11-7-2018

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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.

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

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Short title: Course of fatigue in Cerebral Palsy

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

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

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

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

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

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Introduction

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

CP is defined as "a group of permanent disorders of the development of movement and posture, causing activity limitation, that is attributed to non-progressive disturbances that occurred in the developing fetal or infant brain".[6] Although CP is a non-progressive disease, individuals with CP experience a number of secondary health conditions that occur as they age.[5,7–11] Young adults with CP described fatigue as a new experience that emerges in adulthood.[7,11] Furthermore, adults with CP report functional deterioration;[12] increasing pain;[13] and reduced participation in sports and social activities[14] with increasing age.

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

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

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

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

Method

Participants

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

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

Fatigue

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

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

Variables associated with fatigue

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

Statistical analysis

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

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

Results

Participants

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

Fatigue

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

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

Variables associated with fatigue

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

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

Univariate regression analyses did not reveal any significant relationships between change in BMI and change in FISSA scores (r=0.376, p=0.077), change in WC and change in FISSA scores (r=-0.245, p=0.271), or age and change in FISSA scores (r=-0.371, p=0.082). Exploratory analyses via independent samples t-tests for differences in FISSA scores between topographical distribution revealed no significant difference between those who were unilaterally vs. bilaterally affected (p=0.899). Similarly, there was no significant difference in FISSA scores between those who were community ambulatory (GMFCS I-II) versus community non-ambulatory (GMFCS III-V) (p=0.341).

Discussion

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

in the study by McPhee et al.[3] In this study, we did not find changes in BMI or WC to be predictive of changes in FISSA scores. This is likely attributable to the wide range of absolute changes in BMI (min-max 0-14.6 kg/m^2), WC (min-max 0.5-17 cm), and FISSA scores (min-max 4-50), as well as the small sample size in the present study. Russchen et al.[5] found participants with bilateral CP to be more at risk for fatigue. We did not find a statistical difference in FISSA change in the topographical distribution in this study. Whether or not topographical distribution contributes to fatigue in adults with CP remains to be determined in a larger sample.

We were also interested in describing changes in FISSA scores at the subgroup (within each GMFCS level) and individual levels. Similar to findings by McPhee et al.,[3] our follow-up findings suggest that fatigue may be associated with GMFCS level, with GMFCS level II as being the exception (Fig. 1). In GMFCS level II we found a higher FISSA score than levels I and III. Those classified as GMFCS level II may experience difficulty walking long distances on uneven terrain, and may walk with physical assistance or a handheld mobility device.[21] We know from a study by Balemans et al.,[22] in children and adolescents with CP (n=57), that 23% of participants in GMFCS level I; 47% of participants in GMFCS level II; and 71% in GMFCS level III showed a VO_2walk that was higher than their anaerobic threshold. At intensities above the anaerobic threshold one becomes exhausted quickly and muscles become sore and painful and therefore might contribute to fatigue, which might explain the increase in fatigue in GMFCS level II in the present study. This could be of interest for future research.

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

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

On an individual level there are major changes found, for example in one participant the FISSA score changed from 144 to 109, with a BMI decrease from 50 to 43 kg/m^2. In another participant the FISSA score increased from 89 to 139 and BMI changed from 13 to 28 kg/m^2. Both cases seem to reveal a relationship between BMI and FISSA score. Overall, 35% of participants were found to be obese at T1 (defined[24] as a BMI $\geq$30 kg/m^2) versus 30% at T0. Central obesity (WC $\geq$88 cm for females or $\geq$102 cm for males[24]) was found in 32% (T1) versus 36% (T0). In a previous study,[5] WC has been considered a more sensitive parameter, as compared to BMI, and as being predictive for fatigue. For future research, more insight is needed in the relationship between BMI/WC and fatigue by use of a gold standard measure of body composition (i.e. dual energy x-ray absorptiometry) in order to differentiate which should be used, as the best (and most practical) indicator for body composition, in individuals with CP.

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

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

Study limitations

While a strength of this study is the use of a fatigue questionnaire (FISSA) that has been validated in adults with CP, there are some factors that should be taken into consideration while interpreting our findings. Of note, we took advantage of the opportunity to follow-up with a cohort that was previously recruited for a cardiovascular study[3] through purposeful sampling aimed at more or less equal representation at each GMFCS level. Therefore our sample is not a population based sample in which a higher proportion of people would have been expected in GMFCS level I. The response rate was reasonable (56%), with no evidence of systematic selection bias, leaving us with 23 participants representing all GMFCS levels for analysis of change in FISSA scores. To re-test our primary objective, that the relationship between FISSA scores and time is greater than zero (i.e. two-tailed; p-value ≤0.05), with 0.8 power, a sample size of 59 is required. Nevertheless, our study is the first longitudinal study that provides valuable information about stability of FISSA scores over time in 23 adults with CP.

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

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

Conclusion

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

Acknowledgements

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

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Figures and Tables

Table I: Participant classifications and characteristics

Characteristic		Total (*n*=23)
Sex, *n* (%)		
	Males	9 (39)
	Females	14 (61)
Type, *n* (%)		
	Spastic CP	17 (74)
	Mixed CP	6 (26)
Distribution, *n* (%)		
	Unilateral CP	7 (30)
	Bilateral CP	16 (70)
GMFCS, *n* (%)		
	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

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

Figure 1: 'Oude Lansink Figure 1.pdf'

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

Figure 2: 'Oude Lansink Figure 2.pdf'

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