Exercise Training Program in Children and Adolescents With Cerebral Palsy

A Randomized Controlled Trial

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Objective: To evaluate the effects of an 8-month training program with standardized exercises on aerobic and anaerobic capacity in children and adolescents with cerebral palsy.

Design: Pragmatic randomized controlled clinical trial with blinded outcome evaluation between July 2005 and October 2006.

Setting: Participants were recruited from 4 schools for special education in the Netherlands.

Participants: A total of 86 children with cerebral palsy (aged 7-18 years) classified at Gross Motor Function Classification System level I or II.

Intervention: Participants were randomly assigned to either the training group (n=32) or the control group (n=33). The training group met twice per week for 45 minutes to circuit train in a group format that focused on aerobic and anaerobic exercises.

Main Outcome Measures: Aerobic capacity was assessed by the 10-m shuttle run test, and anaerobic capacity was assessed by the Muscle Power Sprint Test. Secondary outcome measures included agility, muscle strength, self-competence, gross motor function, participation level, and health-related quality of life.

Results: A significant training effect was found for aerobic (P<.001) and anaerobic capacity (P=.004). A significant effect was also found for agility (P<.001), muscle strength (P<.001), and athletic competence (P=.005). The intensity of participation showed a similar effect for formal (P<.001), overall (P=.002), physical (P=.005), and skilled-based activities (P<.001). On the health-related quality of life measure, a significant improvement was found for the motor (P=.001), autonomy (P=.02), and cognition (P=.04) domains.

Conclusions: An exercise training program improves physical fitness, participation level, and quality of life in children with cerebral palsy when added to standard care.

Trial Registration: isrctn.org Identifier: ISRCTN77274716


Cerebral Palsy (CP) describes a group of disorders that affect the development of movement and posture, causing activity limitation, and are attributed to nonprogressive disturbances that occurred in the developing fetal or infant brain.1 The motor disorders of CP are often accompanied by disturbances of sensation, cognition, communication, perception, and/or behavior and/or by a seizure disorder.1 Improving the ability to walk or perform other functional activities are often the primary therapeutic goals for children with CP.2 Because of existing impairments, many children and adolescents with CP have difficulty with activities such as walking independently, negotiating stairs, running, or navigating safely over uneven terrain.3 Additionally, children with CP have distinctly subnormal aerobic and anaerobic capacity in comparison with typically developing peers.4-6 Also, muscle mass is low,4 and muscle strength is reduced.7-9 Low levels on these fitness components may contribute to the difficulties in motor activities most children with CP encounter in daily life. Moreover, evidence suggests that hypoactive children are more likely to become physically sedentary adults and that encouraging the development of physical activity habits in children will help establish activity patterns that continue into adulthood.10

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In general, aerobic capacity, anaerobic capacity, and muscle strength can be trained in typically developing children of...
Studies of the effects of lower limb exercises in children with CP have demonstrated an increase in muscle strength and function, \(^\text{7,14-16}\) as well as in aerobic capacity, \(^\text{17,18}\) while no negative effects on spasticity or abnormal movements have been reported. \(^\text{14,15,19,20}\) To date, none of the training programs in children with CP have focused on anaerobic capacity, which is important because almost all daily childhood activities are more short-term (<15 seconds) and high intensity than long-term. \(^\text{21,22}\)

Activities of daily childhood life consist of well-balanced aerobic, anaerobic, and muscle strength components. The principle of specificity of learning states that learning is optimized by practice that approximates the target skill. \(^\text{23}\) To date, we know of no study that trained the aerobic and anaerobic fitness components as well as muscle strength in task-specific, functional activities in children with CP. Therefore, we conducted a pragmatic randomized controlled clinical trial to determine the effects of a standardized 8-month exercise program with an extra 4 months of follow-up after the training.

Accordingly, the primary objective of this study was to determine the specific (ie, aerobic and anaerobic capacity) effects of an exercise program in addition to usual care at the end of the training period. Moreover, we investigated the effects of this program on all International Classification of Functioning, Disability, and Health (ICF) levels and health-related quality of life (HRQOL) at the end of the training period and at 4 months' follow-up.

## METHODS

### PARTICIPANTS

Children and adolescents with CP were recruited from 4 schools for special education in the Netherlands. To be included, participants had to be 7 to 20 years of age, diagnosed with spastic CP, and classified at level I or II on the Gross Motor Function Classification System (GMFCS). \(^\text{24}\) Children older than 12 years were classified using the same criteria as those used for 6- to 12-year-olds. All children in the study were able to follow simple verbal commands. They were all receiving rehabilitation services at the time of the study. Participants were excluded if they had had orthopedic surgery or neurosurgery and/or botulinum toxin injection(s) within 6 months prior to study entry or cardiac or respiratory conditions that could negatively be affected by exercise.

### STUDY DESIGN

A pragmatic randomized controlled clinical trial was conducted between June 2005 and October 2006. The Institutional Ethics Committee of the University Medical Center Utrecht approved the study. Participating schools for special education were informed about the study and the inclusion and exclusion criteria. Based on clinical examination, pediatric physiatrists working in these schools referred suitable participants.

The Dutch translation of the GMFCS \(^\text{25}\) was used to classify the children with CP into 2 groups based on their functional ability. Participants were randomly assigned to 2 groups using a 4-block randomization protocol. Each block represented all participants from 1 school. The groups within each block consisted of children classified at level I or II on the GMFCS. \(^\text{25}\) From each block and group, every participant was randomly allocated to the training group or the control group. An independent off-site researcher not involved in the assessments used a concealed method for allocation. The children in the control group received their usual rehabilitation care. Because this might affect functional gains among participants, provided care was tracked from the medical progress records. Usual care ranged from no treatment to various therapeutic approaches. There was no difference in usual care between both groups.

### OUTCOME MEASURES ACCORDING TO THE INTERNATIONAL CLASSIFICATION OF FUNCTIONING, DISABILITY, AND HEALTH

Primary outcome measures were aerobic and anaerobic capacity measured at the end of the training period. Additional measurements included agility, muscle strength, body mass index, self-perception, gross motor function, participation, and HRQOL.

#### Body Function

Aerobic capacity was reflected by the achieved level on the 10-m shuttle run test. \(^\text{26}\) This test requires children to walk or run between 2 markers delineating the respective course of 10 m at a set incremental speed determined by a signal (every minute). The achieved level was recorded and used for analysis. Anaerobic capacity was measured using mean power (measured in watts) derived from the Muscle Power Sprint Test. \(^\text{27}\) For the Muscle Power Sprint Test, the subjects were instructed to complete six 15-m runs at a maximum pace. Between each run, the subject was allowed a timed 10-second rest. Power output (measured in watts) for each sprint was calculated. Agility was assessed by the 10 × 5-m Sprint Test. \(^\text{27}\) Muscle strength of the lower extremities was measured with the 30-second repetition maximum. \(^\text{28}\)

The body mass index was calculated as weight in kilograms divided by height in meters squared. Participants’ weight and height were measured using a standard protocol. Each child
was weighed to the nearest 100 g on electronic scales (Seca, Hamburg, Germany). Height measurements were taken to the nearest 0.5 cm while the child was standing with his or her back against a wall.

We used the Self-Perception Profile for Children\(^\text{29}\) to evaluate the self-concept of the children. This scale is designed to assess children’s perceptions of themselves. We assessed the domains of athletic competence, physical appearance, and global perception of their worth or esteem as a person.

Activity

In this study, gross motor function was assessed using dimensions D and E of the Gross Motor Function Measure (GMFM),\(^\text{30}\) which measures activities in a standing position and walking, running, and jumping, respectively. These dimensions were chosen because they represent areas that many young people with CP who are able to walk have difficulty with.\(^\text{7}\)

Participation

The Children’s Assessment of Participation and Enjoyment (CAPE)\(^\text{31}\) was used to document change in how children and youth participate in everyday activities outside mandated school activities. The CAPE provides 3 levels of scoring: (1) overall participation scores; (2) domain scores reflecting participation in formal and informal activities; and (3) scores reflecting participation in 5 types of activity (recreational, active physical, social, skill-based, and self-improvement activities). The intensity scores reflect the average amount of time that a child spends participating in different activities. The intensity scores of all types of activity were measured.

Health-Related Quality of Life

The TNO-AZL Questionnaire for Children’s Health-Related Quality of Life (TACQOL) is a generic, multidimensional instrument.\(^\text{32}\) It asks about health problems in the past few weeks using a 3-point Likert scale and about the emotional response to these problems on a 4-point scale. This instrument contains 7 scales: (1) pain and symptoms, (2) basic motor functioning, (3) autonomy, (4) cognitive functioning, (5) social functioning, (6) global positive emotional functioning, and (7) global negative emotional functioning. We used the TACQOL Parent Form (TACQOL-PF) because we expected that a part of the participants were too young or too low functioning to complete the TACQOL themselves.

To reduce bias, 8 assessors who were not the treating therapist and who were blinded for the treatment modality undertook the testing without review of previous scores. Prior to data collection, the assessors had formal training and were given written instructions in the application and scoring of all tests and measurements.

Assessments were performed at baseline, after 4 months, and directly after the 8 months of the intervention period in both groups. There was a follow-up assessment with the same measures in both groups at 12 months after baseline.

STATISTICAL ANALYSIS

The sample size for the trial was determined by the most demanding hypothesis to detect effects of treatment on aerobic capacity. The sample size was calculated from data for the 10-m shuttle run test,\(^\text{34}\) with a significance level of .05, a d of 0.65, and a power of 80%. Therefore, 30 participants were required for each group. To compensate for dropouts, we planned to enroll 35 patients per group.

All data analyses were carried out according to a preestablished analysis plan and were performed according to the intention-to-treat principle. To assess the effects of training as compared with standard care, we used repeated-measures analysis of variance (group [2] × time [3]) with repeated aerobic and anaerobic capacity measures as dependent variables and treatment group as an independent variable. We used the same analysis to assess our secondary objective. In case of violation of the sphericity assumption, we used the Greenhouse-Geisser correction.

To assess the effects during and after the fitness program as compared with control treatment effects, repeated-measures analysis of variance (group [2] × time [4]) with repeated contrasts was used. Violations of assumptions were checked in all cases.

The α level was set at .05. Data were analyzed using SPSS, version 13.0 (SPSS Inc, Chicago, Illinois).

RESULTS

Age-eligible participants were recruited from July 2005 to September 2005. Figure 1 shows the participant flow from initial recruitment to assessment after training and follow-up (October 2006). A total of 86 participants were assessed for eligibility for the study. One of the children did not meet the inclusion criteria, 9 of the eligible participants did not take part for logistical reasons (year of

Figure 1. Flow of participants through the trial. T0 indicates baseline; T1, after 4 months; T2, after 8 months.
their graduation), and 8 children and adolescents refused to participate for unknown reasons. Sixty-eight patients and their parents gave written informed consent before entering the study and were randomized to either the control group or the training group. Baseline demographic and clinical characteristics of each group are listed in Table 1. At baseline, there were no significant differences between study groups. During baseline measurements, 3 children (boys aged 11.3, 15.1, and 16.1 years) discontinued the study because of personal reasons, such as lack of motivation, and were lost to follow-up. These children completed the fitness measures but did not complete most of the other measures. Sixty-five participants completed the entire study. The median attendance in exercise training was 56 of 60 sessions (93%), with all children attending at least 85% of the training sessions. During a training session, 1 child fell and fractured her radius; she missed 4 training sessions because she was wearing a cast.

EFFECTS OF TRAINING

Body Function

As shown in Figure 2, improvements on aerobic (+38%) and anaerobic capacity (+25%) were found for the training group. Moreover, as shown in Table 2, improvements were found on agility (+15%), muscle strength of the lower extremities (left and right side, +20% and +23%, respectively), and athletic competence on the Self-Perception Profile for Children (+11%).

Activity and Participation

A positive training effect was present for dimension D (standing) of the GMFM. For participation, there were significant differences in favor of the training group on the overall, formal, physical, and skill-based activities of the CAPE.

Health-Related Quality of Life

For HRQOL, significant changes over time that differed by group were found for the motor, autonomy, and cognition domains of the TACQOL-PF (Table 2). The contrasts between all measurements (at baseline, 4 months, and 8 months) show that during the first 4 months of the training period (focused on aerobic capacity) there was a significant change in favor of the training group for aerobic capacity, agility, athletic competence, and GMFM dimension D (standing). Moreover, a similar change was also found for overall, formal, physical, skill-based, and self-improvement activities as measured with the CAPE. On the TACQOL-PF, significant changes in favor of the training group were found for the motor, autonomy, cognition, and social domains during the first 4 months. No significant effects were observed for muscle power.

During the last 4 months of the training period (focused on anaerobic capacity), we found an improvement in the training group for aerobic capacity, anaero-
bic capacity, agility, muscle strength, and GMFM dimension E (walking, running, and jumping). There was no improvement on the CAPE. On the TACQOL-PF, the only significant change in favor of the training group was for the autonomy domain.

FOLLOW-UP EFFECTS AT 12 MONTHS

At follow-up (4 months after the training period), we noted a significant difference in outcome measures, as measured with repeated contrasts between groups. The training group reached levels that were similar to the levels after 4 months of training; there was a decrease in aerobic capacity (−8.4%), anaerobic capacity (−8.5%), agility (−4.3%), muscle strength (left side, −4.4%; right side, −8.3%), and athletic competence (−9.8%). The control group remained stable on these measures. Significant differences between groups were found at follow-up on dimension D (−1.1% for the training group) of the GMFM and on the skill-based activities of the CAPE (−20% for the training group). These measures remained stable in the control group. For HRQOL, there were no significant differences between groups at follow-up.

To our knowledge, this is the first randomized multi-center clinical trial studying the effects of an exercise program with emphasis on movements of daily childhood life, nowadays called “functional exercises,” combining 3 fitness elements (aerobic and anaerobic capacity and muscle strength) in a large number of participants with CP. This study provides evidence that a group circuit-training program can be an effective and feasible strategy for increasing both aerobic capacity and anaerobic capacity in young people with CP.

To date, only 1 randomized controlled study investigating the efficacy of aerobic training in children with CP has been published. After an aerobic training program of 9 months, observed improvements were 35% (4 sessions a week) and 21% (2 sessions a week) in aerobic capacity and there were no training-related improvements in anaerobic capacity. Our study showed improvements of 38% in aerobic capacity and significant improvements of 11% in anaerobic capacity in the second 4 months of the exercise program when anaerobic exercises were incorporated.

The main focus during the last 4 months of the fitness program was anaerobic capacity. The significant interaction in favor of the training group during the last 4 months of training shows that a fitness training program with a predominantly anaerobic nature can improve the anaerobic capacity of children with CP. These improvements also led to significant changes in muscle strength (22%). Other studies focusing specifically on improving muscle strength have reported similar results after training in children with CP.7,11-16

Children with CP often have difficulties changing direction of the body abruptly or quickly shifting the direction of movement without losing balance (agility). During both training periods, with different main foci (aero-

<table>
<thead>
<tr>
<th>Difference T0-T2, Mean (SD)</th>
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<tbody>
<tr>
<td>Training Group</td>
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<tr>
<td>----------------</td>
</tr>
<tr>
<td>BMI 0.7 (2.1)</td>
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<tr>
<td>Aerobic capacity: level on 10-m shuttle run test, min 2.4 (1.9)</td>
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<tr>
<td>Anaerobic capacity: mean power, W 20.4 (38.0)</td>
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<tr>
<td>Agility: 10 × 5-m Sprint Test −4.5 (4.1)</td>
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<tr>
<td>Muscle strength left 6.9 (7.2)</td>
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<tr>
<td>Muscle strength right 7.7 (9.0)</td>
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<tr>
<td>Self-concept (SPPC)²⁰ Athlete performance 2.0 (4.2)</td>
</tr>
<tr>
<td>Physical appearance 0.03 (4.4)</td>
</tr>
<tr>
<td>Global perception of worth 0.5 (3.3)</td>
</tr>
<tr>
<td>Gross motor function (GMFM)²⁰ Dimension D (standing) 2.6 (5.4)</td>
</tr>
<tr>
<td>Dimension E (walking, running, and jumping) 1.5 (6.4)</td>
</tr>
<tr>
<td>Participation (CAPE)²¹ Overall activities 0.0 (0.5)</td>
</tr>
<tr>
<td>Formal activities 0.2 (0.4)</td>
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<tr>
<td>Informal activities 0.0 (0.7)</td>
</tr>
<tr>
<td>Recreational activities −0.2 (1.0)</td>
</tr>
<tr>
<td>Physical activities 0.3 (0.8)</td>
</tr>
<tr>
<td>Social activities −0.1 (0.8)</td>
</tr>
<tr>
<td>Skill-based activities 0.2 (0.5)</td>
</tr>
<tr>
<td>Self-improvement activities −0.1 (0.9)</td>
</tr>
<tr>
<td>Health-related quality of life (TACQOL-PF)²⁰ Pain and symptoms −0.4 (3.8)</td>
</tr>
<tr>
<td>Basic motor functioning 2.1 (4.3)</td>
</tr>
<tr>
<td>Autonomy 0.5 (4.3)</td>
</tr>
<tr>
<td>Cognitive functioning −0.9 (4.7)</td>
</tr>
<tr>
<td>Social functioning 0.7 (4.0)</td>
</tr>
<tr>
<td>Global positive emotions 0.3 (3.9)</td>
</tr>
<tr>
<td>Global negative emotions 0.7 (2.9)</td>
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</tbody>
</table>

Abbreviations: BMI, body mass index (calculated as weight in kilograms divided by height in meters squared); CAPE, Children’s Assessment of Participation and Enjoyment; GMFM, Gross Motor Function Measure; SPPC, Self-Perception Profile for Children; T0, baseline; T2, after 8 months; TACQOL-PF, TNO-AZL Questionnaire for Children’s Health-Related Quality of Life Parent Form.

aP values are for the repeated-measures analysis of variance (group [2] × time [3]).
Another randomized study demonstrated an increase in self-concept of the children after training. Possible explanations for these differences might be explained by differences in settings, the illness severity of the children included in either study, the length of the exercise program, and the type of exercises. These and other possible factors need to be studied.

Participation in everyday childhood activities plays an important role in the development of a child’s social relationships and skills and also influences long-term mental and physical health. Participation enables children to explore their social, intellectual, emotional, communicative, and physical potential and is an important predictor of future life satisfaction. Margalit and Stevenson et al looked at children with CP and found significant limitations in participation. Law et al identified several child factors that affect participation, including that higher levels of physical function are associated with increased intensity of participation. Therefore, it could be expected that involvement in an exercise program might lead to more permanent changes in everyday physical activities that in turn might lead to maintenance of the fitness benefits. Based on the findings of this study, we conclude that having a higher physical function does lead to an increased intensity of participation. Overall, formal, physical, and skill-based activities were improved at the end of the training period. Unfortunately, most of these improvements were not maintained at follow-up. In future studies, factors that may contribute to a positive long-term change in lifestyle habits should be taken into account.

Health-related quality of life is an important outcome of treatment for chronic conditions such as CP. This is, to our knowledge, the first study that assessed the effects of an exercise intervention program on the quality of life in children with CP. Improvements in HRQOL in favor of the training group were seen for 3 domains. The parents of the children with CP noticed improvements for the motor, autonomy, and cognition domains.

The amount of time spent in physical activity has decreased markedly over the past decades. Our study shows aerobic and anaerobic capacity, agility, and muscle strength decreased during the intervention period in the control group. Given this decline in fitness levels, children and adolescents with CP are at risk of a sedentary lifestyle that will continue throughout their lives. The adverse effects of this decreased fitness may be reduced with involvement in a fitness program.

The findings of this study suggest that the benefits that children gained during training were only partially maintained at follow-up. A decrease in fitness after an exercise training program in children with CP was previously reported. It seems very difficult for children with CP to maintain the gains of an exercise program. Consequently, it can be concluded that children with CP must continue their training to maintain their fitness levels. Limited exercise and sport opportunities in their neighborhoods might impede transfer of the gains in fitness into extra participation in regular physical activities. Supervised training programs at schools or in the community might provide fitness training for children and adolescents with CP. These possibilities need to be studied in future research.

An 8-month, standardized, functionally based exercise program significantly improved physical fitness, the intensity of activities, and HRQOL in children with CP when added to standard care.

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CONCLUSIONS

An 8-month, standardized, functionally based exercise program significantly improved physical fitness, the intensity of activities, and HRQOL in children with CP when added to standard care.

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