A lifestyle intervention improves fatigue, mental health and social support among adolescents and young adults with cerebral palsy: focus on mediating effects

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Abstract
Objective: To evaluate the effect of a lifestyle intervention on fatigue, participation, quality of life, gross motor functioning, motivation, self-efficacy and social support, and to explore mediating effects of physical behavior and physical fitness.
Design: A randomized controlled trial with intention to treat analysis.
Setting: Rehabilitation centers in university hospitals in the Netherlands.
Subjects: Adolescents and young adults with spastic cerebral palsy.
Interventions: A six-month lifestyle intervention that consisted of physical fitness training combined with counseling sessions focused on physical behavior and sports participation.
Main measures: Fatigue, social participation, quality of life and gross motor functioning.
Results: The lifestyle intervention was effective in decreasing fatigue severity during the intervention (difference = –6.72, p = 0.02) and in increasing health-related quality of life with respect to bodily pain (difference = 15.14, p = 0.01) and mental health (difference = 8.80, p = 0.03) during follow-up. Furthermore, the domain participation and involvement of the social support increased during both the intervention (difference = 5.38, p = 0.04) and follow-up (difference = 4.52, p = 0.03) period. Physical behavior or physical fitness explained the observed effects for 22.6%, 9.7% and 28.1% of improvements on fatigue, bodily pain and mental health, but had little effect on social support (2.6%).
Interpretation: Fatigue, bodily pain, mental health and social support can be improved using a lifestyle intervention among adolescents and young adults with cerebral palsy. Furthermore, substantial mediating effects were found for physical behavior and physical fitness on fatigue, bodily pain and mental health.

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Keywords
Participation (WHO ICF), cerebral palsy, fatigue, motor performance, quality of life

Trial registration
Nederlands Trial Register; NTR1785.

Received: 7 January 2014; accepted: 20 September 2014

Introduction
Increasing exercise and physical activity in adolescents and young adults with cerebral palsy is important because they are known to have unfavorable physical behavior profiles, consisting of low physical activity, high sedentary time and subnormal physical fitness. These unfavorable factors are associated with adverse effects on medical state and can affect perceived level of fatigue, social participation, quality of life and gross motor functioning. Because physical behavior and physical fitness are modifiable, increases may positively affect levels of fatigue, social participation, health-related quality of life and gross motor functioning. Furthermore, personal and environmental factors, such as self-efficacy, intrinsic motivation and social support, for exercise behavior are likely to be linked with changes in physical behavior and physical fitness among persons with physical disabilities. However, there is only little evidence on the effectiveness of interventions to modify physical behavior and physical fitness among adolescents and young adults with cerebral palsy and therefore, persons with cerebral palsy tend not to receive regular treatment in this area.

The Learn2Move 16–24 study evaluates effects of the active lifestyle and sports participation intervention in adolescents and young adults with spastic cerebral palsy. This lifestyle intervention aims to optimize participants’ physical behavior and increase physical fitness levels. Effects of the intervention on these primary outcomes are presented in previous publications. The current study focuses on the intervention’s secondary effects on fatigue, participation, health-related quality of life and gross motor functioning. Also, additional changes of self-efficacy, intrinsic motivation and Social Support for Exercise Behaviour were studied. In case of significant effects of the active lifestyle and sports participation lifestyle intervention for any of these secondary outcome measures, the mediating effects of physical behavior and physical fitness on this specific outcome measure were explored.

Methods
The study used a multicenter, single blind, randomized controlled design. The measurements were performed by assessors who were blind for group allocation. The study design has been described in detail elsewhere.

Setting and participants
To determine eligibility, we reviewed health records at four rehabilitation centers and two rehabilitation departments at university hospitals throughout the western and central regions of the Netherlands. Persons with spastic cerebral palsy were eligible if they met each of the following inclusion criteria: (1) age 16 to 24 years; and (2) Gross Motor Functioning Classification System (GMFCS) Level I to IV.

Persons were excluded if they had any of the following: (1) disabilities other than cerebral palsy that affect daily physical activity or cardiopulmonary fitness; (2) contraindication to (maximal) exercise; (3) physical activity level at baseline exceeds the mean physical activity level +2 SD of a cerebral palsy population, corresponding with 263 minutes of physical activity per day; or (4) severe cognitive disorder or insufficient comprehension of the Dutch language that would impede understanding of instructions for the intervention and assessments.

An informational letter and invitation to participate were sent to eligible persons. A reminder letter was sent four weeks later to non-responders. All
participants provided written informed consent. The medical ethics committee of the Erasmus Medical Center approved the study and local approval was granted by all participating centers.

**Randomization and intervention**

Following baseline measurement, participants were stratified according to GMFCS level to obtain an equal distribution of gross motor functioning between the experimental and control groups. Within each stratum and for each participating center, participants were randomly allocated (1:1) to these groups. Participants were assigned in chronological order of enrolment by using a series of numbers and each number had a randomly allocated group associated with it. As the patient was registered, he was allocated to the next number and then the group was revealed. The experimental group received the active lifestyle and sports participation intervention. The control group received no intervention to improve physical behavior and cardiopulmonary fitness, which is usual care in the Netherlands.

The active lifestyle and sports participation intervention, which was developed for adolescents and young adults with physical disabilities, lasted six months. This intervention aimed to permanently improve physical behavior and cardiopulmonary fitness, which is usual care in the Netherlands.

The active lifestyle and sports participation intervention, which was developed for adolescents and young adults with physical disabilities, lasted six months. This intervention aimed to permanently improve physical behavior and increase physical fitness. It consisted of: (1) counseling on daily physical activity and sedentary behavior, which was guided by a personal coach to discuss barriers and facilitators of physical behavior; (2) physical fitness training, which consisted of supervised center and home-based training and focused on increasing cardiopulmonary fitness and muscle strength; and (3) counseling on sports participation to find suitable, accessible and appropriate sports and sports facilities in the person’s day-to-day environment. This intervention has been described in detail elsewhere.15

**Measurements**

All measurements were performed thrice: (1) prior to starting the intervention; (2) immediately following the intervention, which was six months after the start of the intervention; and (3) a follow-up measurement six months after finishing the intervention.

**Fatigue**

Two widely used measures of fatigue were applied in this study: the Dutch version of the Fatigue Severity Scale (FSS)20 and the fatigue subscale (CIS-f) of the Checklist Individual Strength (CIS-20r).21 These two questionnaires likely measure different aspects of perceived fatigue. The FSS focuses on the impact of fatigue on specific types of functioning and the CIS-f measures the severity of perceived fatigue.22 The FSS is a 9-item, one-week recall, self-administered questionnaire with scores ranging from 1 to 7. The total score is the mean of nine items and ranges from 1 (no signs of fatigue) to 7 (most disabling fatigue). Internal consistency, reliability, validity and sensitivity of the FSS have been established.20 The CIS-f is a two-week recall questionnaire, assessing fatigue severity during the two weeks prior to assessment, with scores ranging from 1 to 7. The score of the CIS-f is the sum score of these eight questions and ranges from 8 to 56, with higher scores indicating more fatigue. Reliability, validity and sensitivity of the CIS have been established.23

**Social participation**

Social participation was assessed using the short version of the Life Habits Questionnaire (LIFE-H 3.0), which includes 69 life habits covering 12 categories (daily activities and social roles): nutrition, fitness, personal care, communication, housing, mobility, responsibilities, interpersonal relationships, community life, education, employment and recreation.24 Scoring is based on two specific aspects of participation: (1) the degree of difficulty in performing life habits (no difficulty, with difficulty, with substitution or not accomplished); and (2) the type of assistance required performing the habit (no help, technical assistance or adaptation, or human assistance). Both elements are combined in a scale ranging from 0 to 9, with 0 indicating total handicap and 9 indicating optimal activity or participation. The mean scores for the two subdomains (i.e. daily activities and social roles) were calculated. The LIFE-H has been shown to have moderate to good psychometric properties in adults with physical impairments.25
Health-related quality of life

The 36-item Short-Form health survey (SF-36) was used to measure health-related quality of life. The SF-36 is a validated, self-administered questionnaire used internationally to measure health status with respect to several domains: physical functioning, social functioning, role limitations due to physical problems, role limitations due to emotional problems, pain, mental health, vitality and general health perception. All raw scores were linearly converted to a 0 to 100 scale, with higher scores indicating higher levels of functioning or well-being. The Dutch language version of the SF-36 has shown good reliability and validity.

Gross motor functioning

Gross motor functioning was measured using the GMFM-66 item set. These item sets derive a GMFM-66 score by testing a subsample of GMFM-66 items and entering them into the Gross Motor Ability Estimator computer scoring program. The GMFM-66 item set has shown good reliability and validity.

Social Support for Exercise Behaviour

Social support is measured with the Social Support for Exercise Behaviour Scale. This scale consists of 18 items, with scores ranging from 1 to 5, covering three domains that address the social support of family and friends – Family support: participation and involvement; Family support: rewards and punishments; and Friends support: exercising together. Total score and domain scores were calculated by summing up the scores of the questions in the particular domains. The Dutch language version of the Social Support for Exercise Behaviour Scale has shown moderate reliability.

Self-efficacy and motivation

Self-efficacy is determined using the General Self-efficacy scale. This scale consists of 10 questions with scores ranging from 1 to 4. All responses are summed up to obtain the total score. Intrinsic motivation is measured with the Intrinsic Motivation Inventory. This inventory consists of 29 items on a 7-point scale. The response on all items were summed up to obtain the total score.

Potentially mediating variables

Four potentially mediating variables on intervention effects were specified: peak oxygen uptake, objectively measured physical activity level, objectively measured sedentary time, and self-reported physical activity level. Peak oxygen uptake was measured during progressive maximal exercise testing, and defined as the highest mean oxygen uptake during 30 seconds of exercise. An accelerometry-based ambulatory monitoring system (VitaMove) was used to objectively quantify physical behavior over a three-day period. From these measurements, the amounts of physical activity and sedentary time were determined. Self-reported physical activity level was measured using the Physical Activity Scale for Individuals with Physical Disabilities. More detail on how these potential mediating variables were measured is given elsewhere.

Statistical analyses

The current study is part of the Learn2Move study; therefore, the power analysis was performed on physical activity. A total of 50 participants were required to detect a clinically relevant difference of 30 minutes per day in physical activity between control and intervention groups, with a power of 0.8 and an alpha of 0.05.

Chi square tests and independent sample t-tests were applied to test for differences between groups at baseline. General Estimation Equation (GEE) analyses with exchangeable correlation structures were used to analyze the effect of the intervention. Group allocation, baseline values, measurement time and an interaction variable between group allocation and measurement time were added to the GEE model to compare the outcomes of the intervention group with the control group for specific time intervals. These time intervals were specified as the intervention period and the total period. The control group was specified as the reference group for all analyses. IBM SPSS Statistics version 20
(Chicago, USA) was used to perform statistical analyses.

In case of significant intervention effects between the control and intervention groups, additional analyses were performed to analyze possible mediating effects of peak oxygen uptake, objectively measured physical activity level, objectively measured sedentary time and self-reported physical activity level. Mediation was expressed as the percentage of change in the intervention effect(s) after adding the potential mediator to the GEE model.

**Results**

Between October 2009 and September 2011, we identified a target population of 456 adolescents and young adults with cerebral palsy in the registers of participating centers. Many eligible persons had not visited the rehabilitation center for many years. Therefore, the accuracy of their address information was uncertain. A total of 183 potential participants responded to our invitation, of whom 57 (31%) consented to participate, and 41 completed the study (Figure 1). Personal and clinical characteristics at baseline are presented in Table 1. No significant differences were found between the control and intervention group at baseline.

Participants who completed the intervention completed a mean of 89% of the supervised counselling and fitness training sessions.

**Intervention effects**

The observed data over time are presented in Table 2, and corresponding GEE analysis results are shown in Table 3. During the intervention period, fatigue severity, as measured by the CIS-f, decreased in the intervention group compared with the control group (difference = −6.72, \( p = 0.02 \)). An intervention effect was noted for bodily pain during the total period (difference = 15.14, \( p = 0.01 \)) in favor of the intervention group. Perceived mental health differed between groups during the intervention period (difference = 8.00, \( p = 0.03 \)) and total period (difference = 8.80, \( p = 0.03 \)). Furthermore, family support for exercise behavior (participation and involvement) increased during both the intervention (difference = 5.38, \( p = 0.04 \)) and total (difference = 4.52, \( p = 0.03 \)) period. No intervention effects were noted for fatigue, as measured by FSS, gross motor functioning, self-efficacy, motivation, limitation in participation in life areas or health-related quality of life domains other than mental health and bodily pain.

**Mediating effects**

Results of the additional analyses on mediating effects are shown in Table 4. Physical behavior explains intervention effects on fatigue severity (22.6%) (CIS-f), bodily pain (9.3%) and perceived mental health (28.1%), whereas physical fitness explains 16.0%, 9.7% and 22.6%, respectively, of intervention effects for those same outcome measures. Intervention effects on social support were mediated for only 2.6% by physical fitness, whereas no mediating effects of physical behavior was found.

**Discussion**

To our knowledge, this is the first longitudinal study to evaluate the effect of a lifestyle intervention on fatigue, social participation, gross motor functioning and health-related quality of life in adolescents and young adults with cerebral palsy, and study the mediating effects of physical behavior and fitness. The lifestyle intervention was effective in reducing fatigue severity and in increasing health-related quality of life with respect to bodily pain and mental health in the intervention group compared with the control group. In addition, the intervention increased the family support for the person’s exercise behavior, by participating and being involved in planning exercise activities. No intervention effects were noted for limitations in social participation, gross motor functioning or other health-related quality of life domains. Furthermore, motivation and generic self-efficacy were not altered by following the lifestyle intervention.

Additional analyses showed that the observed differences in family support for exercise between the intervention and control groups could be explained to a little extent by specific variables of physical fitness and physical behavior. On the other
Figure 1. Flow of participants through the study. T0, T6 and T12, respectively, represent the baseline, end of intervention and follow-up measurement.

Table 1. Baseline personal and clinical characteristics. P-values refer to differences between the control and intervention groups.

<table>
<thead>
<tr>
<th></th>
<th>Total (N = 57)</th>
<th>Control group (N = 29)</th>
<th>Intervention group (N = 28)</th>
<th>P-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Gender (M/F)</td>
<td>27/30</td>
<td>15/14</td>
<td>12/16</td>
<td>0.50</td>
</tr>
<tr>
<td>Age (years)</td>
<td>20 ±3</td>
<td>20 ±</td>
<td>20 ±3</td>
<td>0.64</td>
</tr>
<tr>
<td>Height (cm)</td>
<td>170 ±10</td>
<td>170 ±9</td>
<td>169 ±11</td>
<td>0.66</td>
</tr>
<tr>
<td>Body mass (kg)</td>
<td>67 ±18</td>
<td>65 ±18</td>
<td>70 ±18</td>
<td>0.24</td>
</tr>
<tr>
<td>CP type (unilateral/bilateral)*</td>
<td>29/27</td>
<td>15/14</td>
<td>14/13</td>
<td>0.79</td>
</tr>
<tr>
<td>GMFCS** level (I/II/III/IV)</td>
<td>33/18/5/1</td>
<td>16/9/3/1</td>
<td>17/9/2/0</td>
<td>0.75</td>
</tr>
</tbody>
</table>

*CP type was unknown for one person in the control group.
**Gross Motor Function Classification System.
### Table 2. Fatigue, participation and health-related quality of life outcome measures for intervention and control groups.

<table>
<thead>
<tr>
<th>Outcome measure</th>
<th>Intervention group</th>
<th>Control group</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>T0</td>
<td>T6</td>
</tr>
<tr>
<td>Fatigue (FSS)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>&lt;sup&gt;0.01&lt;/sup&gt;</td>
<td></td>
<td></td>
</tr>
<tr>
<td>T0: baseline measurement; T6: measurement directly after intervention completion (6 months after inclusion); T12: follow-up measurement (12 months after inclusion); FSS: Fatigue Severity Scale; CIS-f: fatigue subscale of the Checklist Individual Strength; HRQoL: health-related quality of life; SSEBS: Social Support for Exercise Behaviour scale; IMI: intrinsic motivation inventory; GSE: general self-efficacy scale; GMFM: gross motor function measure.</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
Table 3. GEE analysis results for T0–T6 differences and T0–T12 differences. All analyses were adjusted for baseline differences between groups on each variable. The control group is the reference group for all GEE analyses.

<table>
<thead>
<tr>
<th>Outcome measure</th>
<th>Difference T0–T6 (95% CI)</th>
<th>Difference T0–T12 (95% CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Fatigue (FSS)</td>
<td>−0.19 (−0.63, 0.25)</td>
<td>−0.53 (−1.08, 0.02)</td>
</tr>
<tr>
<td>Fatigue (CIS-f)</td>
<td>−6.72 (−12.44, −0.99)</td>
<td>−5.84 (−12.93, 1.26)</td>
</tr>
<tr>
<td>Participation, daily activities</td>
<td>−1.04 (−0.34, 0.13)</td>
<td>−0.22 (−0.50, 0.07)</td>
</tr>
<tr>
<td>Participation, social roles</td>
<td>−0.21 (−0.55, 0.12)</td>
<td>0.04 (−0.60, 0.53)</td>
</tr>
<tr>
<td>HRQoL, physical functioning</td>
<td>3.11 (−8.31, 14.53)</td>
<td>5.45 (−5.13, 16.04)</td>
</tr>
<tr>
<td>HRQoL, role physical</td>
<td>4.15 (−15.10, 23.40)</td>
<td>16.27 (−8.65, 41.20)</td>
</tr>
<tr>
<td>HRQoL, bodily pain</td>
<td>5.47 (−7.12, 18.06)</td>
<td>15.14 (3.44, 26.85)</td>
</tr>
<tr>
<td>HRQoL, general health</td>
<td>7.41 (−3.81, 18.62)</td>
<td>10.28 (−1.42, 21.98)</td>
</tr>
<tr>
<td>HRQoL, vitality</td>
<td>1.64 (−4.96, 8.23)</td>
<td>−0.40 (−6.92, 7.71)</td>
</tr>
<tr>
<td>HRQoL, social functioning</td>
<td>1.76 (−5.88, 9.41)</td>
<td>−3.08 (−12.64, 6.49)</td>
</tr>
<tr>
<td>HRQoL, role emotional</td>
<td>5.94 (−5.01, 16.90)</td>
<td>11.09 (−1.22, 23.39)</td>
</tr>
<tr>
<td>HRQoL, mental health</td>
<td><strong>8.00 (0.96, 15.05)</strong></td>
<td><strong>8.80 (0.99, 16.61)</strong></td>
</tr>
<tr>
<td>Gross motor functioning</td>
<td>−1.94 (−4.69, 0.82)</td>
<td>−0.08 (−1.99, 1.83)</td>
</tr>
<tr>
<td>Social support, total score</td>
<td>5.50 (−12.84, 1.83)</td>
<td>4.87 (−0.89, 10.62)</td>
</tr>
<tr>
<td>Family support: participation and involvement</td>
<td><strong>5.38 (0.03, 10.74)</strong></td>
<td><strong>4.52 (0.39, 8.65)</strong></td>
</tr>
<tr>
<td>Family support: rewards and punishments</td>
<td>−0.31 (−1.12, 0.49)</td>
<td>0.34 (−0.32, 1.00)</td>
</tr>
<tr>
<td>Friends support: exercising together</td>
<td>1.80 (−1.24, 4.83)</td>
<td>−0.61 (−3.71, 2.49)</td>
</tr>
<tr>
<td>Motivation</td>
<td>0.34 (−10.45, 11.13)</td>
<td>−0.20 (−9.24, 8.83)</td>
</tr>
<tr>
<td>Self-efficacy</td>
<td>0.91 (−2.11, 3.92)</td>
<td>0.96 (−1.86, 3.77)</td>
</tr>
</tbody>
</table>

T0: baseline measurement; T6: measurement directly after intervention completion (6 months after inclusion); T12: follow-up measurement (12 months after inclusion); FSS: Fatigue Severity Scale; CIS-f: fatigue subscale of the Checklist Individual Strength; HRQoL: health-related quality of life; GMFM: gross motor function measure; SSEBS: Social Support for Exercise Behaviour scale; IMI: intrinsic motivation inventory; GSE: general self-efficacy scale.

Note: Bold text indicates statistical significance.

Table 4. Mediating effects of physical behavior and fitness on fatigue, bodily pain and mental health.

<table>
<thead>
<tr>
<th>Mediating variables*</th>
<th>Outcome measures</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Fatigue (CIS-f)</td>
</tr>
<tr>
<td>Peak oxygen uptake</td>
<td><strong>16%</strong></td>
</tr>
<tr>
<td>Objective measured physical activity level</td>
<td><strong>6.2%</strong></td>
</tr>
<tr>
<td>Objective measured sedentary time</td>
<td><strong>5.9%</strong></td>
</tr>
<tr>
<td>Self-reported physical activity level</td>
<td><strong>22.6%</strong></td>
</tr>
</tbody>
</table>

*Expressed as a percentage of change in the intervention effect after adding the potential mediator to the GEE model.

CIS-f: fatigue subscale of the Checklist Individual Strength; SF-36: 36-item Short-Form health survey; SSEBS: Social Support for Exercise Behaviour scale.

hand, the observed differences between the inter-
vention and control groups on the remaining param-
eters could be explained to a considerable extent by single variables of physical fitness and physical behavior, specifically by self-reported physical activity and physical fitness. Apparently,
self-reported physical activity and physical fitness levels are substantial mediators of the effect of the active lifestyle and sports participation intervention on fatigue, bodily pain and mental health. Therefore, apart from their direct health benefits, these results stress the importance of favorable physical behavior and sufficient physical fitness for adolescents and young adults with cerebral palsy.

At baseline, participants were more fatigued compared with the general population, as measured by the FSS (3.6 vs. 3.0)\textsuperscript{20} and CIS-f (26.0 vs. 24.4).\textsuperscript{32} Thus, fatigue in persons with cerebral palsy may be a problem even at a young age. The active lifestyle and sports participation intervention was effective in decreasing fatigue severity during the intervention period. This finding is consistent with results from Vogtle et al., who found that fatigue levels in ambulatory adults with cerebral palsy decreased following an exercise training program.\textsuperscript{6} However, Vogtle et al. found these effects during both intervention and follow-up periods, whereas observed effects in the present study did not persist into the follow-up period of our current study. To facilitate long-term effectiveness, booster strategies, such as phone, mail or internet support, could augment the active lifestyle and sports participation intervention, as these strategies seem effective for maintaining lifestyle intervention effects over the long term.\textsuperscript{33}

In contrast to CIS-f scores, FSS scores did not change following the active lifestyle and sports participation intervention. This discrepancy may reflect different constructs of fatigue assessed by these questionnaires. The FSS measures the impact of fatigue on specific types of functioning, whereas the CIS-f more specifically measures the severity of the perceived fatigue.\textsuperscript{22} In adults with cerebral palsy, fatigue may not affect functioning comparably with able-bodied persons because their functioning could have been adapted or limited from an early age.

At baseline, mean scores for difficulty in daily activities and social roles on the LIFE-H 3.0 were 7.9 and 7.8, respectively. These scores are somewhat higher than participation levels among adults with bilateral spastic cerebral palsy, who have mean scores of 7.5 and 7.7, respectively, on these domains.\textsuperscript{34} These higher social participation levels could be explained by the large percentage of persons with unilateral cerebral palsy in the current study, as well as their younger age. The active lifestyle and sports participation intervention was not effective in decreasing restrictions in daily activities or social roles. This finding partially contrasts with those of van Wely et al. among school-age children with cerebral palsy, who found positive long-term effects on activities in and around the house (domestic life) following a lifestyle intervention.\textsuperscript{8} Similar to our study, these children did not experience improvements in performance of mobility and leisure activities related to moving outside the house, in the local community, in the wider environment or in sports.

Baseline scores for health-related quality of life domains (except bodily pain) among our adolescent and young adult participants with cerebral palsy were lower compared with Dutch reference values.\textsuperscript{26} This finding is consistent with studies of adolescents\textsuperscript{35} and adults with cerebral palsy,\textsuperscript{10,36,37} which also show subnormal levels of perceived health-related quality of life, specifically in physical functioning. Exercise has been shown to reduce pain in adults with cerebral palsy,\textsuperscript{38} and to increase cerebral palsy-specific health-related quality of life over time in children.\textsuperscript{8} Cerebral palsy-specific health-related quality of life evaluates self-perceived pain and fatigue, in addition to functional levels in movement, balance, upper-limb activities, speech and communication. Positive effects on pain and fatigue were confirmed by results of the present study, as fatigue severity and perceived bodily pain decreased following the active lifestyle and sports participation intervention. In addition, the present lifestyle intervention was effective in improving mental health. Despite low baseline health-related quality of life, other health-related quality of life domains did not change following the active lifestyle and sports participation intervention.

The gross motor functioning score, as assessed with the Gross Motor Function Measure-66 was around 83 at baseline. This is higher compared with another study on adolescents with cerebral palsy.\textsuperscript{39} The relatively high score on gross motor functioning is possibly explained by the relative lack of severity of the included sample of the present study. Bartlett et al. found that pain and gross
motor functioning are related in adolescents with cerebral palsy. In the present study, the observed intervention effect on pain severity did not lead to significant changes in gross motor functioning.

Loss to follow-up was 39% in the intervention group and 34% in the control group (Figure 1). A type II error may have occurred in the longitudinal analysis owing to a higher drop-out rate than expected. We were not able to perform the remaining measurements on the persons that dropped out of the study. Owing to multiple testing in the current study, one should be careful to draw strong conclusions from single significant findings.

Although a sample size of 57 is relatively large considering the prevalence of spastic cerebral palsy, the absolute number is quite small. However, we do not expect that the results would have differed with fewer participant drop-outs, considering that the clinically relevant differences on non-significant outcome measures are not included in the 95% CI of the analyses.

Our study participants had relatively high gross motor functioning (89% had GMFCS Level I or II) and intellectual functioning (IQ > 70). It can be hypothesized that lifestyle intervention effects can be affected by levels of gross motor and intellectual functioning. Future research is required to clarify the effectiveness of lifestyle interventions in study samples with lower levels of gross motor functioning and intellectual functioning.

**Clinical messages**

- The active lifestyle and sports participation intervention was effective in decreasing fatigue and in increasing social support and health-related quality of life regarding bodily pain and self-perceived mental health.
- The observed effects of the active lifestyle and sports participation intervention were to a considerable extent mediated by physical behavior and fitness.

**Conflict of interest**

The authors declare that there is no conflict of interest.

**Funding**

This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

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