Fatigue in children and young adults with physical disabilities: relation with energy demands of walking and physical fitness

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Submitted
**ABSTRACT**

**AIM:** To examine whether general fatigue, and fatigue during or after walking (i.e. walking-induced fatigue), are related to energy demands in walking and physical fitness in children and young adults with physical disabilities who experience problems with walking, and whether these relations differ between younger and older participants, and/or between boys and girls.

**METHODS:** Included were 68 individuals with physical disabilities (mean age 13y1mo; range 7y5mo – 22y7mo). General fatigue was measured with the Checklist Individual Strength (CIS8R) questionnaire. Walking-induced fatigue was measured with the OMNI (OMNIwalk) scale after walking for 6 min at a self-selected speed. Gross and net energy costs of walking, physical strain of walking, and aerobic and anaerobic fitness were measured.

**RESULTS:** Regression analyses showed no relations with the CIS8R. For all participants, a higher net energy cost ($R^2 = 0.134$) was weakly related to an increased OMNIwalk. For older participants only, low anaerobic fitness ($R^2 = 0.228$) and high physical strain values ($R^2 = 0.318$) were moderately related to high OMNIwalk scores.

**INTERPRETATION:** Whereas the energy demands of walking and physical fitness were not related to general fatigue, low anaerobic fitness and high physical strain values were related to more walking-induced fatigue in individuals aged 12 – 22 years. Therefore, interventions aimed at increasing anaerobic fitness and reducing physical strain may reduce walking-induced fatigue in these individuals.
INTRODUCTION

Persons with a physical disability, such as cerebral palsy (CP) and spina bifida (SB), experience many health-related problems, including functional deterioration, pain and fatigue. Fatigue is one of the most common issues experienced by young adults with CP and is more often reported by adolescents and (young) adults with CP as compared with their typically developing peers. Moreover, a study among children with physical disabilities showed that fatigue is a serious issue, especially for girls. Similarly, in clinical practice, many children and young adults with a physical disability visit a rehabilitation physician with complaints about fatigue during walking or other daytime activities.

Fatigue is described as a complex subjective and multidimensional phenomenon, with both mental and physical causes. Although measurement of fatigue is not easy, questionnaires, such as the Checklist Individual Strength (CIS), are used to measure the different dimensions of fatigue. A subscale of the CIS is ‘subjective fatigue’ (CIS8R), which measures the severity of general fatigue over a period of two weeks (where general fatigue is described as the experience of feeling tired, weak or lacking energy). A frequently reported complaint by individuals with physical disabilities is fatigue during or after walking, here referred to as walking-induced fatigue. Since children may have difficulty in recalling their perceptions related to an earlier period of time, the Children’s OMNI Scale of Perceived Exertion (OMNI) can be used (instead of a questionnaire) to measure the rate of perceived exertion real-time for a specific task. Therefore, use of the OMNI after walking (i.e. the OMNIwalk) can be considered a task-specific way to measure walking-induced fatigue.

A qualitative review suggested that fatigue affects the ability of children and young adults with CP to participate in school, leisure and recreation activities. The author of that study concluded that clinicians should help individuals with CP to manage fatigue to prevent loss of ambulation; to this end, it is important to understand which factors are related to fatigue. It is also reported that children and young adults with CP and SB have higher energy demands of walking, and that children and young adults with physical disabilities have a decreased level of physical fitness (i.e. aerobic and anaerobic fitness). Although it seems intuitive that higher energy demands of walking and lower physical fitness are physical causes of fatigue, to the best of our knowledge the relation between fatigue and energy demands of walking has not yet been studied, and inconsistent results are reported for the relation between general fatigue and physical fitness. It is suggested that higher levels of physical strain of walking (defined as the relative oxygen uptake of walking) might cause the onset of early fatigue. The physical
strain combines the oxygen uptake of walking and the peak oxygen uptake (VO\textsubscript{peak}), thereby reflecting the relative intensity of walking.\textsuperscript{18} However, no study has investigated the relation between the physical strain of walking and fatigue in children or young adults with CP.\textsuperscript{18}

Therefore, the present study examines whether general fatigue and walking-induced fatigue are related to the energy demands of walking and physical fitness in children and young adults with physical disabilities who experience problems with walking. Since measuring fatigue immediately after walking is a more direct measure of fatigue compared to general fatigue measured with a questionnaire, we hypothesized that the relation with the energy demands of walking and physical fitness will be stronger for walking-induced fatigue compared to general fatigue. Finally, because differences are reported in the prevalence of fatigue between children and young adults,\textsuperscript{5,19} and between boys and girls,\textsuperscript{6,20} we also investigated whether the relations differ for age and/or gender.

**METHODS**

**Participants**

This study used data from the exercise laboratory of the outpatient clinic of the Department of Rehabilitation Medicine at the VU University Medical Centre (VUmc, Amsterdam), a tertiary referral center. Patients with walking problems (e.g. reduced walking distance and fatigue during/after walking or other daily activities) were referred to the clinical exercise laboratory in the context of their regular health care by their rehabilitation physician. Clinical exercise test results were used to set an appropriate treatment plan to reduce their walking problems. Data were collected between January 2014 and November 2016. Participants with walking problems due to a non-progressive physical disability (e.g. CP, and other diagnoses such as SB or Kabuki syndrome) were included when they were able to walk at least 5 min with or without walking aids, and when they were able to follow simple instructions. Exclusion criteria were: 1) orthopedic or neurosurgical treatment in the past 6 months, or botulinum toxin treatment in the past 3 months, and 2) cardiac abnormalities, mitochondrial abnormalities, unstable epilepsy, or other contraindications for a maximal exercise test. Participants were divided into four different diagnostic groups: gross motor function classification system (GMFCS) levels I, II, III/IV, and other diagnoses. The Medical Ethical committee of the VUmc waived the necessity of official approval, since the procedures were part of a standard clinical procedure.
Procedure and equipment

The energy demands of walking were measured using a resting test and a walking test, and physical fitness was measured using a maximal aerobic and anaerobic fitness test. The participants were given specific instructions not to eat or drink (except for water) 1.5 h prior to the measurements. First, height (cm) and weight (kg) were measured with a wall-fixed measure in standing position (DGI 250D, KERN DE version 3.3 10/2004) combined with an electronic scale (Kern & Sohn GmbH, Balingen-Frommern, Germany). During the walking and maximal aerobic fitness tests, pulmonary gas exchange was measured using a portable gas analysis system (Metamax 3B Cortex Biophysik, Leipzig, Germany). Oxygen uptake (VO₂, ml·kg⁻¹·min⁻¹) and carbon dioxide production (VCO₂, ml·kg⁻¹·min⁻¹) values were measured breath by breath. The respiratory exchange ratio was calculated as VCO₂ divided by VO₂. The maximal aerobic and anaerobic fitness tests were performed on a cycle ergometer (Corival V2, Lode B.V., Groningen, the Netherlands).

Fatigue

General fatigue was measured with the ‘subjective fatigue’ subscale (CIS8R) of the CIS questionnaire. This subscale assesses the severity of general fatigue in the last two weeks by means of 8 items (e.g. ‘I feel tired’, ‘I feel fit’ and ‘I feel rested’). The participants indicated to what extent the particular statement applies to them on a 7-point Likert scale, with higher scores (range 8 – 56) indicating a higher degree of general fatigue. The CIS was originally developed to measure fatigue in an adult population. However, in Dutch studies among healthy children aged 12 – 19 years, and among adults with physical disabilities, the CIS has proven reliable to measure fatigue. Furthermore, the CIS and CIS8R have been used to evaluate fatigue for adolescents with CP, as well as adolescents with hereditary motor and sensory neuropathy. One of these latter studies also showed that, after fitness training for adolescents with CP, the CIS8R was sensitive to change.

In the present study, walking-induced fatigue was measured with the OMNIwalk as the rate of perceived exertion immediately after participants had walked for 6 min at a self-selected speed. The OMNI scale provides a subjective method for systematically reporting perceived effort on a 0 – 10 point scale, whereby 0 indicates ‘not tired at all’ and 10 indicates ‘very, very tired’. The OMNI has been validated for typically developing children aged 8 – 18 years. Moreover, in ambulatory children with CP aged 6 – 18 years, the OMNI was shown to be a clinically feasible and valid instrument.
Energy demands of walking and physical fitness

Walking test
The energy demands of walking were measured during walking at a self-selected speed for 6 min. The test started with a resting period during which participants watched a movie for 5 min in a sitting position. Then, participants walked on an indoor oval track (40 m). VO$_2$ and VCO$_2$ were measured and used to calculate the gross and net energy consumption (J·kg$^{-1}$·min$^{-1}$). Participants were excluded when their walking speed was ≤ 24 m·min$^{-1}$, as these participants appeared not to be functional walkers in daily life. The net energy consumption was calculated as: gross energy consumption minus resting energy consumption.

To calculate the gross and net energy cost, the gross and net energy consumption were divided by walking speed and expressed in J·kg$^{-1}$·m$^{-1}$. Average energy cost values were calculated over 2 min when a steady state has been reached (excluding the first 3 min). Using this protocol, energy costs can be reliably determined in children with CP.$^{26}$

Aerobic fitness test
Aerobic fitness was measured with a maximal exercise test. This test started with a 2 – 4 min warming-up period, followed by a 3 – 5 min submaximal exercise phase. After a 1-min rest, the maximal phase started with 1-min incremental exercise bouts until exhaustion.$^{15}$ The test outcomes were included in the analyses if at least two of the following three criteria for achieving maximal exercise were met: 1) heart rate ≥ 180 b·min$^{-1}$, 2) respiratory exchange ratio ≥ 1.00, and 3) observed signs of exhaustion (e.g. sweating, out of breath, and fatigued) were present. Both the peak oxygen uptake (VO$_{\text{peak}}$, expressed in ml·kg$^{-1}$·min$^{-1}$) and peak aerobic power output (PO$_{\text{peak}}$, expressed in W·kg$^{-1}$) were used as outcomes of the maximal aerobic fitness test. The VO$_{\text{peak}}$ was defined as the highest oxygen uptake over 30 s and the PO$_{\text{peak}}$ as the highest power output maintained for a minimum of 30 s. The test-retest reliability of this protocol was shown to be excellent for children with CP.$^{27}$

Anaerobic fitness test
A 20-s Wingate sprint test was conducted to estimate anaerobic fitness. For familiarization and to determine the optimal breaking torque, three warm-up sprints of 5 s each were performed at different torques, determined by the severity of the motor disorder and the body height of the participant.$^{15}$ The breaking torque for the 20-s Wingate sprint test was the torque at which the participant ended a 5-s sprint test at a cycle frequency of 90 – 100 rpm.$^{15}$ After the three warm-up sprints, participants performed a 20-s full-out
sprint against a constant workload. The mean anaerobic power over 20 s \( \text{P20}_{\text{mean}} \) was calculated as an estimate for the sprint capacity (expressed in \( \text{W} \cdot \text{kg}^{-1} \)). The test-retest reliability of this outcome has been shown to be excellent for children with CP.\(^{28}\)

**Physical strain**
The physical strain was calculated as the oxygen uptake of walking expressed as a percentage of \( \text{VO}_{\text{peak}} \).\(^{18}\)

**Statistical analysis**
Descriptive statistics were applied to describe subject characteristics, general fatigue, walking-induced fatigue, energy demands of walking and physical fitness values for the different diagnostic groups. Distribution of the data was checked using inspection of mean values, standard deviations (SD), and visual inspection of the histograms and normal Q-Q plots. Since the data were merely normally distributed, parametric statistical tests were applied.

Univariate and multiple linear regression analyses were used to evaluate whether the CIS8R and OMNIwalk (dependent variables) were related to the energy demands of walking and physical fitness. The independent variables were gross energy cost, net energy cost, \( \text{VO}_{\text{peak}} \), \( \text{PO}_{\text{peak}} \), anaerobic fitness, and physical strain. To test whether these relations were different for boys and girls, interaction terms were included in the models. Because it was assumed that age could also be an effect modifier, the participants were dichotomised in two age groups: a younger pre-pubertal age group (girls < 11 years\(^{29}\) and boys < 12 years\(^{30}\)) and an older age group. Stratified analyses were presented when the interaction terms were significant. The overall significance level was set at \( p \)-value < 0.05.

**RESULTS**
The study included 68 children and young adults aged 7 – 22 years (mean age 13 years 1 month; SD 3 years 7 months); characteristics of these participants are shown in Table 4.1.

Of the 68 participants, two did not score the OMNIwalk because they were not referred for the walking test. Also, although being able to follow simple instructions was an inclusion criterium, 10 participants were unable to score the CIS8R due to low cognitive functioning; these latter participants were excluded from the analysis. Finally, 66 participants scored the OMNIwalk and 58 scored the CIS8R (Figure 4.1).
Table 4.1  Characteristics of the study participants (n = 68)

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Mean (Range; SD)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age [y mo]</td>
<td>13y1mo (7y5mo to 22y7mo; 3y7mo)</td>
</tr>
<tr>
<td>Gender</td>
<td></td>
</tr>
<tr>
<td>Boy</td>
<td>41 (60%)</td>
</tr>
<tr>
<td>Girl</td>
<td>27 (40%)</td>
</tr>
<tr>
<td>Height [cm]</td>
<td>153.1 (116 – 187; 171)</td>
</tr>
<tr>
<td>Weight [kg]</td>
<td>46.9 (18.8 – 69.0; 13.9)</td>
</tr>
<tr>
<td>Body mass index</td>
<td>19.6 (13.7 – 26.9; 3.3)</td>
</tr>
<tr>
<td>Diagnosis</td>
<td></td>
</tr>
<tr>
<td>CP GMFCS level I</td>
<td>13 (19%)</td>
</tr>
<tr>
<td>CP GMFCS level II</td>
<td>34 (50%)</td>
</tr>
<tr>
<td>CP GMFCS level III/IV</td>
<td>11 (16%)</td>
</tr>
<tr>
<td>Other diagnoses*</td>
<td>10 (15%)</td>
</tr>
</tbody>
</table>

* Other diagnoses were: fatigue without any specific known cause (n = 1); psychomotor retardation (n = 1); Kabuki syndrome (n = 1); Alport syndrome (n = 1); craniostenosis (n = 1); progressive cerebellar atrophy (n = 1); novo mutation CACNA1A (n = 1); cerebellar ataxia ECI (n = 1); spina bifida (n = 2).

GMFCS, Gross Motor Function Classification System.

Mean values of general fatigue, walking-induced fatigue, energy demands of walking and physical fitness are presented in Table 4.2. In participants with CP, both general fatigue and walking-induced fatigue increased with larger motor involvement (i.e. higher GMFCS levels).

Table 4.3 presents results of the linear regression analyses for general fatigue (CIS8R) and walking-induced fatigue (OMNIwalk). The CIS8R was not related to gross or net energy cost, or physical strain. There were trends for a relation between the CIS8R and the VO\textsubscript{peak} (p: 0.055; R\textsuperscript{2}: 0.083), PO\textsubscript{peak} (p: 0.060; R\textsuperscript{2}: 0.082) and anaerobic fitness (p: 0.095; R\textsuperscript{2}: 0.076). There were no interaction effects for age and gender in the relation with the CIS8R.

The OMNIwalk was significantly related to gross energy cost (R\textsuperscript{2}: 0.093), net energy cost (R\textsuperscript{2}: 0.134), VO\textsubscript{peak} (R\textsuperscript{2}: 0.087), physical strain (R\textsuperscript{2}: 0.213) and anaerobic fitness (R\textsuperscript{2}: 0.113). The relations show higher OMNIwalk values for children with high gross energy cost, high net energy cost and high physical strain values, and with low VO\textsubscript{peak} and low anaerobic fitness values. There was a trend for a negative relation between the OMNIwalk and PO\textsubscript{peak}.
Figure 4.1  Recruitment of the study sample and reasons for missing data.
CIS8R, Checklist Individual Strength, subscale Subjective fatigue; OMNIwalk, Children’s OMNI Scale of Perceived Exertion.
Table 4.2  Mean (SD) values of the fatigue scores, and the energy demands during walking and physical fitness of the participants (n = 68)

<table>
<thead>
<tr>
<th></th>
<th>GMFCS level I (n = 13)</th>
<th>GMFCS level II (n = 34)</th>
<th>GMFCS level III/IV (n = 11)</th>
<th>Other diagnosis (n=10)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Fatigue</strong></td>
<td></td>
<td></td>
<td></td>
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</tr>
<tr>
<td>CIS8R</td>
<td>24.8 (9.6) [n = 12]</td>
<td>28.7 (8.6) [n = 28]</td>
<td>34.3 (10.5) [n = 9]</td>
<td>27.8 (9.7) [n = 9]</td>
</tr>
<tr>
<td>OMNIwalk</td>
<td>3.9 (2.0) [n = 13]</td>
<td>5.1 (2.3) [n = 34]</td>
<td>5.7 (3.1) [n = 10]</td>
<td>3.4 (2.1) [n = 9]</td>
</tr>
<tr>
<td><strong>Energy demands during walking</strong></td>
<td></td>
<td></td>
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</tr>
<tr>
<td>Gross EC</td>
<td>5.2 (0.9) [n = 13]</td>
<td>71 (1.6) [n = 33]</td>
<td>9.6 (1.5) [n = 7]</td>
<td>5.5 (1.9) [n = 10]</td>
</tr>
<tr>
<td>Net EC</td>
<td>3.5 (0.5) [n = 13]</td>
<td>5.3 (1.5) [n = 33]</td>
<td>70 (1.3) [n = 7]</td>
<td>4.0 (1.3) [n = 10]</td>
</tr>
<tr>
<td><strong>Aerobic fitness</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>VO_{peak}</td>
<td>40.0 (9.9) [n = 12]</td>
<td>38.1 (7.5) [n = 22]</td>
<td>30.3 (7.0) [n = 6]</td>
<td>32.7 (9.1) [n = 10]</td>
</tr>
<tr>
<td>PO_{peak}</td>
<td>2.5 (0.7) [n = 12]</td>
<td>1.9 (0.6) [n = 22]</td>
<td>1.3 (0.4) [n = 6]</td>
<td>2.1 (0.8) [n = 10]</td>
</tr>
<tr>
<td><strong>Physical strain during walking</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
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<tr>
<td>Physical strain</td>
<td>48.9 (13.3) [n = 12]</td>
<td>58.4 (13.0) [n = 21]</td>
<td>78.0 (18.5) [n = 6]</td>
<td>57.3 (19.9) [n = 10]</td>
</tr>
<tr>
<td><strong>Anaerobic fitness</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>P2O_{mean}</td>
<td>6.0 (1.7) [n = 12]</td>
<td>4.0 (1.2) [n = 19]</td>
<td>2.5 (0.5) [n = 3]</td>
<td>4.8 (2.4) [n = 6]</td>
</tr>
</tbody>
</table>

GMFCS, gross motor function classification system; CIS8R, Checklist Individual Strength, subscale subjective fatigue; OMNIwalk, Children’s OMNI Scale of Perceived Exertion; EC, energy cost; VO_{peak}, peak oxygen uptake; PO_{peak}, peak aerobic power output; P2O_{mean}, mean anaerobic power.
### Table 4.3: Results of regression analysis for the relation between fatigue and energy demands during walking and physical fitness

<table>
<thead>
<tr>
<th>Independent variables</th>
<th>( \beta ) (95% CI)</th>
<th>( p )</th>
<th>( R^2 )</th>
</tr>
</thead>
<tbody>
<tr>
<td>( \beta ) OMNIwalk</td>
<td>0.034 (0.000 / 0.068)</td>
<td>0.0003</td>
<td>0.003</td>
</tr>
<tr>
<td>Gross ( EC ) [J·kg(^{-1})·m(^{-1})]</td>
<td>0.202 (1.081 / 1.485)</td>
<td>0.753</td>
<td>0.002</td>
</tr>
<tr>
<td>Net ( EC ) [J·kg(^{-1})·m(^{-1})]</td>
<td>0.251 (1.271 / 1.774)</td>
<td>0.742</td>
<td>0.002</td>
</tr>
<tr>
<td>VO(_{2})peak [ml·kg(^{-1})·min(^{-1})]</td>
<td>-0.278 (-0.562 / -0.006)</td>
<td>0.691</td>
<td>0.004</td>
</tr>
<tr>
<td>PO(_{20})mean [W·kg(^{-1})]</td>
<td>-1.333 (-2.907 / 0.242)</td>
<td>0.095</td>
<td>0.076</td>
</tr>
</tbody>
</table>

**Results**

- **Independent variables** with a significant relation with fatigue (\( p < 0.05 \)).
- **Italic** variables that differ between the younger and older age group.
- \( \beta \), regression coefficient; CI, confidence interval; EC, energy cost; VO\(_{2}\)peak, peak oxygen uptake; PO\(_{20}\)mean, mean anaerobic power.
Figure 4.2a  Relation between OMNIwalk and physical strain for younger (grey line = $\beta$: 0.005; $p$: 0.877; $R^2$: 0.002) and older participants (black line = $\beta$: 0.089; $p$: 0.023; $R^2$: 0.350).
Grey dots, younger participants; Black dots, older participants (girls aged over 11 years and boys aged over 12 years).

Figure 4.2b  Relation between OMNIwalk and anaerobic fitness for younger (grey line = $\beta$: 0.165; $p$: 0.619; $R^2$: 0.027) and older participants (black line = $\beta$: -0.854; $p$: 0.038; $R^2$: 0.264).
Grey dots, younger participants; Black dots, older participants (girls aged over 11 years and boys aged over 12 years).
The linear regression analyses showed significant interactions of age with anaerobic fitness and physical strain, indicating that these relations are different for the two age groups (Figure 4.2). For the older age group, high rates of OMNIwalk were related to low anaerobic fitness values ($\beta$: -0.854 (95% CI: -1.580 / -0.011); $p$: 0.038; $R^2$: 0.264) and high physical strain values ($\beta$: 0.089 (95% CI: 0.013 / 0.164); $p$: 0.023; $R^2$: 0.350), whereas these relations were not present in the younger age group. There were no interaction effects for gender.

**DISCUSSION**

This study examined whether general fatigue and walking-induced fatigue are related to the energy demands of walking and physical fitness in children and young adults with physical disabilities who experience problems with walking. For general fatigue, measured with the CIS8R questionnaire, no relations were found. However, walking-induced fatigue, measured immediately after walking with the OMNI, was weakly related to high values for gross and net energy cost, and to low VO$_{2peak}$ values. Furthermore, for girls aged ≥ 11 years and boys aged ≥ 12 years, walking-induced fatigue was related to low anaerobic fitness and high physical strain values. The observed relations were similar for boys and girls.

These results indicate that general fatigue, measured with the CIS8R questionnaire, was not related to the energy demands of walking and physical fitness. There were, however, trends for a relation with the VO$_{2peak}$, PO$_{peak}$ and anaerobic fitness. However, since the explained variance ($R^2$) of these relations was weak, these parameters do not (or hardly) contribute to general fatigue. An explanation for the lack of a relation might be that recalling perceived fatigue is difficult, especially for children. Although factors possibly contributing to the *experience of fatigue* among individuals with CP include higher BMI, more pain or being physically inactive, controversy still exists as to which factors contribute to *general fatigue*. Two studies which measured fatigue with a questionnaire support our conclusion regarding the lack of a relationship between fatigue and physical fitness. Thus, there is evidence that deviated energy demands of walking and physical fitness values do not cause general fatigue. This suggests that rehabilitation physicians may need to focus on other causes when children/young adults with physical disabilities complain of general fatigue. Also, since a good understanding of general fatigue is required before effective interventions can be developed, future research should investigate which mental and/or physical factors cause general fatigue.
In contrast to general fatigue, walking-induced fatigue showed a significant relation with gross energy cost, net energy cost and VO\textsubscript{2peak} and, for the older participants, with physical strain and anaerobic fitness. We assume that this discrepancy reflects the different factors that cause general fatigue and walking-induced fatigue because, whereas general fatigue seems to be influenced by both mental and physical causes, walking-induced fatigue seems to be mainly influenced by physical causes. While there is no general consensus on the definition of fatigue, we suggest that general fatigue and walking-induced fatigue are different problems that should be clearly distinguished and measured separately in clinical practice.

The present study revealed clear differences between the younger and older participants. While the relations between walking-induced fatigue and the gross energy cost, net energy cost and VO\textsubscript{2peak} were weak for both younger and older participants, combining both the energy cost and VO\textsubscript{2peak} with the physical strain\textsuperscript{18} showed a moderate relation for older participants only. Furthermore, for the older participants, a moderate relation was found between walking-induced fatigue and anaerobic fitness. An explanation for the differences between the two age groups might be that, particularly younger children, tend to underestimate fatigue; for example, children with CP reported less fatigue than their parents via proxy reports.\textsuperscript{19} In the present study, younger participants indeed showed slightly lower scores for the OMNIwalk (4.2; SD 2.1; range 0 – 7) compared to older participants (5.0; SD 2.6; range 0 – 10). Alternatively, walking-induced fatigue might be less severe in younger children, or other factors may cause walking-induced fatigue in these younger individuals.

Among older participants, improving anaerobic fitness and physical strain may lower walking-induced fatigue. It is not surprising that walking-induced fatigue is related to physical strain because walking at intensities ≥ 60% of the VO\textsubscript{2peak} is fatiguing and our participants (with GMFCS levels II, III/IV and other diagnoses) indeed walked at intensities ≥ 60%. However, in order to improve physical strain, it is important to know whether the increased physical strain is caused by increased energy demands of walking, or by decreased VO\textsubscript{2peak} levels, as different treatments will be required to improve these outcomes.\textsuperscript{34} Thus, orthotics and orthopedic surgery have the potential to reduce energy cost in ambulatory children with CP,\textsuperscript{35,36} while fitness training has the potential to improve the VO\textsubscript{2peak} in children and young adults with physical disabilities.\textsuperscript{37,38} Fitness training might also improve the anaerobic fitness.\textsuperscript{39} However, randomized controlled trials are required to reveal whether improving the physical strain and anaerobic fitness leads to clinically important improvements in walking-induced fatigue.
Limitations

The present study has some limitations that need to be addressed. First, since all participants reported walking problems, the results of this study cannot be generalized to individuals without walking problems.

There were missing values for both the energy demands of walking and for physical fitness. Although some participants were unable to perform a walking test, or an aerobic or anaerobic fitness test, most missing data on the energy demands of walking and physical fitness were because participants were specifically referred for one of the tests (and not the other) as they had already undergone too many tests in the context of their regular health care. These missing values may have caused a type II error.

The psychometric properties of the CIS have been evaluated for typically developing and physically disabled individuals aged ≥ 12 years.20,21,22,24 However, another questionnaire (with items similar to those in the CIS8R) designed to measure fatigue, i.e. the Pediatric Quality of Life Inventory (PedsQL), has also shown good psychometric properties for children with CP aged 5 – 18 years. Furthermore, younger participants were assisted by a parent or caregiver (if required) and, when the researcher doubted the ability of the participant to score the CIS8R (both younger and older participants), these outcomes were excluded from the analyses (n = 10). Therefore, in the present study, it was assumed that the younger children were also able to adequately fill in the CIS8R.

In the present study, due to the small sample size, we did not investigate whether the observed relations between walking-induced fatigue and the energy demands of walking and physical fitness differ between the diagnostic subgroups. Additional studies with a larger number of participants are required to further examine these differences.

CONCLUSION

The results of this study show that general fatigue is not related to the energy demands of walking and physical fitness in children and young adults with physical disabilities who experience problems with walking. However, high rates of walking-induced fatigue relate to low anaerobic fitness and high physical strain values for girls aged ≥ 11 years and for boys aged ≥ 12 years. These results suggest that interventions aimed at improving anaerobic fitness and reducing physical strain might reduce walking-induced fatigue and therefore warrant further investigation.
ACKNOWLEDGMENTS

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Fatigue in children with physical disabilities