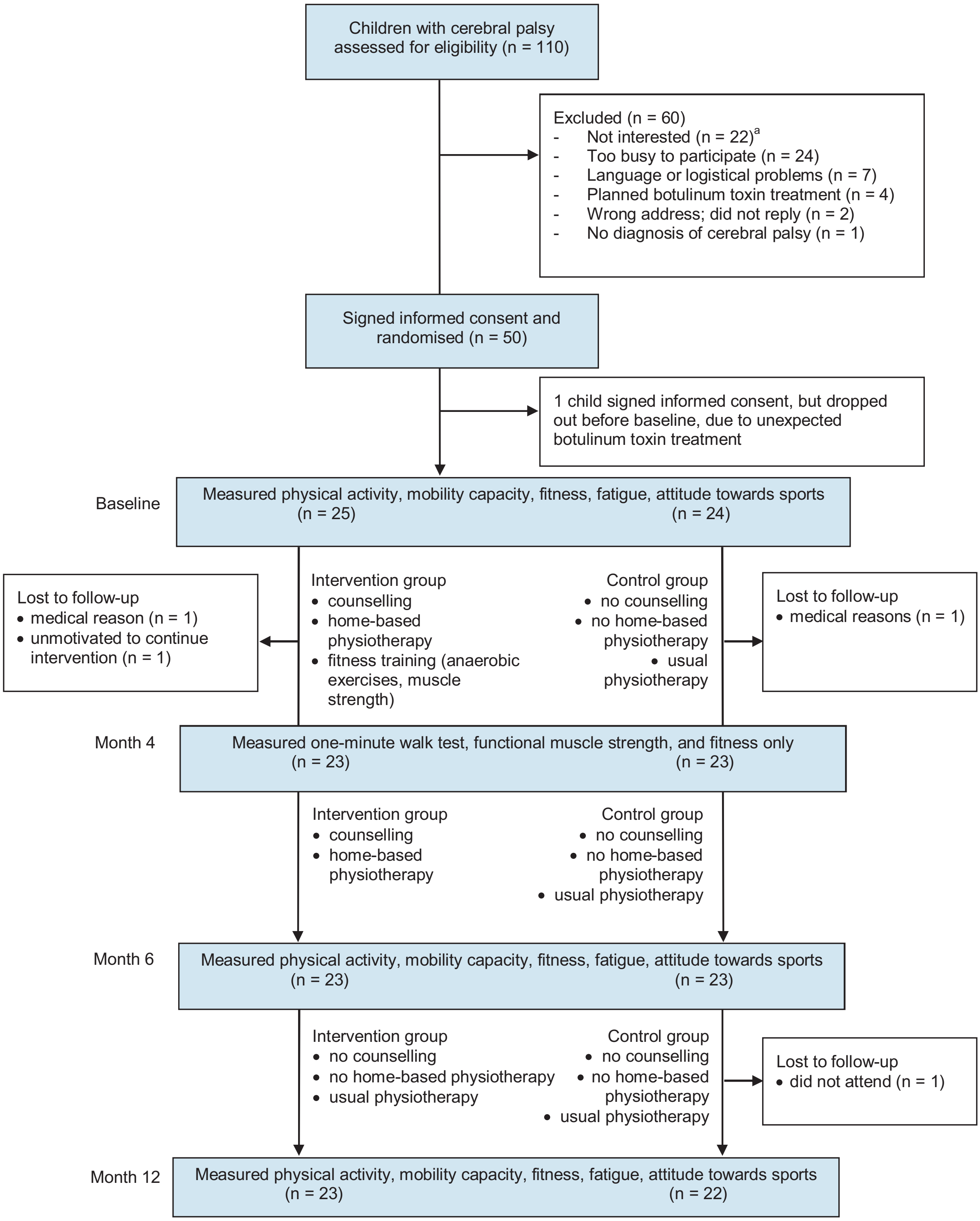


Figure 2. Flow of participants through the trial.
[a]Eight lacked interest because they were to active.

Results

Flow of participants, therapists and centres through the study

In total, 110 children with cerebral palsy were invited to participate, as presented in Figure 2. Fifty children agreed, signed an informed consent form and were randomised to either the experimental (n = 25) or control (n = 25) groups. Children were treated at 13 paediatric physiotherapy practices (n = 27) and three special schools for children with disabilities (n = 23). One child (control group) dropped out before baseline assessments due to unexpected botulinum toxin treatment. Three children (experimental group: n = 2, control group: n = 1) dropped out during the first 4 months of the intervention, and one child (control group) missed the 4-month and 12-month assessments. Reasons for loss to follow-up are presented in Figure 2. The baseline characteristics of the participants are presented in Table 1 and in the first two columns of Tables 2–7.

Compliance with the trial method

The families in the experimental group received a median of five counselling sessions (range three to nine). An inventory of previously experienced mobility-related problems resulted in home-based physiotherapy for 14 of the 23 children in the experimental group. Adherence to the fitness training sessions was 91%, with children attending an average of 22 (SD 2, range 17 to 24) of the 24 training sessions. After a 3-week familiarisation period, training intensity of the loaded sit-to-stand increased from 79% (5.9 kg) of the predicted twelve-repetition maximum (ie, 10.6 kg)[13] in the fourth week, to 116% (8.7 kg) in the eighth week, and to 141% in the final week. The intensity of the anaerobic exercises increased from the fourth to the last week according to the protocol, by reducing the work:rest ratio from 1:4 to 1:3 when performing five sets of 20-second exercises.[13] No serious adverse effects were reported except for one child (female, GMFCS III) who reported hip complaints during the training. After taking rest (omitting two training sessions) and reduction of the training intensity, she was able to resume and complete the training program. Blinding was successful, with the assessor correctly guessing group allocation at a rate similar to chance throughout the trial. Some children did not complete all assessments on each occasion due to motivational problems or time constraints, as illustrated by the number of analysed cases in the tables. One child at 6 months, and four children at 12 months did not wear the accelerometer.

Effect of intervention

Primary outcome

No significant intervention effect was found for walking activity or for parent-reported physical activity at 6 months and 12 months (Tables 2 and 3). A trend in favour of the intervention group was identified at 6 months for parent-reported time at moderate-to-vigorous intensity (between-group change ratio 2.2, 95% CI 1.1 to 4.4), but not at 12 months.

Secondary outcomes

No significant intervention effect was demonstrated for mobility capacity (Table 4), attitude towards sports (Table 5) and the other secondary outcomes (Tables 6 and 7) at 4 months, 6 months or 12 months. (See eAddenda for Tables 6 and 7.) A positive trend was found for the GMFM-66 at 6 months (mean between-group difference 2.8, 95% CI 0.2 to 5.4), but not at 12 months, and for the 1-minute walk test at 4 months (mean between-group difference 5 m, 95% CI 0 to 9), but not at 6 months or 12 months. For attitude towards sports, when compared to the control group, there was also a trend for reporting greater agreement with possible advantages

Table 1
Characteristics of participants.

	Started with the study		Lost to follow-up	
	Exp (n = 25)	Con (n = 24)	Exp (n = 2)	Con (n = 1)
Children				
Age (yr), mean (SD)	9.5 (1.5)	10.0 (1.8)	10.4 (1.8)	11.1 (–)
Gender, n male (%)	12 (48)	16 (67)	1 (50)	1 (100)
Height (m), mean (SD)	1.36 (0.11)	1.38 (0.14)	1.48 (0.07)	1.35 (–)
Weight (kg), mean (SD)	33.4 (8.8)	36.4 (12.6)	39.0 (2.0)	31.0 (–)
Body mass index (kg/m^2)	17.8 (2.7)	18.5 (3.6)	17.9 (0.74)	17.0 (–)
GMFCS level, n (%)				
I	15 (60)	13 (54)	2 (100)	0 (0)
II	6 (24)	6 (25)	0 (0)	0 (0)
III	4 (16)	5 (21)	0 (0)	1 (100)
Type of CP, n (%)				
Unilateral spastic	12 (48)	11 (46)	2 (100)	0 (0)
Bilateral spastic	13 (52)	13 (54)	0 (0)	1 (100)
Selective motor control (0–8), mean (SD)[a]	5 (2)	4 (2)	6 (0)	1 (–)
Wheelchair long distances, n yes (%)	5 (20)	5 (21)	1 (50)	0 (0)
Orthoses, n yes (%)	17 (68)	15 (62)	1 (50)	1 (100)
School type, n (%)				
Regular education	14 (56)	12 (50)	2 (100)	0 (0)
Special school for children with disabilities	11 (44)	12 (50)	0 (0)	1 (100)
Family				
Siblings, n yes (%)	19 (76)	20 (83)	2 (100)	1 (100)
Parental marital status, n (%)				
Living together	22 (88)	21 (87)	2 (100)	0 (0)
Single parent	3 (12)	3 (13)	0 (0)	1 (100)
Highest parental educational level, n (%)				
High school or lower education (low)	14 (56)	13 (54)	1 (50)	0 (0)
College or university degree (high)	111 (44)	11 (46)	1 (50)	1 (100)
Sport frequency immediate family, (1–5)[a]	3 (1)	3 (1)	3 (2)	3 (–)

Exp, experimental; Con, control; CP, cerebral palsy, GMFCS, Gross Motor Function Classification System.
[a] A higher score means better selective motor control or higher sport frequency.

Table 2
Mean (SD) for walking activity outcomes for each group, mean (SD) difference within groups, and mean (95% CI) difference between groups.

Outcome	Groups						Difference between groups[a]				Difference between groups[a]	
	Baseline		Month 6		Month 12		Month 6 minus baseline		Month 12 minus baseline		Month 6 minus baseline	Month 12 minus baseline
	Exp	Con	Exp	Con	Exp	Con	Exp	Con	Exp	Con	Exp minus Con	Exp minus Con
Average weekday	(n = 23)	(n = 22)	(n = 20)	(n = 20)	(n = 19)	(n = 20)	(n = 20)	(n = 19)	(n = 19)	(n = 19)		
Strides (n/day)	5109 (1636)	5181 (1758)	5314 (2250)	6012 (1746)	4746 (1750)	4868 (1557)	268 (1108)	725 (1591)	−162 (1312)	−146 (1827)	−858 (−1819 to 104)	−175 (−1218 to 867)
Medium to high stride rate (minutes/day)	123 (43)	127 (49)	128 (59)	148 (48)	110 (45)	114 (40)	7 (31)	19 (44)	−8 (36)	−8 (49)	−23 (−50 to 3)	−5 (−31 to 22)
High stride rate (minutes/day)	49 (22)	49 (23)	55 (37)	65 (26)	45 (24)	50 (25)	7 (22)	15 (26)	0 (21)	3.4 (30)	−14 (−31 to 3)	−6 (−23 to 11)
Inactive (minutes/day)	347 (98)	354 (95)	369 (97)	347 (77)	393 (92)	381 (81)	13 (58)	−5 (53)	30 (55)	9 (74)	26 (−10 to 61)	22 (−19 to 62)
Schooldays only	(n = 23)	(n = 23)	(n = 22)	(n = 23)	(n = 21)	(n = 20)	(n = 22)	(n = 23)	(n = 21)	(n = 20)		
Strides (n/day)	5462 (1701)	5320 (1928)	5424 (2257)	5887 (2000)	5058 (1794)	5232 (1641)	−96 (1344)	567 (1465)	−373 (1359)	−47 (1769)	−770 (−1593 to 53)	−418 (−1275 to 439)
Medium to highstride rate (minutes/day)	132 (45)	130 (54)	131 (60)	143 (53)	117 (45)	124 (43)	−3 (41)	14 (38)	−15 (40)	−5 (49)	−19 (−42 to 3)	−14 (−36 to 8)
High stride rate (minutes/day)	53 (23)	50 (24)	55 (37)	62 (29)	50 (26)	55 (26)	1 (25)	11 (21)	−3 (22)	5 (26)	−12 (−27 to 3)	−9 (−24 to 7)
Inactive (minutes/day)	343 (100)	356 (102)	362 (101)	354 (79)	390 (91)	376 (82)	19 (62)	−3 (68)	45 (57)	4 (88)	12 (−25 to 49)	37 (−8 to 81)

Exp, experimental group; Con, control group.
[a] Adjusted for baseline values and adjusted for age, sex, selective motor control and sport frequency of the immediate family if necessary.

Table 3
Group averages for parent-reported physical activity for each group, difference within groups, and difference between groups.

Outcome	Groups						Difference within groups				Difference between groups[a]	
	Baseline		Month 6		Month 12		Month 6 minus baseline		Month 6 minus baseline		Month 6 minus baseline	Month 12 minus baseline
	Exp (n = 23)	Con (n = 23)	Exp (n = 21)	Con (n = 2)	Exp (n = 22)	Con (n = 21)	Exp (n = 21)	Con (n = 22)	Exp (n = 22)	Con (n = 21)	Exp minus Con	Exp minus Con
Moderate to vigorous activity, Median (IQR) (minutes/wk)	365 (110–490)	400 (160–635)	780 (303–1093)	317 (71–668)	378 (179–623)	360 (215–590)	300 (−8 to 817)	7 (−213 to 331)	−28 (−123 to 176)	30 (−135 to 206)	2.2[b] (1.1 to 4.4)[c]	1.1[b] (0.7 to 1.8)
Sports club participation, n yes (%)	8 (35)	15 (65)	9 (43)	15 (68)	10 (48) (n = 21)	16 (76)	1 (5)	2 (9)	3 (14) (n = 21)	3 (14)	1[d] (0 to 6)	1[d] (0 to 3)
Physical activity norm, n yes (%)	4 (17)	5 (22)	9 (43)	6 (27)	2 (9)	3 (14)	7 (33)	3 (14)	0 (0)	3 (14)	2.19[d] (0.58 to 8.24)	0.60[d] (0.09 to 4.14)
Inactive, mean (SD), (minutes/wk)	2653 (1065)	2659 (812)	2199 (996)	2170 (1320)	2223 (944)	2783 (1058)	−495 (1047)	−564 (1341)	−508 (934)	181 (1354)	140[e] (−550 to 831)	−494[e] (−1099 to 111)

Exp, experimental group; Con, control group.
[a] Adjusted for baseline values and adjusted for age, sex, selective motor control and sport frequency of the immediate family if necessary.
[b] Change ratio, based on linear regression analysis after logarithmic transformation.
[c] Positive trend in favour of the intervention group ($p = 0.04$).
[d] Odds ratio, based on binary logistic regression analysis.
[e] Mean difference (95% CI).

Table 4
Mean (SD) for outcomes for each group, mean (SD) difference within groups, and mean (95% CI) difference between groups.

Outcome	Groups								Difference within groups						Difference between groups [a]		
	Baseline		Month 4		Month 6		Month 12		Month 4 minus baseline		Month 6 minus baseline		Month 12 minus baseline		Month 4 minus baseline	Month 6 minus baseline	Month 12 minus baseline
	Exp (n = 23)	Con (n = 23)	Exp (n = 23)	Con (n = 21)	Exp (n = 23)	Con (n = 22)	Exp (n = 23)	Con (n = 21)	Exp (n = 23)	Con (n = 21)	Exp (n = 23)	Con (n = 22)	Exp (n = 23)	Con (n = 21)	Exp minus Con	Exp minus Con	Exp minus Con
GMFM-66 (0–100)	77 (14)	80 (14)	–	–	79 (13)	79 (14)	79 (14)	82 (14)	–	–	1.7 (4.5)	– 1.4 (4.2)	1.2 (4.4)	2.0 (3.1)	–	2.8[b] (0.2 to 5.4)	−0.9 (−3.3 to 1.4)
One-minute walk test (m)	86 (20)	92 (20)	92 (22)	94 (20)	92 (25)	96 (17)	91 (25) (n = 22)	93 (19) (n = 20)	6 (7)	1 (9)	6 (11)	3 (9)	5 (11) (n = 22)	2 (10) (n = 20)	5[b] (0 to 9)	2 (−4 to 9)	3 (−43 to 10)
Functional strength (repetitions)	43 (16)	42 (18)	48 (18)	48 (22)	51 (20) (n = 20)	53 (21)	53 (18) (n = 21)	56 (22)	4 (8)	4 (8)	9 (10) (n = 20)	10 (9)	9 (8) (n = 21)	13 (9)	0 (−5 to 5)	0 (−5 to 5)	−4 (−9 to 2)

Exp, experimental group; Con, control group; GMFM-66, Gross Motor Function Measure-66.

[a] Differences between groups are adjusted for baseline values. The intervention effect was not substantially confounded by age, sex, or selective motor control.

[b] Positive trend in favour of the intervention group (GMFM-66: $p = 0.03$; one-minute walk test: $p = 0.06$).

Table 5
Median (IQR) of each group, median (IQR) within each group and difference between groups (Mann–Whitney *U* test, *p*-value) for the children's attitudes towards sports, and mean (SD) of each group, mean (SD) within each group, and (95% CI) difference between groups for the parents' attitudes towards sports.

Outcome	Groups						Difference within groups				Difference between groups [a]	
	Baseline		Month 6		Month 12		Month 6 minus baseline		Month 12 minus baseline		Month 6 minus baseline	Month 12 minus baseline
	Exp	Con	Exp	Con	Exp	Con	Exp	Con	Exp	Con	Exp minus Con	Exp minus Con
Children's attitudes	(n = 23)	(n = 23)	(n = 21)	(n = 22)	(n = 21)	(n = 21)	(n = 21)	(n = 22)	(n = 21)	(n = 21)		
Attitudes towards advantages[b] (1–5)	4.0 (3.7–4.4)	4.0 (3.7–4.6)	4.0 (3.6–4.5)	4.0 (3.4–4.3)	4.1 (3.6–4.4)	3.9 (3.5–4.5)	0.14 (−0.29 to 0.5)	−0.21 (−0.64 to 0)	0.14 (−0.14 to 0.64)	−0.14 (−0.86 to 0.14)	$p = 0.08$	$p = 0.04$[c]
Attitudes towards disadvantages[b] (1–5)	3.5 (3.0–3.8)	3.8 (3.5–4.2)	3.7 (3.1–4.2)	3.7 (3.1–4.2)	3.7 (3.3–4.1)	4.0 (3.6–4.3)	0.33 (−0.42 to 0.58)	−0.25 (−1.0 to 0.33)	0.17 (−0.5 to 0.83)	0 (−0.38 to 0.42)	$p = 0.02$[d]	$p = 0.56$
Parents' attitudes	(n = 20)	(n = 22)			(n = 20)	(n = 22)			(n = 20)	(n = 22)		
Importance of sport[b] (1–5)	4.13 (0.60)	4.22 (0.58)			4.07 (0.56)	4.19 (0.46)			−0.05 (0.60)	−0.03 (0.53)		−0.08 (−0.36 to 0.21)
Opinion towards accessibility of sports facilities[b] (1–5)	3.40 (0.48)	3.47 (0.50) (n = 18)			3.34 (0.67)	3.48 (0.67) (n = 18)			−0.06 (0.62)	0.01 (0.71) (n = 18)		−0.11 (−0.53 to 0.31)

Exp, experimental group, Con, control group.

[a] Adjusted for baseline values.

[b] A higher score reflects a more positive attitude towards sports.

[c] Positive trend in favour of the intervention group ($p = 0.04$).

[d] Significant intervention effect in favour of the intervention group ($p < 0.025$).

of sports at 12 months ($p = 0.04$) but not at 6 months, and a borderline significant greater disagreement with possible disadvantages of sports at 6 months ($p = 0.02$) but not at 12 months.

Discussion

There was no significant effect of the intervention on physical activity, so the hypothesis that counselling, home-based physiotherapy and fitness training would work synergistically to improve physical activity could not be confirmed. This was against our expectations, previous studies in cerebral palsy showed (non-significant) positive trends towards improving physical activity in children and adolescents with cerebral palsy after either counselling,[11] or fitness training only.[9] Nevertheless, the present findings are in agreement with research involving typically developing children where evidence is equivocal. No evidence has been found for the effectiveness of family-based and community-based physical activity interventions that combine exercise programs with the provision of information.[29] Another review has pointed out that physical activity among typically developing children can be increased by means of school-based interventions.[30] The authors of that review indicated that the highest-quality studies with positive effects on physical activity were characterised by a multicomponent intervention (education, focus on behavioural change and involvement of parents) and a minimum intervention duration of one school year. Therefore, it is possible that our 6-month program was too short to elicit changes in such a complex behaviour as physical activity. Whether a longer counselling period, with periodical attention to physical activity, may be needed to improve physical activity in children with cerebral palsy should be examined in further research.

Another explanation for the intervention's lack of effect on physical activity might be insufficient contrast between groups, which could arise from three possible sources. First, the families who chose to participate in the study were likely to be more interested in (increasing) physical activity than those who refused to participate, as illustrated by the parents' already very positive attitude towards sports in both groups. This selection bias may have resulted in all families in the study stimulating physical activity of their children regardless of group allocation. Second, physiotherapists participating in the study were interested in fitness training and physical activity stimulation. Possibly, they (unintentionally) changed the content of the physiotherapy treatment for the control group towards a more pro-active approach, similar to the intervention. Third, the fact that all participants were informed about the aim, relevance and content of the study (for example, increasing physical activity) and that they had to wear an activity monitor and register physical activity might have raised awareness of the importance of physical activity.

The two measures of physical activity demonstrated contrasting results: there was no change for walking activity assessed with the StepWatch™, but there was a positive trend for the parent-reported physical activity assessed with the AQuAA. This might be explained by the different constructs underlying the StepWatch™ and AQuAA assessments. The StepWatch™ objectively measures real-time stride rate during daily walking activities, but does not provide information about other types of activities performed. The AQuAA covers a wide range of activities and may have captured an increase in activities not registered by the StepWatch™. However, self-reports are prone to recall bias and socially desired answering.[31] Socially desired answering may be particularly likely to occur in the intervention group, because they received the physical activity stimulation program. Previous studies that compared the AQuAA to accelerometry,[19] or compared other objective and subjective physical activity measures in typically developing children, found low agreement between the methods, suggesting that these measures are not interchangeable.[32] This indicates that the assessment of physical activity remains challenging.

Since changing physical activity behaviour is a complex process, evaluating the effect of this multi-component physical activity stimulation program on other outcomes may provide valuable information. Because the fitness training incorporated gross motor activities, and the home-based physiotherapy was focused on practising mobility activities in the home, we expected that mobility capacity would improve. Although no significant effects of intervention were demonstrated, the positive trend for gross motor capacity, which is a highly relevant outcome measure in this population, shows that this home-based activity approach may have potential for improving activity capacity. The 2.8-point increase in GMFM-66 scores in favour of the intervention group seems substantial, since it exceeds the minimum clinical important difference reported by Oeffinger et al[33] No conclusions could be drawn about which component of the intervention was responsible for this observed positive trend. However, it is most likely that the individually tailored home-based physiotherapy and the strong focus on gross motor activities in the fitness training contributed to this trend. This is supported by the positive trend found for the 1-minute walk test, directly after ending the fitness program. Although two components of the program may have potential to improve mobility capacity, the added value of improving mobility capacity for increasing physical activity remains unclear. This should be the subject of future research.

An explanation for not demonstrating an intervention effect on fitness and self-reported fatigue might be the scheduled reduction in fitness training frequency to once a week in the third and fourth month of the training period. The reduction was planned to limit the burden on parents and children, and to allow the children to develop physical activities in order to create a transitional period between the organised fitness training and self-developed activities. Since sports club participation did not improve after the physical stimulation program, it is likely that children did not succeed in initiating further physical activities, resulting in insufficient training volume to elicit a significant fitness improvement. However, the beneficial effect of a higher fitness training volume on physical activity is not yet clear. A previous 9-month fitness training program of four times per week only resulted in a positive trend in physical activity, despite an effect on fitness.[9]

The short-term improvement in the children's attitudes towards the disadvantage of sports, and the long-term trend for improving the children's attitudes towards the advantages of sports are promising, considering the lack of effect previously found on the attitude of adolescents with cerebral palsy after counselling.[11] However, the small effect sizes for attitude towards sports in our population, which is already very positive about sports, weaken the clinical relevance of these improvements. Socially desired answering might also have influenced this subjective measure. This is supported by the lack of effect on physical activity or sports participation, which was expected to increase by a more positive attitude.[34] It is possible that the improvement in attitude towards sports was insufficient to improve physical activity. Also, environmental barriers, such as lack of transportation and availability of facilities,[35] may have restricted starting up (sports) activities despite small improvements in attitude. Future studies aimed at improving physical activity should assess the presence of environmental barriers and systematically examine whether influencing these barriers contributes to a more active lifestyle.

An important study limitation is that it was not possible to draw any conclusion about the effectiveness of the separate components of the intervention. More insight into the contribution of the separate components of the program is needed, in order to understand how they influence physical activity, by varying one component at the same time. Another limitation is that the variability of the activity monitor data was higher than anticipated, so study power was less than planned. However, based on the results for the activity monitor, it is unlikely that a larger sample size would have resulted in a positive intervention effect for walking activity.

A strength of this study was the location of the program in the children's homes, in paediatric physiotherapy practices or special schools for children with disabilities. While different characters, motivational skills and training facilities might have influenced the effects of training, this variety increases the generalisability of our results to other paediatric practices.

In conclusion, a physical activity stimulation program combining counselling through motivational interviewing, home-based physiotherapy and fitness training was not effective for increasing children's physical activity, or improving mobility capacity, fitness, fatigue, and attitude towards sports. Further research should be performed to determine the separate contribution of each component of the program for improving physical activity.

What is already known on this topic: Children with cerebral palsy have lower levels of physical activity and fitness compared to their typically developing peers. Physical activity patterns may persist into adolescence and adulthood. Exercise programs can improve the fitness of children with cerebral palsy. Studies of interventions to promote physical activity in this population have shown favourable, but non-significant, trends.
What this study adds: A physical activity stimulation program consisting of fitness training, counselling and home-based therapy was not effective in children with cerebral palsy. Although the program improved the children's attitude to sports, the effect was small.

Footnotes: [a] StepWatch™ Activity Monitor 3.0, Orthocare Innovations, Seattle, USA. [b] MicroFet dynamometer, Biometrics, Almere, The Netherlands. [c] Corival V2 Lode B.V., Groningen, The Netherlands. [d] Cosmed, Rome, Italy.

eAddenda: Tables 6 and 7 can be found online at doi:10.1016/j.jphys.2013.12.007

Ethics: The Medical Ethical Board of the VU University Medical Center, Amsterdam, approved this study. Parents and children aged 12 years and over gave written informed consent before data collection began.

Competing interests: Nil. Grant providers were not involved in the design of the study, data collection, data analysis, manuscript preparation and publication decisions.

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