

11-7-2018

Fatigue in adults with cerebral palsy: A three-year follow-up study.

Irene L B Oude Lansink

Patrick G McPhee

Laura Kristine Brunton

Jan Willem Gorter

Follow this and additional works at: <https://ir.lib.uwo.ca/ptpub>

 Part of the [Physical Therapy Commons](#)

Citation of this paper:

Lansink, I. O. L., McPhee, P. G., Brunton, L. K., & Gorter, J. W. (2018). Fatigue in adults with cerebral palsy: A three-year follow-up study. *Journal of Rehabilitation Medicine*, 50(10), 886-891.

1 **Fatigue in adults with cerebral palsy: a 3-year follow-up study**

2 Irene L.B. Oude Lansink¹, Patrick G. McPhee², Laura K. Brunton³, Jan Willem Gorter^{2,4,*}

3

4

5 **Affiliations**

6 1. Wilhelmina Children's Hospital, University Medical Center Utrecht, Department of
7 Rehabilitation Medicine, Utrecht, the Netherlands;

8 2. School of Rehabilitation Science, McMaster University, Hamilton, ON, Canada;

9 3. School of Physical Therapy, Western University, London, ON, Canada;

10 4. Department of Pediatrics, CanChild Centre for Childhood Disability Research, McMaster
11 University, Hamilton, ON, Canada.

12

13 **Short title:** Course of fatigue in Cerebral Palsy

14

15 **AIM(S)** (1) to describe the course of fatigue over a 3-year follow-up period in adults with
16 cerebral palsy (CP); and (2) to investigate the association of known determinants of fatigue
17 (i.e. demographic characteristics and/or body composition) with change in fatigue.

18 **METHOD** Forty-one adults with CP from a previous study examining fatigue were invited to
19 participate in a follow-up study. Twenty-three adults with CP (GMFCS levels I-V; mean age
20 38y 2m, standard deviation [SD 14y 1m]) agreed to participate (convenience sample).
21 Fatigue was measured with the Fatigue Impact and Severity Self-Assessment (FISSA, range
22 31 to 157) questionnaire. The course of fatigue is described at group, subgroup (GMFCS)
23 and individual levels.

24 **RESULTS** The mean (SD) FISSA score for all participants was 84.0 (27.7) at baseline and
25 91.7 (26.7) at follow-up. Despite variations among individuals in the change of fatigue, there
26 was no statistically significant difference in FISSA score over time ($p=0.087$, 95% CI -16.7 to
27 1.22). We did not find any known determinants of fatigue to be predictive of change in FISSA
28 scores.

29 **INTERPRETATION** Fatigue appears to be relatively stable within adults with CP over time,
30 with a variable presentation between individuals and across GMFCS levels. Care providers
31 should monitor and discuss fatigue with young individuals with CP to attenuate fatigue later
32 in life.

33
34 **Key words:** cerebral palsy, fatigue, adult, longitudinal study, body composition

35
36 **Address for correspondence**

37 * Correspondence to Jan Willem Gorter at Department of Pediatrics, CanChild Centre for
38 Childhood Disability Research, McMaster University, 1400 Main St. W., Hamilton, ON,
39 Canada, L8S 1C7. E-mail: gorter@mcmaster.ca

40

41 **Introduction**

42
43 Fatigue is a term used to describe a reduced capacity to sustain power output over time, and
44 is the experience of feeling tired, weak, or lacking energy.¹ There is growing recognition that
45 fatigue is a common problem in adults with cerebral palsy (CP).²⁻⁴ Findings from one study
46 documented that 41% of adults with bilateral CP were severely fatigued (Fatigue Severity
47 Scale [FSS] ≥ 5.1).² Russchen et al.⁵ found that adults with bilateral CP were more at risk for
48 high levels of fatigue than unilaterally affected adults. However, in a different study, 30% of
49 participants experienced substantial fatigue (Fatigue Questionnaire [FQ]), regardless of
50 topographical distribution of CP.⁴

51 CP is defined as “a group of permanent disorders of the development of movement and
52 posture, causing activity limitation, that is attributed to non-progressive disturbances that
53 occurred in the developing fetal or infant brain”.⁶ Although CP is a non-progressive disease,
54 individuals with CP experience a number of secondary health conditions that occur as they
55 age.^{5,7-11} Young adults with CP described fatigue as a new experience that emerges in
56 adulthood.^{7,11} Furthermore, adults with CP report functional deterioration;¹² increasing pain;¹³
57 and reduced participation in sports and social activities¹⁴ with increasing age.

58 It is of great importance to improve our knowledge about fatigue experienced by individuals
59 with CP as they age, because of the association of fatigue with bodily pain, deterioration of
60 functional skills, and low life-satisfaction in individuals with CP.⁴ Individuals with CP need to
61 know whether their fatigue is likely to improve, stay the same or get progressively worse over
62 time to plan their future. Healthcare providers need to know the factors associated with
63 change in fatigue, if any, to inform surveillance, prevention, and management.

64 To our knowledge, there is only one published longitudinal study on fatigue in adults with
65 CP.⁹ This 7-year follow-up study concluded that fatigue did not change, as measured using
66 the FQ, for adults with CP (n=149), Gross Motor Function Classification System (GMFCS)¹⁵
67 level I-IV. However, these results need to be interpreted with caution due to some limitations

68 in the study. First, the study was not specifically designed to measure fatigue over time.
69 Second, the authors used the FQ and the FSS, both of which have not been validated for
70 individuals with CP. Finally, they did not include individuals who were GMFCS level V, a
71 subgroup that is often not included in clinical research. A recent follow-up study in adults with
72 CP (GMFCS levels I-V)¹⁶ described longitudinal changes in perceived health, presence of
73 health issues and functional level in adults with CP. They concluded pain and severe fatigue
74 (dichotomized as a FSS ≥ 5.1) to be the most common health issues in 31 adults with CP and
75 found them to be predictive of perceived poor health.

76 McPhee et al.³ measured fatigue in adults with CP using the Fatigue Impact and Severity
77 Self-Assessment (FISSA), which is validated for use with adults with CP.¹⁷ They concluded
78 that participants experienced at least some fatigue across all levels of the GMFCS. Also,
79 they discovered a significant negative relationship between moderate-to-vigorous physical
80 activity (MVPA) per hour and FISSA scores, meaning higher levels of MVPA were associated
81 with decreased levels of fatigue. Furthermore, a significant positive relationship between BMI
82 and FISSA scores was discovered, meaning that increased fatigue was experienced in
83 people with a higher BMI.³ To our understanding, no one has assessed fatigue over time in
84 adults with CP using a tool that has been validated for this population. The primary objective
85 of the current study is to describe the course of fatigue over a 3-year follow-up period in
86 adults with cerebral palsy (CP) using the FISSA. The secondary objective is to investigate
87 the association between possible, previously identified, determinants of fatigue (i.e.
88 demographic characteristics [including age, distribution of CP and community ambulation]
89 and/or body composition [i.e. BMI and waist circumference (WC)]) and change in fatigue.

90 **Method**

91 ***Participants***

92 This study is part of a larger on-going program of research of cardiovascular health and
93 physical activity in adolescents and adults with CP: the Stay-FIT research program at
94 CanChild, McMaster University (www.canchild.ca).¹⁸ Participants who took part in the cross-

95 sectional study by McPhee et al. were contacted and invited to participate in this study. Our
96 study sample is a convenience sample. Persons with CP were eligible for inclusion if they
97 met each of the following criteria: ≥18 years of age and able to respond to questions with
98 some degree of independence (either independently or with assistance from another person
99 varying from having someone to help them to physically mark their answers to having
100 someone help them to think about their answers); questionnaires completed entirely by
101 parental proxy were excluded from analyses). Participants did not participate in any
102 intervention as part of the study and were not previously enrolled in any type of intervention
103 within the Stay-FIT program. Participant or parent/caregiver written consent was obtained
104 before study commencement. The study was approved by the Hamilton Integrated Research
105 Ethics Board.

106

107 ***Fatigue***

108 The FISSA is a newly developed fatigue questionnaire that has been validated for use with
109 youth and adults with CP.¹⁷ This new measure comprises 37 items aimed to provide an
110 overall total fatigue score and information specific to the impact and severity (Impact
111 Subscale) and the management (Management and Activity Modification Subscale) of
112 fatigue.¹⁷ Responses to the first 31 items are summed to provide the total score (ranging
113 from 31 (minimum) to 157 (maximum)) and are generally scored on a 5-point Likert scale
114 from Completely Disagree (1) to Completely Agree (5). A higher score indicates greater
115 fatigue.¹⁷ One question, related to the number of days of the week that fatigue is
116 experienced, is scored on a 7-point Likert scale. The remaining 6 questions are qualitative in
117 nature and are not included as part of the FISSA scores.¹⁷ The FISSA contains a framing
118 definition for participants to think about fatigue in terms of physical tiredness, muscle
119 soreness, exhaustion of your muscles and body or any related feeling. The questionnaire can
120 be used to promote discussion between individuals with CP and their clinicians about fatigue.
121 The FISSA was intended to be a clinical tool used at the individual level, to facilitate the
122 understanding of the individualized nature of fatigue. Evidence of construct validity of the

123 FISSA was provided by the ability to discriminate between groups expected to have more
124 fatigue based on functional ability and pain experiences. The FISSA demonstrated adequate
125 test-retest reliability $ICC(3,1)=0.74$ (95% CI 0.53-0.87).¹⁷

126

127 ***Variables associated with fatigue***

128 All participants were asked to self-report their GMFCS level (GMFCS Self Report
129 Questionnaire, www.canchild.ca)¹⁹, their type of motor impairment (spastic or mixed) and
130 topographical distribution (unilateral or bilateral), during the initial assessment. Type of motor
131 impairment (spastic or mixed) and topographical distribution (unilateral or bilateral) were
132 classified according to the Surveillance of Cerebral Palsy in Europe guidelines.²⁰ Height,
133 body mass and WC were re-measured by one researcher (PM) at follow-up, during the same
134 time in which the FISSA was administered. BMI (kg/m^2) was calculated, and measurement of
135 WC was performed supinely at 4 cm above the umbilicus, as previously reported.³

136

137 ***Statistical analysis***

138 Statistical analyses were performed using STATA (version 13) statistical software.
139 Descriptive summary statistics were calculated and reported as mean, standard deviation,
140 minimum, lower quartile, median, upper quartile, and maximum values for each continuous
141 variable. Nominal data (i.e. topographical distribution, GMFCS level, type of motor
142 impairment) were reported as percentages. All continuous variables were assessed for
143 normality using the Shapiro-Wilk descriptive test. A series of paired-samples t-tests were
144 performed to assess the difference between baseline (T0) and follow-up (T1) time points for
145 FISSA scores, BMI, and WC. Bivariate correlations were conducted for FISSA scores, BMI,
146 and WC between the two time points (T0 & T1). Univariate linear regression analyses were
147 performed to investigate the relationship between change in FISSA scores (dependent
148 variable) and age, change in FISSA scores and BMI, and change in FISSA scores and WC.
149 Percent variance attributable to change in FISSA scores within each univariate regression
150 analysis was tested using an analysis of variance to determine the significance of each

151 model. Exploratory analyses were performed via independent-samples t-tests to assess
152 change in FISSA scores between topographical distributions (unilateral or bilateral) as well
153 as between community ambulatory (GMFCS I-II) and community non-ambulatory (GMFCS
154 III-V) participants. As this study was a follow-up to cross-sectional research conducted by
155 McPhee et al.,³ independent-samples t-tests were conducted between participants and non-
156 participants for variables of FISSA score, age, BMI, and WC. A minimum criterion alpha level
157 of p-value ≤ 0.05 was used to determine statistical significance. A Bonferroni correction was
158 performed to prevent type I error.

159

160 **Results**

161 ***Participants***

162 FISSA questionnaires were completed by 23 (mean age 38y 2m [SD 14y 1m]; min-max 21-
163 78) of the eligible 41 participants. The remaining 18 participants did not respond to our
164 request to participate. Mean (SD) follow-up period was 3y 8m (5m). A non-responder
165 analysis showed no difference between responders and non-responders in FISSA score,
166 age, BMI, or WC (data not shown). Participant characteristics are presented in Table I. Age
167 was not normally distributed at both time points ($p < 0.05$), therefore natural log
168 transformations of the data were performed resulting in normal distributions. All other
169 continuous variables were normally distributed. It was not possible to obtain WC
170 measurements in four participants at the site of 4 cm above the umbilicus, which was
171 attributable to the presence of an intrathecal baclofen pump, enteral feeding tube, or other
172 obscurity (i.e. bandages). In these participants, WC was measured at the border of the
173 anterior superior iliac crest. In one participant WC was not measured, due to other practical
174 inconveniences.

175

176 ***Fatigue***

177 The mean (SD) FISSA score for all participants at T0 was 84 (27.7) and 91.7 (26.7) at T1.
178 There was no significant difference in mean total FISSA scores between the two time points

179 (mean difference=7.74, $p=0.087$, 95% CI -16.7 to 1.22). The minimum FISSA score at T0
180 was 34, and 36 at T1. The maximum FISSA score at T0 was 144, and 139 at T1. Overall
181 mean FISSA scores by GMFCS level are displayed in Figure 1. Individual FISSA scores for
182 each GMFCS level are depicted in Figure 2.

183 ***Variables associated with fatigue***

184 There was no significant difference in BMI between T0 and T1 (mean difference=1.65,
185 $p=0.084$, 95% CI -2.54 to 0.24). There was a significant increase in WC in the follow-up
186 cohort (mean difference=4.10, $p=0.016$, 95% CI -7.34 to -.084).

187 Pearson correlation analyses revealed significant associations between FISSA scores at T0
188 vs. T1 ($r=0.71$, $p<0.001$), BMI at T0 vs. T1 ($r=0.86$, $p<0.001$), and waist circumference at T0
189 vs. T1 ($r=0.93$, $p<0.001$).

190 Univariate regression analyses did not reveal any significant relationships between change in
191 BMI and change in FISSA scores ($r=0.376$, $p=0.077$), change in WC and change in FISSA
192 scores ($r=-0.245$, $p=0.271$), or age and change in FISSA scores ($r=-0.371$, $p=0.082$).

193 Exploratory analyses via independent samples t-tests for differences in FISSA scores
194 between topographical distribution revealed no significant difference between those who
195 were unilaterally vs. bilaterally affected ($p=0.899$). Similarly, there was no significant
196 difference in FISSA scores between those who were community ambulatory (GMFCS I-II)
197 versus community non-ambulatory (GMFCS III-V) ($p=0.341$).

198

199 **Discussion**

200 The primary objective of this study was to describe the course of fatigue, using the FISSA,
201 over a 3-year time period in adults with CP. At the group level, mean FISSA scores were not
202 statistically significantly different between baseline and follow-up time points. Secondary
203 objectives were to investigate relationships between demographical characteristics and body
204 composition and changes in FISSA scores over time. Potential determinants (WC and BMI)
205 were tested as predictors of change in FISSA scores over time, as was previously concluded

206 in the study by McPhee et al.³ In this study, we did not find changes in BMI or WC to be
207 predictive of changes in FISSA scores. This is likely attributable to the wide range of absolute
208 changes in BMI (min-max 0-14.6 kg/m²), WC (min-max 0.5-17 cm), and FISSA scores (min-
209 max 4-50), as well as the small sample size in the present study. Russchen et al.⁵ found
210 participants with bilateral CP to be more at risk for fatigue. We did not find a statistical
211 difference in FISSA change in the topographical distribution in this study. Whether or not
212 topographical distribution contributes to fatigue in adults with CP remains to be determined in
213 a larger sample.

214 We were also interested in describing changes in FISSA scores at the subgroup (within each
215 GMFCS level) and individual levels. Similar to findings by McPhee et al.,³ our follow-up
216 findings suggest that fatigue may be associated with GMFCS level, with GMFCS level II as
217 being the exception (Fig. 1). In GMFCS level II we found a higher FISSA score than levels I
218 and III. Those classified as GMFCS level II may experience difficulty walking long distances
219 on uneven terrain, and may walk with physical assistance or a handheld mobility device.²¹
220 We know from a study by Balemans et al.,²² in children and adolescents with CP (n=57), that
221 23% of participants in GMFCS level I; 47% of participants in GMFCS level II; and 71% in
222 GMFCS level III showed a VO₂walk that was higher than their anaerobic threshold. At
223 intensities above the anaerobic threshold one becomes exhausted quickly and muscles
224 become sore and painful and therefore might contribute to fatigue, which might explain the
225 increase in fatigue in GMFCS level II in the present study. This could be of interest for future
226 research.

227 In the current study, the GMFCS level III group was the only one to exhibit a decrease in total
228 FISSA score, albeit non-significant. As mentioned in the study of McPhee et al.³ and also in
229 the study above,²² it is likely that those who function at GMFCS level III and use arm
230 crutches and/or a manual wheelchair may experience greater fatigue. However, GMFCS
231 level III is a subgroup of individuals with a great variation in mobility, depending on their
232 physical abilities and personal/environmental factors. An individual could, for example, have
233 made an effort to keep climbing the stairs, but decided to stop doing that, which could

234 potentially have a positive effect on their fatigue. A previous study²³ described the probability
235 of walking among children with CP. They concluded that the probability of walking, in children
236 classified as GMFCS level III, was highest at age 9 (68%) and the probability of walking at
237 age 18 was approximately 50%. For adults, this specific probability of walking is unknown.
238 Opheim et al.⁹ found 78% of participants in GMFCS level III (n=23) to have reported a
239 deteriorated walking function over time. This variation in physical abilities and change in
240 assisted mobility over time is less likely to occur in other GMFCS levels, and might therefore
241 explain the decreased FISSA score in level III. It also reflects on individual variability within
242 the population and within the GMFCS levels, which makes it hard to give a generalizable
243 conclusion with the small sample size in our study.

244 On an individual level there are major changes found, for example in one participant the
245 FISSA score changed from 144 to 109, with a BMI decrease from 50 to 43 kg/m². In another
246 participant the FISSA score increased from 89 to 139 and BMI changed from 13 to 28 kg/m².
247 Both cases seem to reveal a relationship between BMI and FISSA score. Overall, 35% of
248 participants were found to be obese at T1 (defined²⁴ as a BMI ≥ 30 kg/m²) versus 30% at T0.
249 Central obesity (WC ≥ 88 cm for females or ≥ 102 cm for males²⁴) was found in 32% (T1)
250 versus 36% (T0). In a previous study,⁵ WC has been considered a more sensitive parameter,
251 as compared to BMI, and as being predictive for fatigue. For future research, more insight is
252 needed in the relationship between BMI/WC and fatigue by use of a gold standard measure
253 of body composition (i.e. dual energy x-ray absorptiometry) in order to differentiate which
254 should be used, as the best (and most practical) indicator for body composition, in individuals
255 with CP.

256 The current study underscores the importance of the use of the FISSA for surveillance of
257 fatigue in clinical practice and the importance of promoting discussion between clinicians and
258 their patients about fatigue. An important clinical implication of the results here is that adults
259 with CP that experience fatigue, are likely to still experience fatigue 3 years later. Moreover,
260 there is great value associated with preventing the development or worsening of fatigue to
261 maximize functional abilities and avoid deterioration with age. Care providers should

262 consider educating patients about fatigue, helping (young) adults to cope with fatigue and
263 discuss risk factors that might be modifiable (i.e. body mass).

264

265 **Study limitations**

266 While a strength of this study is the use of a fatigue questionnaire (FISSA) that has been
267 validated in adults with CP, there are some factors that should be taken into consideration
268 while interpreting our findings. Of note, we took advantage of the opportunity to follow-up
269 with a cohort that was previously recruited for a cardiovascular study³ through purposeful
270 sampling aimed at more or less equal representation at each GMFCS level. Therefore our
271 sample is not a population based sample in which a higher proportion of people would have
272 been expected in GMFCS level I. The response rate was reasonable (56%), with no
273 evidence of systematic selection bias, leaving us with 23 participants representing all
274 GMFCS levels for analysis of change in FISSA scores. To re-test our primary objective, that
275 the relationship between FISSA scores and time is greater than zero (i.e. two-tailed; p-value
276 ≤ 0.05), with 0.8 power, a sample size of 59 is required. Nevertheless, our study is the first
277 longitudinal study that provides valuable information about stability of FISSA scores over
278 time in 23 adults with CP.

279 Secondly, we do not yet know the minimal clinically important difference (MCID) of the
280 FISSA. To our knowledge, our study is the first to describe a follow-up of fatigue in adults
281 with CP using the FISSA, a fatigue tool validated in this population, and including all levels of
282 the GMFCS. Future studies should consider multi-centre studies with large samples to gain a
283 better understanding of the evaluative capacity and MCID of the FISSA.

284 Thirdly, we understand that waist circumference can be measured using different techniques
285 and there are various ways to measure body composition. Future studies should consider
286 developing a standard outcome set for body composition.

287

288 **Conclusion**

289 The results of this study indicate that fatigue remains an ongoing challenge for adults with
290 CP inclusive of all five levels of the GMFCS, with a variable presentation between
291 individuals. We did not find age or body composition (i.e. BMI, WC) to be predictive of
292 change in FISSA scores. However, the clinically important implication of our study is that
293 adults with CP who experience fatigue, are likely to remain fatigued in the future. We would
294 therefore advise care providers to monitor and talk about fatigue with adults with CP. To
295 attenuate fatigue later in life, we would suggest to begin the discussion with younger people
296 with CP.

297

298 **Acknowledgements**

299 This project is part of the larger Stay-FIT research program within *CanChild* Centre for
300 Childhood Disability Research. This work was undertaken while I.L.B. Oude Lansink was at
301 CanChild for an elective research placement in 2017 during her residency at the University
302 Medical Centre Groningen. Dr. Gorter holds the Scotiabank Chair in Child Health Research.
303 We would like to acknowledge the study participants for taking part in the study. The authors
304 have stated that they had no interests which may be perceived as posing a conflict or bias.

- 305 1. Kaasa S, Loge JH, Knobel H, Jordhøy MS, Brenne E. Fatigue. Measures and relation
306 to pain. *Acta Anaesthesiol Scand* 1999;43:939–947.
- 307 2. van der Slot WMA, Nieuwenhuijsen C, van den Berg-Emons RJG, Bergen MP,
308 Hilberink SR, Stam HJ, et al. Chronic pain, fatigue, and depressive symptoms in adults
309 with spastic bilateral cerebral palsy. *Dev Med Child Neurol* 2012;54:836–842.
- 310 3. McPhee PG, Brunton LK, Timmons BW, Bentley T, Gorter JW. Fatigue and its
311 relationship with physical activity, age, and body composition in adults with cerebral
312 palsy. *Dev Med Child Neurol* 2017;59:367–373.
- 313 4. Jahnsen R, Villien L, Stanghelle JK, Holm I. Fatigue in adults with cerebral palsy in
314 Norway compared with the general population. *Dev Med Child Neurol* 2003;45:296–
315 303.
- 316 5. Russchen HA, Slaman J, Stam HJ, van Markus-Doornbosch F, van den Berg-Emons
317 RJ, Roebroek ME. Focus on fatigue amongst young adults with spastic cerebral
318 palsy. *J Neuroeng Rehabil* 2014;11:161.
- 319 6. Rosenbaum P, Paneth N, Leviton A, Goldstein M, Bax M, Damiano D, et al. A report:
320 The definition and classification of cerebral palsy April 2006. *Dev Med Child Neurol*
321 2007;49:8–14.
- 322 7. Sandström K. The lived body - experiences from adults with cerebral palsy. *Clin*
323 *Rehabil* 2007;21:432–441.
- 324 8. Hirsh AT, Gallegos JC, Gertz KJ, Engel JM, Jensen MP. Symptom burden in
325 individuals with cerebral palsy. *J Rehabil Res Dev* 2010;47:863–876.
- 326 9. Opheim A, Jahnsen R, Olsson E, Stanghelle JK. Walking function, pain, and fatigue in
327 adults with cerebral palsy: A 7-year follow-up study. *Dev Med Child Neurol*
328 2009;51:381–388.
- 329 10. Nieuwenhuijsen C, van der Slot WMA, Dallmeijer AJ, Janssens PJ, Stam HJ,
330 Roebroek ME, et al. Physical fitness, everyday physical activity, and fatigue in
331 ambulatory adults with bilateral spastic cerebral palsy. *Scand J Med Sci Sport*
332 2011;21:535–542.
- 333 11. Brunton LK, Bartlett DJ. The bodily experience of cerebral palsy: a journey to self-
334 awareness. *Disabil Rehabil* 2013;35:1981–1990.
- 335 12. Andersson C, Mattsson E. Adults with cerebral palsy: a survey describing problems,
336 needs, and resources, with special emphasis on locomotion. 2001;43:76–82.
- 337 13. Hilberink SR, Roebroek ME, Nieuwstraten W, Jalink L, Verheijden JMA, Stam HJ.
338 Health issues in young adults with cerebral palsy: Towards a life-span perspective. *J*
339 *Rehabil Med* 2007;39:605–611.
- 340 14. L van der D, Nieuwstraten W, Roebroek M, HJ S. Functional level of young adults
341 with cerebral palsy. *Clin Rehabil* 2001;15:84–91.
- 342 15. Palisano R, Rosenbaum P, Walter S, Russell D, Wood E, Galuppi B. Development
343 and reliability of a system to classify gross motor function in children with cerebral
344 palsy. *Dev Med Child Neurol* 1997;39:214–223.
- 345 16. Benner JL, Hilberink SR, Veenis T, Stam HJ, van der Slot WM, Roebroek ME. Long-
346 Term Deterioration of Perceived Health and Functioning in Adults With Cerebral Palsy.
347 *Arch Phys Med Rehabil* 2017;98:2196–2205.
- 348 17. Brunton LK, Bartlett DJ. Construction and validation of the fatigue impact and severity
349 self-assessment for youth and young adults with cerebral palsy. *Dev Neurorehabil*
350 2017;20:274–279.
- 351 18. [https://www.canchild.ca/en/research-in-practice/current-studies/stay-fit-program-of-](https://www.canchild.ca/en/research-in-practice/current-studies/stay-fit-program-of-research-physical-activity-and-active-lifestyle-program-for-youth-with-cerebral-palsy)
352 [research-physical-activity-and-active-lifestyle-program-for-youth-with-cerebral-palsy.](https://www.canchild.ca/en/research-in-practice/current-studies/stay-fit-program-of-research-physical-activity-and-active-lifestyle-program-for-youth-with-cerebral-palsy)
- 353 19. Jahnsen R, Aamodt G, Rosenbaum P. Gross Motor Function Classification System
354 used in adults with cerebral palsy : agreement of self-reported versus professional
355 rating Gross Motor Function Classification System used in adults with cerebral palsy :
356 agreement of self-reported versus profes 2015;(2006):734–738.
- 357 20. Cans C. Surveillance of cerebral palsy in Europe: a collaboration of cerebral palsy
358 surveys and registers. *Dev Med Child Neurol* 2007;42:816–824.
- 359 21. Palisano R, Rosenbaum P, Bartlett D, Livingston M, Walter S, Russell D, et al. Gross

360 Motor Function Classification System Expanded and Revised. Ref Dev Med Child
361 Neurol 2007;39:214–223.

362 22. Balemans AC, Bolster EA, Brehm MA, Dallmeijer AJ. Physical Strain: A New
363 Perspective on Walking in Cerebral Palsy. Arch Phys Med Rehabil 2017;98:2507–
364 2513.

365 23. Palisano RJ, Hanna SE, Rosenbaum PL, Tieman B. Probability of walking, wheeled
366 mobility, and assisted mobility in children and adolescents with cerebral palsy. Dev
367 Med Child Neurol 2010;52:66–71.

368 24. National Institutes Of Health. The Practical Guide to the identification, evaluation, and
369 treatment of overweight and obesity in adults. Obesity. 2000.

370

371

372 **Figures and Tables**
 373
 374

Table I: Participant classifications and characteristics

Characteristic							Total (n=23)
Sex, <i>n</i> (%)							
	Males						9 (39)
	Females						14 (61)
Type, <i>n</i> (%)							
	Spastic CP						17 (74)
	Mixed CP						6 (26)
Distribution, <i>n</i> (%)							
	Unilateral CP						7 (30)
	Bilateral CP						16 (70)
GMFCS, <i>n</i> (%)							
	I						4 (17)
	II						5 (22)
	III						5 (22)
	IV						6 (26)
	V						3 (13)
Characteristic T0 – T1	Mean T0 (SD)	Mean T1 (SD)	Min. T0	Min. T1	Max. T0	Max. T1	
Age, y	34.6 (14.2)	38.2 (14.1)	18.0	21.0	75.0	78.0	
BMI, kg/m ²	26.1 (8.4)	27.8 (8.1)	13.3	15.7	50.0	42.6	
Waist circumference, cm ^a	83.5 (21.1)	87.6 (20.1)	54.0	56.0	142.0	135.0	
FISSA scores	84.0 (27.7)	91.7 (26.7)	34.0	36.0	144.0	139.0	

375 ^an=22, due to 1 missing measurement.
 376 Mixed CP consisted of a combination of spastic motor disorder and either dyskinetic or ataxic motor disorder.
 377 CP, cerebral palsy; GMFCS, Gross Motor Function Classification System; SD, standard deviation; Min., minimum;
 378 Max., maximum; BMI, body mass index; FISSA, Fatigue Impact and Severity Self-Assessment.
 379

380 **Figure 1: 'Oude Lansink Figure 1.pdf'**

381

382

383 **Figure 1.** Mean (SD) Fatigue Impact and Severity Self-Assessment (FISSA) scores at each Gross
384 Motor Function Classification System (GMFCS) level at T0, baseline; and T1, follow-up.

385

386

387
388
389
390

Figure 2: 'Oude Lansink Figure 2.pdf'

391 **Figure 2.** Change in Fatigue Impact and Severity Self-Assessment (FISSA) score per participant
392 (between the two time points), categorized per Gross Motor Function Classification System (GMFCS)
393 level, including mean FISSA score per GMFCS level. Each solid line represents one participant, and
394 the dashed line represents the mean FISSA score per GMFCS level. T0, baseline; T1, follow-up.

395