UHASSELT

KNOWLEDGE IN ACTION

# Faculteit Geneeskunde en Levenswetenschappen

master in de revalidatiewetenschappen en de kinesitherapie

***Masterthesis***

***Does muscle fatigability impact on upper limb function and activity measures in children with unilateral cerebral palsy?***

**Sandra Geerits**
**Stephanie Pareyn**

Scriptie ingediend tot het behalen van de graad van master in de revalidatiewetenschappen en de kinesitherapie, afstudeerrichting revalidatiewetenschappen en kinesitherapie bij musculoskeletale aandoeningen

**PROMOTOR :**
Prof. dr. Katrijn KLINGELS

**COPROMOTOR :**
Mevrouw Lieke BRAUERS

**2017**
**2018**

UHASSELT

KNOWLEDGE IN ACTION

**www.uhasselt.be**
Universiteit Hasselt
Campus Hasselt:
Martelarenlaan 42 | 3500 Hasselt
Campus Diepenbeek:
Agoralaan Gebouw D | 3590 Diepenbeek

UHASSELT

KNOWLEDGE IN ACTION

# Faculteit Geneeskunde en Levenswetenschappen

## master in de revalidatiewetenschappen en de kinesitherapie

***Masterthesis***

***Does muscle fatigability impact on upper limb function and activity measures in children with unilateral cerebral palsy?***

**Sandra Geerits**
**Stephanie Pareyn**

Scriptie ingediend tot het behalen van de graad van master in de revalidatiewetenschappen en de kinesitherapie, afstudeerrichting revalidatiewetenschappen en kinesitherapie bij musculoskeletale aandoeningen

**PROMOTOR :**
Prof. dr. Katrijn KLINGELS

**COPROMOTOR :**
Mevrouw Lieke BRAUERS

**Acknowledgements**

This thesis is one of the most important requirements to obtain our master diploma in Rehabilitation sciences and physiotherapy at Hasselt University.

First, we gratefully thank Prof. dr. K. Klingels and Dra. L. Brauers for their guidance throughout the process of this thesis. We are also thankful to Cristina Simon Martinez for sharing data and to Prof dr. E. Rameckers for his critical remarks and advice. We also want to thank all the children who participated in this study. Finally, we also would express our thanks to give us the opportunity to participate in the therapy camp which was a very pleasant experience.

Panhovenstraat 9, 3990, Peer, Belgium, 3 June ’18 S.G.

Hoogstraat 52, 3950, Bocholt, Belgium, 3 June ’18 S.P.

**Research context**

This thesis fits in the research field of pediatric rehabilitation at the Faculty of Rehabilitation sciences and Physiotherapy at Hasselt University and REVAL Rehabilitation research group (BIOMED). It is part of the ongoing research line of Prof. dr. K. Klingels, focusing on upper limb function in children with cerebral palsy (CP), in collaboration with the Department of Rehabilitation Sciences at KU Leuven (Prof. H. Feys) and Adelante Valkenburg (Prof. Dr. E. Rameckers). Specifically, this study will focus on muscle fatigability in children with unilateral CP (UCP). So far, the amount of literature about upper limb muscle fatigability in children with UCP is limited[1,2,3]. Most of the published articles have shown inconclusive results and have not investigated the impact of muscle fatigability on activity level[4,5,6]. As it may be hypothesized that muscle fatigability, together with muscle weakness, impact children's functional level, it is essential to determine the relationship between muscle fatigability and body function and activity measures. These insights can result in new developments in targeted interventions and thus further optimize treatment implementations.

The design of this study was determined by Prof. dr. K. Klingels. The two master students Rehabilitation Sciences and Physiotherapy of Hasselt University actively participated in a two week therapy camp in Diepenbeek investigating the effects of constraint-induced movement therapy (CIMT) and action observation therapy (AOT) for children with UCP. Besides, they assisted the researchers of KU Leuven with the clinical assessments of these children. The pre-assessment data from the therapy camps between 2014 and 2017 were used for this cross-sectional study. L. Brauers and C. Simon Martinez converted all data to a usable and clear dataset. The two students performed the statistical analyses, interpreted and discussed the results to form a conclusion. Proofreading and supervision was performed by promotor Prof. dr. K. Klingels and copromotor L. Brauers.


1. Eken, M. M., Dallmeijer, A. J., Doorenbosch, C. A., Dekkers, H., Becher, J. G., & Houdijk, H. (2014). Assessment of muscle endurance of the knee extensor muscles in adolescents with spastic cerebral palsy using a submaximal repetitions-to-fatigue protocol. *Arch Phys Med Rehabil, 95*(10), 1888-1894. doi:10.1016/j.apmr.2014.05.010
2. Eken, M. M., Dallmeijer, A. J., Houdijk, H., & Doorenbosch, C. A. (2013). Muscle fatigue during repetitive voluntary contractions: a comparison between children with cerebral palsy, typically developing children and young healthy adults. *Gait Posture, 38*(4), 962-967. doi:10.1016/j.gaitpost.2013.05.004

3. Moreau, N. G., Li, L., Geaghan, J. P., & Damiano, D. L. (2008). Fatigue resistance during a voluntary performance task is associated with lower levels of mobility in cerebral palsy. *Arch Phys Med Rehabil, 89*(10), 2011-2016. doi:10.1016/j.apmr.2008.03.012

4. Brauers, L., Geijen, M. M., Speth, L. A., & Rameckers, E. A. (2017). Does intensive upper limb treatment modality Hybrid Constrained Induced Movement Therapy (H-CIMT) improve grip and pinch strength or fatigability of the affected hand? *J Pediatr Rehabil Med, 10*(1), 11-17. doi:10.3233/prm-170406

5. Doix, A. C., Gulliksen, A., Braendvik, S. M., & Roeleveld, K. (2013). Fatigue and muscle activation during submaximal elbow flexion in children with cerebral palsy. *J Electromyogr Kinesiol, 23*(3), 721-726. doi:10.1016/j.jelekin.2012.12.005

6. van Meeteren, J., van Rijn, R. M., Selles, R. W., Roebroeck, M. E., & Stam, H. J. (2007). Grip strength parameters and functional activities in young adults with unilateral cerebral palsy compared with healthy subjects. *J Rehabil Med, 39*(8), 598-604. doi:10.2340/16501977-0095

**Does muscle fatigability impact on upper limb function and activity measures in children with unilateral cerebral palsy?**

BACKGROUND: Children with unilateral cerebral palsy (UCP) not only experience muscle weakness, spasticity and impaired control but also muscle fatigability. However, studies exploring the impact of muscle fatigability on body function and activity measures in children with UCP are currently lacking.

OBJECTIVES: (1) To gain insights into muscle fatigability according to anthropometric characteristics; (2) to investigate the relation between muscle fatigability and upper limb impairments; (3) to study the impact of muscle fatigability on activity measures in children with UCP.

PARTICIPANTS: Forty-one children with UCP (25 males, 16 females; mean age 9 years, 5 months (SD 1 year 11 months); range 6-15 years) met the inclusion criteria. According to the Manual Ability Classification System (MACS), eight children were classified as level I, 14 as level II and 19 as level III.

MEASUREMENTS: To measure muscle fatigability, a sustained contraction during 30 seconds with a digital JAMAR dynamometer was performed. Afterwards the static fatigue index (SFI) was calculated. According to body functions, passive range of motion, muscle tone, grip strength, muscle strength and sensory function were measured. The Jebsen-Taylor Test of Hand Function, Abilhand-Kids, Melbourne Assessment and Assisting Hand Assessment were used to evaluate unimanual capacity and bimanual performance.

RESULTS: The mean SFI of the affected hand (AH) and non-affected hand (NAH) of the total group were 51.75% (SD 16.50%) and 46.54% (SD 11.16%) respectively (p=0.007). SFI correlated fairly with grip strength ($r_p$=–0.43, p=0.005), shoulder muscle strength ($r_s$=–0.50, p=0.001), total muscle strength ($r_p$=–0.32, p=0.044) and stereognosis ($r_s$=–0.38, p=0.018). For activity measures, only Abilhand-kids correlated significantly with SFI ($r_p$=−0.31, p=0.05). Shoulder muscle strength and stereognosis were significant predictors, explaining 33.60% of variance in SFI.

CONCLUSION: Our findings suggest that shoulder muscle strength and stereognosis are fairly related to muscle fatigability in children with UCP. No correlations were found for most of the activity measurements.

**Introduction**

Cerebral palsy (CP) is defined as a group of permanent disorders of the development of movement and posture, causing activity limitations, that are attributed to non-progressive disturbances occurring in the developing fetal or infant brain[1]. The motor disorders of CP are often accompanied by disturbances of sensation, perception, cognition, communication, behavior or seizures[1]. The prevalence of CP in Europe is approximately three per 1000 newborns. Unilateral CP (UCP) occurs in 29% to 44% of these children[2,3,4].

In children with UCP, increased muscle tone and weakness in the distal muscles are the most noticeable motor impairments in the upper limb[5]. According to sensory function, children with UCP in particular experience deficits in stereognosis and two-point discrimination[5,6,7]. These motor and sensory impairments result in difficulties in upper limb activities like eating, drinking or carrying objects[8]. Nevertheless, children with UCP not only experience weakness, spasticity and impaired control but also fatigability[9,10,11]. Muscle fatigability can be defined as a reduction in muscle force-generating capacity in the neuromuscular system that occurs during prolonged or ongoing activity[12]. It may be hypothesized that besides adequate strength levels, also the ability to sustain a muscle contraction is important in daily life activities[13]. However, insights in muscle fatigability and its impact on activity measures in children with UCP are currently lacking.

The inability to produce and sustain sufficient muscle strength may be both centrally, as well as peripherally originated in this population[14]. Centrally originated deficits are related to the inadequate recruitment of motor neurons by the central nervous system. This results in poor coordination between agonist and antagonist muscle activation and altered patterns of muscle recruitment[15]. On peripheral level, research has shown that children with CP have more type 1 fibers and less type 2 fibers compared to typically developing children[16]. Also, children with CP have reduced cross-sectional area of spastic muscles, increased elastic modulus and sarcomere length, muscle fiber atrophy and increased levels of intramuscular fat and connective tissue. This results in impaired grip-lift force synergies and increased force variability which may lead to inaccurate daily life movements[17,18,19].

So far, the amount of literature about muscle fatigability in children with UCP is limited[20,21,22]. Only three studies have investigated muscle fatigability in the upper limbs in individuals with

UCP and reported conflicting results[23,24,25]. In the study of van Meeteren et al. (2007), young adults with UCP showed less maximum grip strength, coordination and endurance in both hands compared to healthy young adults[25]. On the contrary, Doix et al. (2013), found no difference in endurance time and electromyogram (EMG) median frequency slope between adolescents with UCP and typically developing (TD) adolescents during an isometric elbow flexion. They only found a decrease in EMG-amplitude in adolescents with UCP[24]. Higher EMG-amplitude is the result of increasing amounts of recruited motor units. This indicates impaired motor unit recruitment occurring in adolescents with CP[26]. The conflicting results in these studies could be explained by the differences in methodology in quantifying muscle fatigability and the small sample sizes. Finally, an intervention study from Brauers et al. (2017) investigated the effects of Hybrid-Constrained Induced Movement Therapy (H-CIMT) on grip and pinch strength and fatigability. They found a significant increase in pinch strength of the affected hand and a tendency towards a decrease in muscle fatigability during the pinch test in children with UCP[23].

In patients with multiple sclerosis (MS), upper limb muscle fatigability has been studied more extensively. Studies showed its impact on ADL, participation and quality of life[27,28,29]. In this population, muscle fatigability has been calculated by the ''Static Fatigue Index''. This index is quantified during a 30 seconds maximum isometric grip strength task and is defined as the ratio between the area under the curve and the hypothetical area under the curve if no strength loss would occur. The index has proven to be valid and reliable in patients with MS[27,30].

In conclusion, research on muscle fatigability in the upper limb in children with UCP is limited and the conflicting results and small sample sizes necessitate further research. Moreover, cross-sectional studies exploring the relation between muscle fatigability and body function and activity measures are currently lacking. Therefore, the objectives of this study were 1) To gain insights into muscle fatigability according to anthropometric characteristics; 2) to investigate the relation between muscle fatigability and other upper limb impairments, and 3) to study the impact of muscle fatigability on activity measures in children with UCP. These insights are crucial to predict the child's functional level and may help in optimizing individual treatment interventions.

## Method

### *Participants*

Forty-one children with UCP were recruited at the University Hospital Pellenberg in Leuven, Belgium between 2014 and 2017. Inclusion criteria were:

1) Confirmed diagnosis of UCP;
2) Aged between 6 and 15 years;
3) Sufficiently cooperative to comprehend and complete the test procedures;
4) Minimal ability to actively grasp and stabilize an object with the more impaired hand.

Children were excluded if they underwent a surgical procedure in the upper limb two years prior to testing or Botulinum toxin injections within the last 6 months prior testing. The protocol was approved by the Ethical Committee of the KU Leuven (METC trialnr. ML9913) and the University of Maastricht (METC trialnr. 015095). An informed consent was signed by the parents.

### *Procedure*

Characteristics of the children such as age, sex, affected side and the Manual Ability Classification System (MACS)[31] were obtained. Children were tested at the University Hospital Pellenberg by two well-trained physiotherapists.

Static Fatigue Index and grip strength

*Muscle endurance and maximal grip strength* was measured with the digital JAMAR handgrip dynamometer (E-link, Biometrics Ltd, Newport, UK). All children were measured in the same standardized position. They were seated on a chair with back support and feet flat on the ground. The elbow was placed in 90° flexion with the forearm supported on the table in neutral position between supination and pronation. The wrist was in a neutral position between flexion and extension. The children were asked to squeeze as hard as possible for maximal three seconds. Three trials were performed with a rest of 30 seconds between the different trials. The mean maximal force of each child was used for analysis. Grip strength of the affected hand (AH) showed high inter-rater and test-retest reliability in children with UCP, respectively ICC 0.95 and 0.96[32].

To measure *muscle fatigability*, the children performed a maximal contraction during 30 seconds. The children were asked to squeeze the Jamar dynamometer for 30 seconds as hard as possible. They were verbally encouraged and received visual feedback about the remaining test time. Afterwards the static fatigue index (SFI) was calculated. This index is based on the assumption that the maximum force is reached in the first 10 seconds and after this point muscle fatigability can be measured. According to this assumption, the absolute peak force (kgs) and the time to peak moment (TPM) was determined first. Secondly, the hypothetical area under the curve (HAUC) was defined. The HAUC (area B and C) represents the curve that is obtained when the participant keeps its force constant for 30 seconds and is calculated by multiplying the maximal handgrip strength (in kg) with 30 minus time of maximal hand grip strength (Tmax). Thereafter the actual area under the curve (AUC) was calculated. This calculation is based on the area under the force-time curve from Tmax to 30s (area C). SFI was calculated as follows: $\mathrm{SFI} = 100\% \times \left(1 - \frac{\mathrm{C}}{\mathrm{B+C}}\right)$. The SFI calculations were performed using Matlab (MathWorks Matlab version 2017b). A decrease in maximal grip strength over time results in higher levels of SFI percentage, which indicates higher muscle fatigability. In Figure 1, a schematic overview of the strength-time curve shows the different areas to calculate the SFI. A pilot study from Brauers L. showed a moderate test-retest reliability for the SFI of the handgrip dynamometer with ICC 0.73 in the AH and ICC 0.72 in the non-affected hand (NAH)[33].

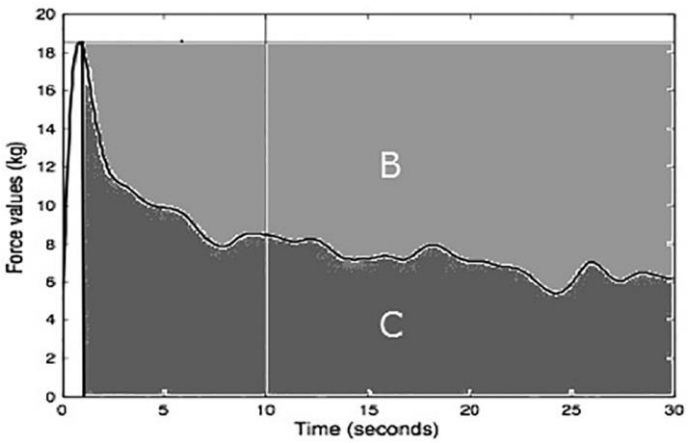


*Figure 1:* Static Fatigue Index: strength-time curve during isometric contraction of 30s: the fatigue index is based on the area under the curve (C) and the hypothetical area under the curve (B+C)[33].

Body function measurements

To investigate upper limb impairments, passive range of motion (PROM), muscle tone, grip strength, muscle strength and sensory function were measured.

*Passive range of motion (PROM)* of the shoulder (anteflexion, abduction, external rotation, internal rotation), elbow (extension, supination), wrist (extension) and fingers (thumb adduction and extension in proximal and distal interphalangeal joints) was measured with a universal goniometer. Data were converted to dichotomous scores, where score 0 and 1 corresponds to 'no movement limitation' and 'a deficit of 10° or more' respectively. A total score (0-10) as well as subscores for the muscle groups of the shoulder (0-4), elbow (0-2), wrist and hand (0-4) were calculated.

The modified Ashworth Scale (MAS) was used to grade the level of *spasticity* on a 6-point scale, ranging from 0 to 4[34]. Following muscle groups were evaluated: shoulder abductors, adductors, extensors, internal- and external rotators; elbow flexors, extensors and pronators; wrist flexors and extensors; finger flexors and thumb abductors. A total score was calculated (0-48) as well as muscle groups of the shoulder (0-20), elbow (0-12) and wrist and hand (0-16).

Manual muscle testing (MMT) was used to determine *muscle strength* of the shoulder flexors, abductors and adductors; elbow extensors, flexors, supinators and pronators and wrist extensors and flexors. This is an 8-point ordinal scale ranging from 0 to 5[35]. A total score was calculated (0-45) as well as subscores for the muscle groups of the shoulder (0-15), elbow (0-20) and wrist (0-10).

To assess *sensory function*, four modalities were evaluated: exteroception, proprioception, two-point discrimination (TPD) and stereognosis. Exteroception was examined by assessing tactile sense of the index finger, thumb and hand palm. The examiner lightly touched three times each site. The child was asked to reply 'yes' each time a stimulus was felt. Proprioceptive sense was examined by moving the distal index finger, the child was asked to respond immediately if motion was perceived. When small amplitudes were not felt, larger amplitudes were used. Both modalities were scored on a 3-level-scale, where score 0 means absent, score 1 reduced and score 2 normal. First the NAH was measured and thereafter the AH. An aesthesiometer was used to evaluate the TPD. The distal phalanx of the index finger was tested. In five consecutive trials, the minimal distance in millimeters (mm) at which the child could correctly distinguish one or two discrete points was reported. Following

scoring criteria were applied: less or equal than 5 mm corresponds with score 2, between 6 and 10 mm score 1 and more than 10 mm score 0. Stereognosis was evaluated by recognizing and identifying six different objects through tactile manipulation. If all objects where recognized with the affected hand a score 2 was given. A score 1 means recognizing 4 or 5 out of 6 objects and score 0 if less than 3 objects were recognized.

For all these motor and sensory assessments, Klingels et al. (2010) showed a moderate to very high interrater and test-retest reliability in children with UCP[32].

Activity level measurements

To evaluate the activity level, the Jebsen-Taylor Test of Hand Function (JTTHF), Abilhand-Kids, Melbourne Assessment of Unilateral Upper Limb Function (MUUL) and the Assisting Hand Assessment (AHA) were used.

*The Jebsen-Taylor Test of Hand Function* is used to measure speed and dexterity in functional tasks. This test includes six timed tasks to evaluate speed in each hand[36]. This test has a very high interrater reliability and has shown to be responsive in children with CP[37].

*The Melbourne Assessment-2* is an unilateral qualitative evaluation tool which measures the capacity of the AH. The test evaluates different functions like manipulation, grasping, reaching and releasing. Fourteen tasks are filmed and then scored via four items on a 3 or 5-level-scale[38]. The test has shown a high internal consistency, test-retest- intra- and interrater reliability[38].

The bimanual performance was measured with the *Assisting Hand Assessment*, version 4.4 or 5.0. The spontaneous use of the AH was evaluated on 4-level-scale[39] in several items such as the general use, use of the arms, grasping and releasing, fine-motor adjustments, coordination and speed. Raw scores were converted to logit-based 0-100 AHA units[40]. This test is a valid and has an excellent inter- and intrarater reliability[41].

The performance of activities in daily life was evaluated with *the Abilhand-Kids Questionnaire*, which contains 21 mainly bimanual activities for which parents score the difficulty on a 3-level-scale (impossible, difficult or easy to perform)[42]. Raw scores were converted to logits. High levels of reliability and validity were shown for these outcome measures[43].

***Statistical analysis***

The general and clinical characteristics of the subjects were documented using descriptive statistics. Data distribution was tested by the Shapiro-Wilk test. The data of grip strength AH and JTTHF were not normally distributed, therefore a logarithmic transformation was performed. Descriptive statistics (mean values and standard deviation (SD)) and one-way analysis of variance (ANOVA) were applied to determine differences between the MACS levels. In case of significant differences between groups, post-hoc Holm-Bonferroni tests were performed. To investigate the correlation between muscle fatigability and age, upper limb impairments and activity measures, correlation coefficients were calculated. Depending on the type of the data Spearman's rank ($r_s$) correlations or Pearson correlations ($r_p$) were used. Following criteria were considered: correlation coefficients >0.70: high association, 0.50 – 0.70: good association, 0.30 - 0.50: fair association, <0.30: weak or no association[44]. The significant correlations were entered in a stepwise multiple linear regression model to determine which parameters could explain muscle fatigability. The level of statistical significance was set at $p < 0.05$. All statistical analyses were conducted using IBM SPSS Statistics V25.0 software.

## Results

### *Participants*

Forty-one children diagnosed with UCP (25 boys, 16 girls; 24 left-side affected, 17 right-side affected) were included in this study. Mean age was 9 years 5 months (SD 1 year 11 months). MACS-level II and III were most common, respectively 14 (34%) and 19 (46%) children. Eight (20%) children had MACS-level I.

### *Static Fatigue Index and grip strength*

Descriptive characteristics

Descriptive statistics of the SFI and grip strength are shown in Table 1.

The mean SFI of the AH for the boys and girls were 49.45% (SD 17.15%) and 55.34% (SD 15.26%) respectively ($p = 0.271$). For the total group children, the mean SFI of the AH was significantly higher than the NAH, with mean (SD) values of 51.75% (16.50%) and 46.54% (11.16%) ($p = 0.007$). The mean ratio for SFI of the AH to the NAH was 111% (SD 25%). There was no significant correlation between SFI and age for the AH ($r_s = –0.23$, $p = 0.141$) (Figure 2A). However, the SFI of the NAH was significantly correlated with age ($r_s = −0.32$, $p = 0.045$), with younger children showing higher muscle fatigability.

The mean grip strength of the AH for the boys and girls were 4.92 kg (SD 3.14 kg) and 4.89 kg (SD 3.12 kg) ($p = 0.894$).The mean grip strength of the AH and NAH for the total group children was respectively 4.91 kg (SD 3.09 kg) and 13.50 kg (SD 4.93 kg) ($p < 0.0001$). The mean ratio for grip strength of the AH to the NAH was 40% (SD 20%). For grip strength and age, a significant positive correlation was found for AH and NAH, respectively $r_s = 0.51$ ($p = 0.001$) and $r_s = 0.68$ ($p < 0.001$) (Figure 2B).

**Table 1: .**
*Descriptive statistics of the Static Fatigue Index and grip strength.*

| | SFI (%) Mean (SD) | Grip strength (kg) Mean (SD) |
|---|---|---|
| **AH** | 51.75 (16.50) | 4.91 (3.09) |
| **NAH** | 46.54 (11.16) | 13.50 (4.93) |
| **p-value[a]** | 0.007 | <0.0001 |
| **Boys (N= 25)** | 49.45 (17.15) | 4.92 (3.14) |
| **Girls (N= 16)** | 55.34 (15.26) | 4.89 (3.12) |
| **p-value[b]** | 0.271 | 0.894 |
| **MACS I (N= 8)** | 53.71 (8.80) | 7.84 (3.81) |
| **MACS II (N= 14)** | 47.41 (16.04) | 5.50 (3.12) |
| **MACS III (N=9)** | 54.12 (19.15) | 3.24 (1.24) |
| **p-value[b]** | 0.490 | < 0.0001 |

SFI, Static Fatigue Index; SD, standard deviation; AH, Affected hand; NAH, Non-affected hand; N, total number of children; MACS, Manual Ability Classification System;
[a] paired samples statistics
[b] one-way analysis of variances (ANOVA)

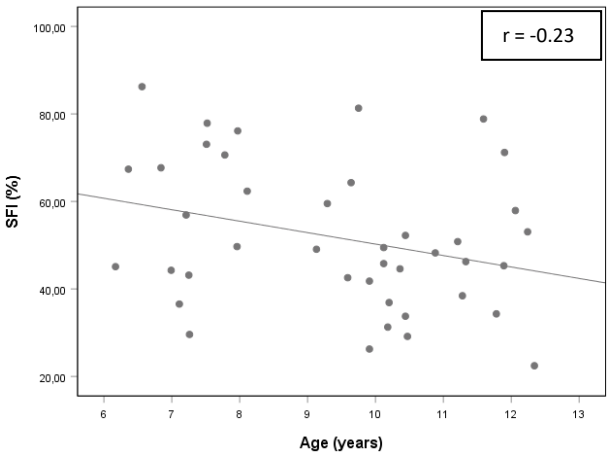


*Figure 2A:* Scatter plot Static Fatigue Index affected hand (%) by age (years).

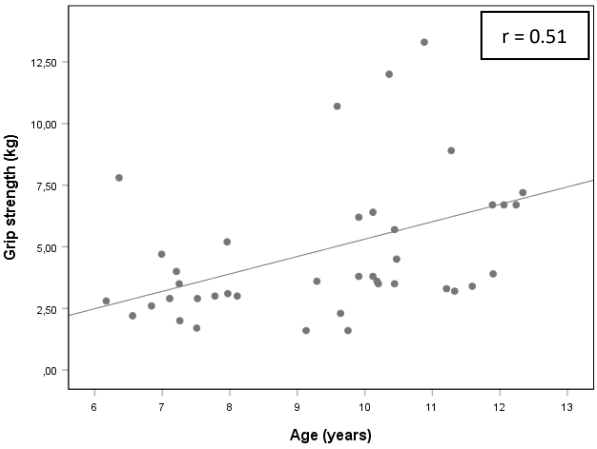


*Figure 2B:* Scatter plot grip strength affected hand (kg) by age (years).

Comparison between MACS levels

Figure 3A-B and Table 1 presents mean values and SD of grip strength and SFI across the different MACS levels. The highest mean SFI was found in MACS III (54.12%, SD 19.15%). The SFI of MACS I and II were 53.71% (SD 8.80%) and 47.41% (SD 16.04%) respectively. However, one-way analysis of variance showed no significant differences between all levels ($p = 0.490$). For grip strength, significant differences were found ($p < 0.0001$). Post-hoc analyses showed significant differences between MACS I and III ($p < 0.0001$) and MACS II and III ($p = 0.019$).

Children in MACS III showed lower levels of grip strength compared with MACS I and II, indicating that grip strength decreases as limitations in manual ability increase.

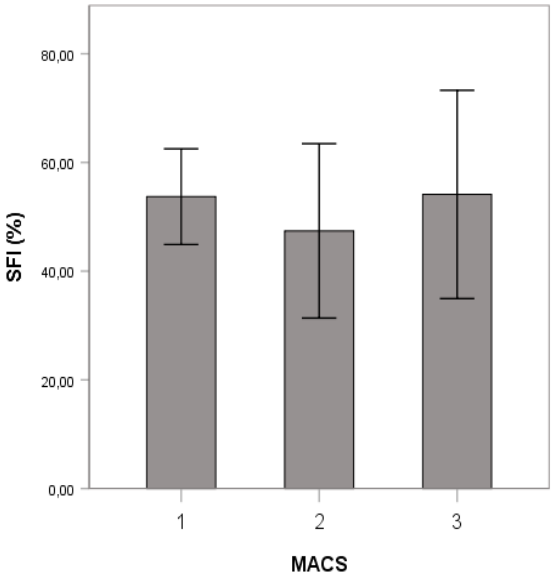


*Figure 3A*: Mean values and standard deviation of the Static Fatigue Index affected hand (%) by MACS.

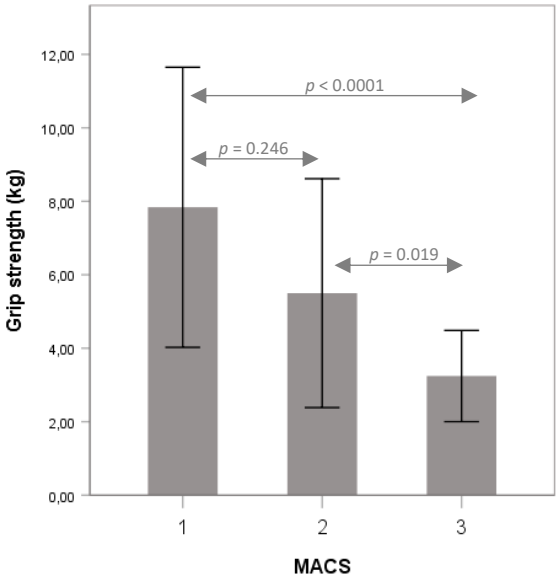


*Figure 3B*: Mean values and standard deviation of grip strength affected hand (kg) by MACS. The p-values of the post-hoc analyses are presented.

***Relationship between SFI and body function***

Appendix 1 shows an overview of descriptive values of the outcome measures at body function and activity level. Correlations between muscle fatigability of the AH and PROM, muscle tone, grip strength, muscle strength and sensory function are presented in Table 2. For the PROM and muscle tone, no to weak correlations were found with muscle fatigability for total scores as well as for subscores ($r < 0.13$, $p > 0.409$). A fair correlation was found for total muscle strength ($r_p = –0.32$, $p = 0.044$) and a good correlation between SFI and shoulder strength ($r_s = –0.50$, $p = 0.001$). Elbow and wrist muscle strength showed no to weak correlations, $r_s = –0.25$, $p = 0.166$ and $r_s = –0.01$, $p = 0.965$, respectively. A fair correlation was found with grip strength ($r_p = –0.43$, $p = 0.005$).

For sensory measures, a fair correlation between SFI and stereognosis was found ($r_s = –0.38$, $p = 0.018$). Other sensibility modalities were poorly correlated with SFI ($r_s > –0.27$, $p > 0.10$). Scatterplots of the significant correlations are shown in Figures 4A-D.

To investigate to which degree the variability of SFI was explained by body function measures a forward stepwise multiple regression analysis was performed. The significant correlations were entered in a stepwise linear regression model, which revealed that shoulder muscle strength ($R^2$ = 24.20%, adjusted $R^2$ = 22.10%, p = 0.002) and stereognosis ($R^2$ = 9.40%, adjusted $R^2$ = 7.60%, p = 0.035) were significant predictors, explaining 33.60% of the variance in SFI.

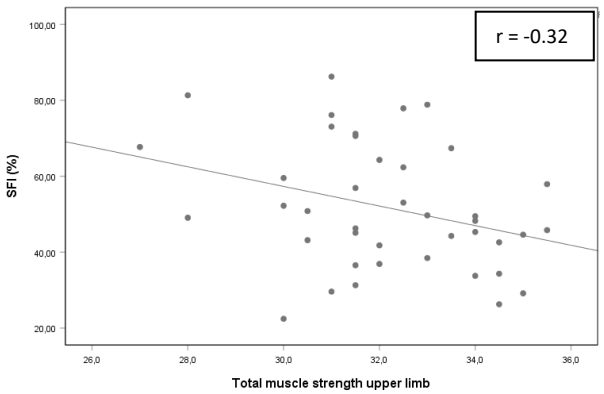


*Figure 4A:* Scatter plot Static Fatigue Index (%) by total muscle strength upper limb.

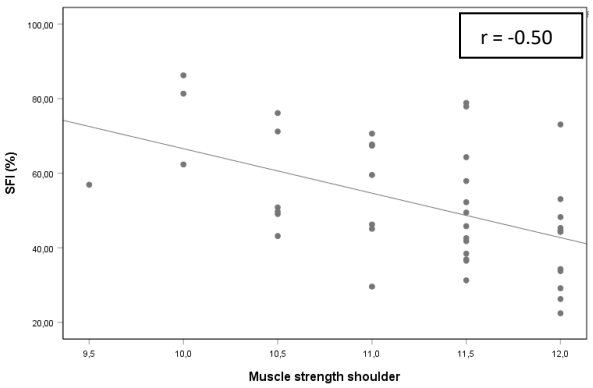


*Figure 4B:* Scatter plot Static Fatigue Index (%) by muscle strength shoulder.

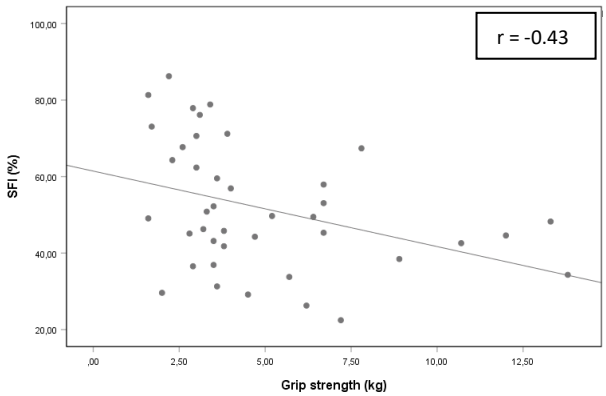


*Figure 4C*: Scatter plot Static Fatigue Index (%) by grip strength (kg).

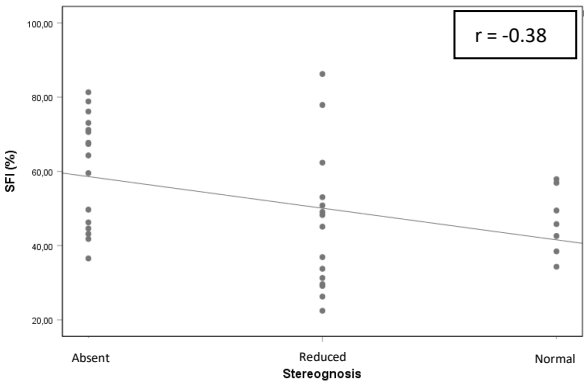


*Figure 4D*: Scatter plot Static Fatigue Index (%) by stereognosis.

### *Relationship between SFI and activity measures*

Pearson correlation coefficients between SFI and activity measures are shown in Table 2. For the Abilhand-Kids, a fair correlation was found ($r_p$ = –0.31, p = 0.050) (Figure 5). No to weak correlations were found with JTTHF, AHA and Melbourne Assessment ($r_p$ < 0.29, p > 0.077).

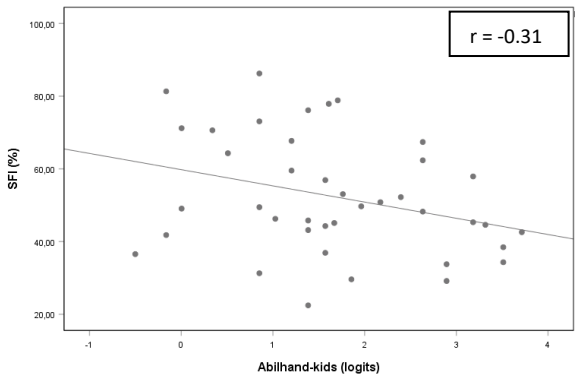


*Figure 5:* Scatter plot SFI (%) by Abilhand-kids (logits).

**Table 2:**
*Correlation coefficients between body function and activity measures and the Static Fatigue Index (SFI).*

| | N | SFI (%) |
|---|---|---|
| **BODY FUNCTION MEASURES** | | |
| PROM (0-10)[b] | 41 | 0.07 |
| - Shoulder[b] | | 0.07 |
| - Elbow[b] | | 0.13 |
| - Wrist + hand[b] | | −0.02 |
| Muscle tone (0-48)[a] | 41 | −0.12 |
| - Shoulder[b] | | −0.11 |
| - Elbow[b] | | –0.03 |
| - Wrist + hand[b] | | −0.01 |
| Muscle strength (0-45)[a] | 41 | −0.32* |
| - Shoulder[b] | | −0.50** |
| - Elbow[b] | | −0.25 |
| - Wrist[b] | | –0.01 |
| Log Grip strength[a] | 41 | −0.43** |
| Sensibility | 38 | |
| - Exteroception[b] | | −0.03 |
| - Proprioception[b] | | −0.27 |
| - TPD[b] | | −0.21 |
| - Stereognosis[b] | | −0.38* |
| **ACTIVITY MEASURES** | | |
| Log JTTHF[a] | 41 | 0.29 |
| Abilhand Logits[a] | 40 | –0.31* |
| Melbourne assessment[a] | 41 | –0.28 |
| AHA Units[a] | 39 | –0.22 |

N, total number of children; SFI, Static Fatigue Index; PROM, passive range of motion; log, logaritm; TPD, two-point discrimination; JTTHF, Jebsen-Taylor Test of Hand Function; Abilhand, Abilhand-Kids Questionnaire; AHA, Assisting Hand Assessment.
*p<0.05; **p<0.01.
[a], pearson correlation coefficient
[b], Spearman's rho correlation coefficient

**Discussion**

The aim of this study was to gain insights into muscle fatigability according to anthropometric characteristics and to investigate the relation between muscle fatigability and body and activity measures in 41 children with UCP. So far, these relationships have not been addressed extensively. Although according to Klingels et al. (2012) and Jahnsen et al. (2003), it may be hypothesized that muscle fatigability, together with muscle weakness, have an important impact on children's functional level and daily activities[5,9].

First, descriptive statistics of the SFI and grip strength were performed to achieve a general overview of muscle fatigability in children with UCP. Our results showed a clear difference of 60% in grip strength between the AH and NAH ($p < 0.0001$), which was in line with previous studies[5,25]. Probably, this difference can be explained by both centrally as peripherally originated deficits in children with UCP. Centrally originated deficits are related to damage in the motor cortex or cerebellum which results in inadequate control of the monosynaptic reflex and low levels of voluntary activation. Consequently, this leads to impairments such as spasticity and muscle weakness in the AH[14,15,18,45]. On peripheral level, changes in biomechanical properties of the muscle (muscle fiber distribution, sarcomere length, …) may contribute to impaired force production[17,18,19].

For the SFI, a significant difference was found between the AH and NAH, with 11% more muscle fatigability in the AH compared to the NAH. Previous studies[25] found no significant difference for muscle fatigability between both hands in young adults with UCP, measured by using the SFI. Compared to typically developing children, two studies[8,21] found less muscle fatigability in children and young adults with CP compared to typically developing children, measured with an isokinetic dynamometer in knee flexors and extensors. These authors attributed the lower muscle fatigability to lower muscle strength in people with CP. Several mechanisms can be taken into account. As described previously, children with CP have lower levels of voluntary activation which lead to difficulties in motor unit recruitment[45]. Because type I muscle fibers can be recruited with lower firing rates, these children preferentially recruit these fibers[8]. Furthermore, children with CP have more type 1 fibers and less type 2 fibers compared to typically developing children[16,46] and the former are characterized to be more fatigue resistant.

This study also investigated the correlation between age, SFI and grip strength. The results showed a decrease of SFI of the AH with an increase of age, but this was a non-significant correlation. As literature has shown that children with CP have more type 1 fibers with increasing age[16,46], this seems a reasonable explanation for our findings. This relationship has not been investigated yet in typically developing children and children with CP. In contrast, our study showed a significant positive correlation between grip strength of the AH and age. This age-related increase is in line with previous studies[47,48]. Klingels et al. (2012) showed that grip strength of the affected hand increased significantly over one year in children with UCP[47]. Also for finger grip strength, Blank et al. (2009) found an increase by 10-15% in both hands of preschool children with spastic CP after a one-year follow up[48]. It is possible that with increasing age, children develop more muscle tissue and become stronger[49].

A further comparison was performed between the different MACS levels. According to our clinical expectations the highest mean SFI was seen in MACS III, however no significant differences were found between MACS levels. The fact that children in MACS I experience more fatigability compared to those of MACS II is unclear. However, it should be considered that MACS I consisted of approximately 50% less children compared to MACS II. Also, results showed a large variability in SFI across the MACS levels. Moreover, the MACS is a classification system based on the ability to handle objects in daily activities in children with CP. It does not categorizes them according to their degree of impairment. The distribution across the three MACS levels was in contrast with the study of Brauers et al. (2017). They found the highest mean SFI in MACS II (57.87%, SD 10.87%), followed by MACS III (56.86%, SD 3.38%) and MACS I (46.24%, SD 8.93%)[23]. An expected relation was found between grip strength and MACS levels whereas grip strength decreases with increasing MACS levels. This was consistent with previous studies[5,23].

The second aim of this study was to investigate the relation between SFI and body function impairments. Significant relations were found between the SFI and grip strength, shoulder strength, total muscle strength and stereognosis. Our results indicated that both proximal and distal muscle weakness were related to muscle fatigability. However, correlations were only moderate and only shoulder muscle strength was a significant predictor. Interestingly, there was a fair negative correlation between muscle fatigability and grip strength meaning that increasing SFI correlates with decreasing grip strength. This is an important clinical finding

because it is reported that grip strength is a strong predictor for unimanual and bimanual activities[5,50]. However, this finding is not in line with previous studies[21,51]. Meldrum et al. (2007), who investigated maximum muscle strength and muscle fatigability in healthy adults, found no correlation between these parameters[51]. On the other hand, Eken et al. (2013) found that less muscle fatigability corresponded with less maximum muscle strength in the knee flexors and extensors in children with CP[21].

Furthermore, our findings revealed that children with impaired stereognosis experience more fatigability. This was the first study to examine the relationship between sensory measures and muscle fatigability in children with UCP. Also in other neurological disorders, the relation between stereognosis and fatigability seems unclear. Therefore, comparisons with other studies are difficult. However, in children with CP, Arnould et al. (2007) found significant positive correlations for grip strength with stereognosis and proprioception[52]. This indicates a better sensory function with increasing grip strength. In addition, previous studies demonstrated that stereognosis is most affected of all sensory functions in children with UCP, probably due to hierarchical functional anatomical organization of the central nervous system[5-7]. Stereognosis appeared to be a more cortical function compared to proprioception and exteroception which are labelled as basic sensory functions[5].

Finally, the population in this study demonstrated few deficits for PROM and muscle tone possibly explaining the no to weak correlations with SFI.

A further aim was to study the relation between SFI and activity measures. The current literature investigating the relationship between other upper limb impairments and manual ability suggested that motor functions such as grip strength, dexterity and spasticity interfere with manual ability[5,52]. In addition, Jahnsen et al. (2003) found that fatigability plays an important role in children with CP. They found bodily pain, deterioration of functional skills, limitations in emotional and physical role function, and low life satisfaction as the strongest predictors associated with fatigability[9]. Therefore, it is essential to explore the relationship between muscle fatigability and unimanual capacity and bimanual performance. To our knowledge, this is the first study to examine this relationship in children with CP. However, in persons with MS it has been demonstrated that distal muscle strength and proximal muscle endurance are related with the use of the arm in daily life activities measured with the Manual Ability Measures-36[53]. Our results showed only a significant correlation between SFI and

Abilhand-Kids. It appears that muscle fatigability is no predictive factor in daily life activities in children with UCP. This might be explained by the fact that activity measurements used in this study includes predominantly submaximal and short activities such as opening a crisp bag (Abilhand-Kids) or turning over cards (JTTHF). Considering the importance of endurance in daily life activities, we believe that there is a need for alternative activity measurements. However, in the Netherlands a new measurement tool, the Task-oriented Arm Hand Capacity (TAAC), has been developed which can be a valuable addition to current activity measurements. This tool includes functional tasks and is used for measuring proximal and distal muscle strength and muscle fatigability[54].

In summary, our results indicate that shoulder muscle strength and stereognosis are significant predictors for muscle fatigability explaining 33.60% of its variance. Unimanual capacity and bimanual performance are weakly related with muscle fatigability. Nevertheless, our results suggest that muscle fatigability should be considered as an important underlying impairment in children with UCP.

The study includes some limitations. The first limitation to discuss is the fact that the achieved results cannot be generalized to the total CP population. The population of this study predominantly had mild to moderate functional impairments (MACS I-III).

Secondly, the design was a cross-sectional study and therefore we cannot determine causality in relationships among fatigability and daily life activities. Prospective studies are recommended to evaluate how changes in muscle fatigability impact manual abilities. This study was a non-probability convenience sample and involved a selective including of participants. Children were only recruited at the University Hospital Pellenberg in Leuven implying a risk of selection bias. Future research with participants across different clinical settings are necessary to provide a more representative sample.

Thirdly, a critical reflection of the used measurement instruments must be considered. The reliability of the Static Fatigue Index in children with CP was only investigated in a pilot study[33]. Furthermore, it is difficult to evaluate muscle fatigability in an objective and optimal way because it is a complex phenomenon[25]. Both the protocol as well as the calculation of the SFI measurement should be critically considered. Factors such as child's motivation and rater's verbal encouragement can influence the performance. Another consideration is that the calculation does not include the amount of force variability. This leads to the fact that

different curve shapes can have the same fatigability value. As the literature showed that children with UCP experience increased force variability[17,18,19], this should be considered in future calculations of muscle fatigability.

Measurements of PROM, spasticity, MMT and sensory functions were based on nominal or ordinal rating scales which made the results less sensitive to subtle changes. However, Klingels et al. (2010) showed for these assessments moderate to very high test-retest and interrater reliability[32]. As suggested by the International Classification of Functioning, Disability and Health[55] (ICF) several other factors such as motivation, cognition, age and comorbidities can influence the performance of the measurements and should be considered.

In conclusion, our results showed that the AH had 11% more muscle fatigability and 60% less grip strength compared to the NAH. Also, there was a significant positive correlation between grip strength of the AH and age. Body functions impairments such as grip strength, shoulder muscle strength, total muscle strength but also sensibility (stereognosis) impact muscle fatigability. According to activity measures, a significant but low correlation between SFI and manual ability was found. These new insights are meaningful to gain a more complete impression of children's functional level. Further research with larger sample sizes and optimization of the measurement protocol of muscle fatigability are important to improve these insights.

## Reference list


1. Bax, M., Goldstein, M., Rosenbaum, P., Leviton, A., Paneth, N., Dan, B., . . . Damiano, D. (2005). Proposed definition and classification of cerebral palsy, April 2005. *Dev Med Child Neurol, 47*(8), 571-576.
2. Himmelmann, K., & Uvebrant, P. (2014). The panorama of cerebral palsy in Sweden. XI. Changing patterns in the birth-year period 2003-2006. *Acta Paediatr, 103*(6), 618-624. doi:10.1111/apa.12614
3. SCPE. (2000). Surveillance of cerebral palsy in Europe: a collaboration of cerebral palsy surveys and registers. Surveillance of Cerebral Palsy in Europe (SCPE). *Dev Med Child Neurol, 42*(12), 816-824.
4. Westbom, L., Hagglund, G., & Nordmark, E. (2007). Cerebral palsy in a total population of 4-11 year olds in southern Sweden. Prevalence and distribution according to different CP classification systems. *BMC Pediatr, 7*, 41. doi:10.1186/1471-2431-7-41
5. Klingels, K., Demeyere, I., Jaspers, E., De Cock, P., Molenaers, G., Boyd, R., & Feys, H. (2012). Upper limb impairments and their impact on activity measures in children with unilateral cerebral palsy. *Eur J Paediatr Neurol, 16*(5), 475-484. doi:10.1016/j.ejpn.2011.12.008
6. Van Heest, A. E., House, J., & Putnam, M. (1993). Sensibility deficiencies in the hands of children with spastic hemiplegia. *J Hand Surg Am, 18*(2), 278-281. doi:10.1016/0363-5023(93)90361-6
7. Krumlinde-Sundholm, L., & Eliasson, A. C. (2002). Comparing tests of tactile sensibility: aspects relevant to testing children with spastic hemiplegia. *Dev Med Child Neurol, 44*(9), 604-612.
8. Moreau, N. G., & Gannotti, M. E. (2015). Addressing muscle performance impairments in cerebral palsy: Implications for upper extremity resistance training. *J Hand Ther, 28*(2), 91-99; quiz 100. doi:10.1016/j.jht.2014.08.003
9. Jahnsen, R., Villien, L., Stanghelle, J. K., & Holm, I. (2003). Fatigue in adults with cerebral palsy in Norway compared with the general population. *Dev Med Child Neurol, 45*(5), 296-303.
10. Rose, J., & McGill, K. C. (2005). Neuromuscular activation and motor-unit firing characteristics in cerebral palsy. *Dev Med Child Neurol, 47*(5), 329-336.
11. Rosenbaum, P., Paneth, N., Leviton, A., Goldstein, M., Bax, M., Damiano, D., . . . Jacobsson, B. (2007). A report: the definition and classification of cerebral palsy April 2006. *Dev Med Child Neurol Suppl, 109*, 8-14
12. Moreau, N. (2007). *Quantification of muscle fatigue in cerebral palsy and its relationship to impairments and function* (Doctoral dissertation). Louisiana State University Medical Center, Department of kinesiology
13. Bigland-Ritchie, B., Johansson, R., Lippold, O. C., & Woods, J. J. (1983). Contractile speed and EMG changes during fatigue of sustained maximal voluntary contractions. *J Neurophysiol, 50*(1), 313-324.
14. Tilton, A. H. (2006). Therapeutic interventions for tone abnormalities in cerebral palsy. *NeuroRx, 3*(2), 217-224. doi:10.1016/j.nurx.2006.01.008
15. Damiano, D. L., Quinlivan, J., Owen, B. F., Shaffrey, M., & Abel, M. F. (2001). Spasticity versus strength in cerebral palsy: relationships among involuntary resistance, voluntary torque, and motor function. *Eur J Neurol, 8 Suppl 5*, 40-49.
16. Ito, J., Araki, A., Tanaka, H., Tasaki, T., Cho, K., & Yamazaki, R. (1996). Muscle histopathology in spastic cerebral palsy. *Brain Dev, 18*(4), 299-303.

17. Castle, M. E., Reyman, T. A., & Schneider, M. (1979). Pathology of spastic muscle in cerebral palsy. *Clin Orthop Relat Res*(142), 223-232.

18. Smits-Engelsman, B. C., Rameckers, E. A., & Duysens, J. (2005). Muscle force generation and force control of finger movements in children with spastic hemiplegia during isometric tasks. *Dev Med Child Neurol, 47*(5), 337-342.

19. Valvano, J., & Newell, K. M. (1998). Practice of a precision isometric grip-force task by children with spastic cerebral palsy. *Dev Med Child Neurol, 40*(7), 464-473.

20. Eken, M. M., Dallmeijer, A. J., Doorenbosch, C. A., Dekkers, H., Becher, J. G., & Houdijk, H. (2014). Assessment of muscle endurance of the knee extensor muscles in adolescents with spastic cerebral palsy using a submaximal repetitions-to-fatigue protocol. *Arch Phys Med Rehabil, 95*(10), 1888-1894. doi:10.1016/j.apmr.2014.05.010

21. Eken, M. M., Dallmeijer, A. J., Houdijk, H., & Doorenbosch, C. A. (2013). Muscle fatigue during repetitive voluntary contractions: a comparison between children with cerebral palsy, typically developing children and young healthy adults. *Gait Posture, 38*(4), 962-967. doi:10.1016/j.gaitpost.2013.05.004

22. Moreau, N. G., Li, L., Geaghan, J. P., & Damiano, D. L. (2008). Fatigue resistance during a voluntary performance task is associated with lower levels of mobility in cerebral palsy. *Arch Phys Med Rehabil, 89*(10), 2011-2016. doi:10.1016/j.apmr.2008.03.012

23. Brauers, L., Geijen, M. M., Speth, L. A., & Rameckers, E. A. (2017). Does intensive upper limb treatment modality Hybrid Constrained Induced Movement Therapy (H-CIMT) improve grip and pinch strength or fatigability of the affected hand? *J Pediatr Rehabil Med, 10*(1), 11-17. doi:10.3233/prm-170406

24. Doix, A. C., Gulliksen, A., Braendvik, S. M., & Roeleveld, K. (2013). Fatigue and muscle activation during submaximal elbow flexion in children with cerebral palsy. *J Electromyogr Kinesiol, 23*(3), 721-726. doi:10.1016/j.jelekin.2012.12.005

25. van Meeteren, J., van Rijn, R. M., Selles, R. W., Roebroeck, M. E., & Stam, H. J. (2007). Grip strength parameters and functional activities in young adults with unilateral cerebral palsy compared with healthy subjects. *J Rehabil Med, 39*(8), 598-604. doi:10.2340/16501977-0095

26. Rose, J., & McGill, K. C. (2005). Neuromuscular activation and motor-unit firing characteristics in cerebral palsy. *Dev Med Child Neurol, 47*(5), 329-336.

27. Schwid, S. R., Thornton, C. A., Pandya, S., Manzur, K. L., Sanjak, M., Petrie, M. D., . . . Goodman, A. D. (1999). Quantitative assessment of motor fatigue and strength in MS. *Neurology, 53*(4), 743-750.

28. Benedict, R. H., Wahlig, E., Bakshi, R., Fishman, I., Munschauer, F., Zivadinov, R., & Weinstock-Guttman, B. (2005). Predicting quality of life in multiple sclerosis: accounting for physical disability, fatigue, cognition, mood disorder, personality, and behavior change. *J Neurol Sci, 231*(1-2), 29-34. doi:10.1016/j.jns.2004.12.009

29. Severijns, D., Lamers, I., Kerkhofs, L., & Feys, P. (2015). Hand grip fatigability in persons with multiple sclerosis according to hand dominance and disease progression. *J Rehabil Med, 47*(2), 154-160. doi:10.2340/16501977-1897

30. Surakka, J., Romberg, A., Ruutiainen, J., Virtanen, A., Aunola, S., & Maentaka, K. (2004). Assessment of muscle strength and motor fatigue with a knee dynamometer in subjects with multiple sclerosis: a new fatigue index. *Clin Rehabil, 18*(6), 652-659. doi:10.1191/0269215504cr781oa

31. Compagnone, E., Maniglio, J., Camposeo, S., Vespino, T., Losito, L., De Rinaldis, M., Trabacca, A. (2014). Functional classifications for cerebral palsy: correlations between the gross motor

function classification system (GMFCS), the manual ability classification system (MACS) and the communication function classification system (CFCS). *Res Dev Disabil, 35*(11), 2651-2657. doi:10.1016/j.ridd.2014.07.005

32. Klingels, K., De Cock, P., Molenaers, G., Desloovere, K., Huenaerts, C., Jaspers, E., & Feys, H. (2010). Upper limb motor and sensory impairments in children with hemiplegic cerebral palsy. Can they be measured reliably? *Disability and Rehabilitation, 32*(5), 409-416. doi:10.3109/09638280903171469

33. Brauers L. (2015). *Test-retest reliability of a new fatigue index to quantitatively evaluate fatigue in children with cerebral palsy* (Unpublished master's thesis). Adelante rehabilitation Center, Valkenburg, The Netherlands

34. Bohannon, R. W., & Smith, M. B. (1987). Interrater reliability of a modified Ashworth scale of muscle spasticity. *Phys Ther, 67*(2), 206-207.

35. Hislop H.J., & Montgomery J. (1995). Daniels and Worthingham's Muscle Testing: techniques of manual examination. Philadelphia: Harcourt Brace & Company

36. Taylor, N., Sand, P. L., & Jebsen, R. H. (1973). Evaluation of hand function in children. *Arch Phys Med Rehabil, 54*(3), 129-135.

37. Charles, J. R., Wolf, S. L., Schneider, J. A., & Gordon, A. M. (2006). Efficacy of a child-friendly form of constraint-induced movement therapy in hemiplegic cerebral palsy: a randomized control trial. *Developmental Medicine and Child Neurology, 48*(8), 635-642. doi:10.1017/s0012162206001356

38. Randall, M., Carlin, J. B., Chondros, P., & Reddihough, D. (2001). Reliability of the Melbourne assessment of unilateral upper limb function. *Dev Med Child Neurol, 43*(11), 761-767.

39. Krumlinde-Sundholm L., Eliasson A (2003). Development of the Assisting Hand Assessment, a Rasch built measure intended for children with unilateral upper limb impairments. Scand J Occup Ther, 10, 16-26

40. Holmefur, M. M., & Krumlinde-Sundholm, L. (2016). Psychometric properties of a revised version of the Assisting Hand Assessment (Kids-AHA 5.0). *Dev Med Child Neurol, 58*(6), 618-624. doi:10.1111/dmcn.12939

41. Krumlinde-Sundholm, L., Holmefur, M., Kottorp, A., & Eliasson, A. C. (2007). The Assisting Hand Assessment: current evidence of validity, reliability, and responsiveness to change. *Dev Med Child Neurol, 49*(4), 259-264. doi:10.1111/j.1469-8749.2007.00259.x

42. Arnould, C., Penta, M., Renders, A., & Thonnard, J. L. (2004). ABILHAND-Kids: a measure of manual ability in children with cerebral palsy. *Neurology, 63*(6), 1045-1052.

43. Vandervelde, L., Van den Bergh, P. Y., Penta, M., & Thonnard, J. L. (2010). Validation of the ABILHAND questionnaire to measure manual ability in children and adults with neuromuscular disorders. *J Neurol Neurosurg Psychiatry, 81*(5), 506-512. doi:10.1136/jnnp.2009.177055

44. Hinkle, D.E., Wiersma W., & Jurs S.G. (1998). *Applied statistics for behavioral science*. 4th ed. Boston: Houghton Mifflin Company

45. Stackhouse, S. K., Binder-Macleod, S. A., & Lee, S. C. (2005). Voluntary muscle activation, contractile properties, and fatigability in children with and without cerebral palsy. *Muscle Nerve, 31*(5), 594-601. doi:10.1002/mus.20302

46. Marbini, A., Ferrari, A., Cioni, G., Bellanova, M. F., Fusco, C., & Gemignani, F. (2002). Immunohistochemical study of muscle biopsy in children with cerebral palsy. *Brain Dev, 24*(2), 63-66.

47. Klingels, K., Feys, H., De Wit, L., Jaspers, E., Van de Winckel, A., Verbeke, G., Molenaers, G. (2012). Arm and hand function in children with unilateral cerebral palsy: a one-year follow-up study. *Eur J Paediatr Neurol, 16*(3), 257-265. doi:10.1016/j.ejpn.2011.08.001

48. Blank, R., & Kluger, G. (2009). Changes in elementary finger-hand functions over time in preschool children with spastic cerebral palsy. *Neurosci Lett, 455*(1), 30-35. doi:10.1016/j.neulet.2009.03.058

49. Ploegmakers, J. J., Hepping, A. M., Geertzen, J. H., Bulstra, S. K., & Stevens, M. (2013). Grip strength is strongly associated with height, weight and gender in childhood: a cross sectional study of 2241 children and adolescents providing reference values. *J Physiother, 59*(4), 255-261. doi:10.1016/s1836-9553(13)70202-9

50. Braendvik, S. M., Elvrum, A. K., Vereijken, B., & Roeleveld, K. (2010). Relationship between neuromuscular body functions and upper extremity activity in children with cerebral palsy. *Dev Med Child Neurol, 52*(2), e29-34. doi:10.1111/j.1469-8749.2009.03490.x

51. Meldrum, D., Cahalane, E., Conroy, R., Guthrie, R., & Hardiman, O. (2007). Quantitative assessment of motor fatigue: normative values and comparison with prior-polio patients. *Amyotroph Lateral Scler, 8*(3), 170-176. doi:10.1080/17482960701223113

52. Arnould, C., Penta, M., & Thonnard, J. L. (2007). Hand impairments and their relationship with manual ability in children with cerebral palsy. *J Rehabil Med, 39*(9), 708-714. doi:10.2340/16151977-0111

53. Severijns, D., Van Geel, F., & Feys, P. (2018). Motor fatigability in persons with multiple sclerosis: Relation between different upper limb muscles, and with fatigue and the perceived use of the arm in daily life. *Mult Scler Relat Disord, 19*, 90-95. doi:10.1016/j.msard.2017.11.016

54. Geijen, M. (2016). *Reliability and validity of the TAAC instrument during bimanual activities in children with unilateral Cerebral Palsy* (Unpublished master's thesis). Maastricht University, The Netherlands

55. ICF (2001*). International classification of functioning, disability and health*. Geneva, Switzerland: World Health Organisation, http://www.who.int/classifications/icf/en/

## Appendix 1: descriptive statistics

**Table 1:**
*Descriptive statistics of the outcome measures.*

| | | |
|---|---|---|
| **Body Function** | | |
| PROM | N (0/1) | |
| - Shoulder | | |
| o Flexion | | 41/0 |
| o Abduction | | 41/0 |
| o External rotation | | 41/0 |
| o Internal rotation | | 40/1 |
| - Elbow | | |
| o Extension | | 38/3 |
| o Supination | | 27/14 |
| - Wrist + hand | | |
| o Wrist extension | | 40/1 |
| o Finger extension PIP | | 41/0 |
| o Finger extension DIP | | 41/0 |
| o Thumb abduction | | 26/15 |
| Muscle tone (0-48) | Me (IQR) | 7.5 (5.5-9) |
| - Shoulder (0-20) | | 3 (1-4) |
| - Elbow (0-12) | | 2.5 (2-3.5) |
| - Wrist + hand (0-16) | | 2 (2-3) |
| Muscle strength (0-45) | Me (IQR) | 32 (31-34) |
| - Shoulder (0-15) | | 11.5 (11-12) |
| - Elbow (0-20) | | 14.5 (14-15.5) |
| - Wrist (0-10) | | 6.5 (6-7) |
| Grip strength | Mean (SD) | 4.91 (3.09) |
| Sensory function | N (0/1/2) | |
| - Exteroception | | 1/4/33 |
| - Proprioception | | 0/10/28 |
| - Two-point discrimination | | 16/5/17 |
| - Stereognosis | | 16/15/7 |
| **Activity Level** | | |
| JTTHF | Mean (SD) | 237.68 (161.40) |
| Abilhand-kids | | 1.75 (1.81) |
| Melbourne | | 75.53 (13.89) |
| AHA | | 57.18 (11.21) |

PROM, passive range of motion; N, total number of children; Me, Median; IQR, Inter Quartile Range; SD, standard deviation; JTTHF, Jebsen Taylor Test of Hand Function; AHA, assisting hand assessment

**Appendix 2: progress form**

www.uhasselt.be
Campus Hasselt | Martelarenlaan 42 | BE-3500 Hasselt
Campus Diepenbeek | Agoralaan gebouw D | BE-3590 Diepenbeek
T + 32(0)11 26 81 11 | E-mail: info@uhasselt.be

UHASSELT
KNOWLEDGE IN ACTION

## VOORTGANGSFORMULIER WETENSCHAPPELIJKE STAGE DEEL 2

| DATUM | INHOUD OVERLEG | HANDTEKENINGEN |
|---|---|---|
| 10/10/17 | Bespreking verder verloop MP deel 2. Afspraken maken rond sorteren van de data. | Promotor:<br>Copromotor:<br>Student(e):<br>Student(e): |
| 25/01/18 | Bespreking inleiding + methode. Data-set op punt stellen. | Promotor:<br>Copromotor:<br>Student(e):<br>Student(e): |
| 21/02/18 | Bespreking resultaten | Promotor:<br>Copromotor:<br>Student(e):<br>Student(e): |
| 05/04/18 | Bespreking resultaten + discussie | Promotor:<br>Copromotor:<br>Student(e):<br>Student(e): |
| 25/05/18 | Bespreking discussie + presentatie | Promotor:<br>Copromotor:<br>Student(e):<br>Student(e): |
| | | Promotor:<br>Copromotor:<br>Student(e):<br>Student(e): |
| | | Promotor:<br>Copromotor:<br>Student(e):<br>Student(e): |
| | | Promotor:<br>Copromotor:<br>Student(e):<br>Student(e): |
| | | Promotor:<br>Copromotor:<br>Student(e):<br>Student(e): |
| | | Promotor:<br>Copromotor:<br>Student(e):<br>Student(e): |

# Auteursrechtelijke overeenkomst

Ik/wij verlenen het wereldwijde auteursrecht voor de ingediende eindverhandeling:
**Does muscle fatigability impact on upper limb function and activity measures in children with unilateral cerebral palsy?**

Richting: **master in de revalidatiewetenschappen en de kinesitherapie-revalidatiewetenschappen en kinesitherapie bij musculoskeletale aandoeningen**
Jaar: **2018**

in alle mogelijke mediaformaten, - bestaande en in de toekomst te ontwikkelen - , aan de Universiteit Hasselt.

Niet tegenstaand deze toekenning van het auteursrecht aan de Universiteit Hasselt behoud ik als auteur het recht om de eindverhandeling, - in zijn geheel of gedeeltelijk -, vrij te reproduceren, (her)publiceren of distribueren zonder de toelating te moeten verkrijgen van de Universiteit Hasselt.

Ik bevestig dat de eindverhandeling mijn origineel werk is, en dat ik het recht heb om de rechten te verlenen die in deze overeenkomst worden beschreven. Ik verklaar tevens dat de eindverhandeling, naar mijn weten, het auteursrecht van anderen niet overtreedt.

Ik verklaar tevens dat ik voor het materiaal in de eindverhandeling dat beschermd wordt door het auteursrecht, de nodige toelatingen heb verkregen zodat ik deze ook aan de Universiteit Hasselt kan overdragen en dat dit duidelijk in de tekst en inhoud van de eindverhandeling werd genotificeerd.

Universiteit Hasselt zal mij als auteur(s) van de eindverhandeling identificeren en zal geen wijzigingen aanbrengen aan de eindverhandeling, uitgezonderd deze toegelaten door deze overeenkomst.

Voor akkoord,

**Geerits, Sandra** **Pareyn, Stephanie**