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## Measuring Fatigue In Adolescents And Young Adults With Cerebral Palsy

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A thesis submitted in partial fulfillment of the requirements for the degree in Doctor of Philosophy  
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MEASURING FATIGUE IN ADOLESCENTS AND YOUNG ADULTS  
WITH CEREBRAL PALSY

(Thesis format: Integrated-Article)

by

Laura K. Brunton

Graduate Program in Health and Rehabilitation Sciences

A thesis submitted in partial fulfillment  
of the requirements for the degree of  
Doctor of Philosophy

The School of Graduate and Postdoctoral Studies  
The University of Western Ontario  
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## Abstract

Fatigue is a significant issue and has been estimated to affect between 30-50% of individuals with cerebral palsy (CP) in various studies; however, there is no validated measure of fatigue for this population. A systematic review revealed no one single measure with adequate psychometric properties for use with individuals with CP. As a result a new tool was created: the Fatigue Impact and Severity Self-Assessment (FISSA). A phenomenology was conducted with youth and young adults with CP to understand the bodily experience of living with CP and as a client-centered approach to item creation for the FISSA. Focus groups with healthcare professionals were used to reduce the number of items on the FISSA and to ensure relevance to the population. A large survey was conducted to assess the validity, reliability and factor structure of the FISSA. The bodily experience of CP revolved around, and emphasized, fatigue that occurs with walking and prolonged activity. Self-awareness of the individuals' own bodies emerged as the most important theme and strategies employed to prevent and manage fatigue were elucidated. In the validation study, individuals who self-classified as level I on the Gross Motor Function Classification System (GMFCS) were shown to experienced less fatigue than individuals in any other GMFCS level (II-V) ( $p < .001$ ). Individuals with higher pain (both impact and severity) also reported higher fatigue scores ( $p < .001$ ). The FISSA was shown to be reliable ( $\alpha = 0.95$ ;  $ICC(3,1)=0.74$  (95% CI 0.53-0.87)) and contains 31 items related to two factors (Impact of Fatigue and Management/Activity Modification) that together explain 48.7% of the variance in fatigue scores. The FISSA was created to examine the severity, impact and management of fatigue for youth and young adults with CP. The FISSA is reliable and was able to discriminate between groups expected to experience more fatigue including those with a more severe motor disability according to the GMFCS and individuals with a higher degree of pain. The FISSA allows for individualized identification of the activities of daily living that may be compromised by fatigue, which may enhance collaborative goal setting and intervention planning by clinicians and their clients.

**Key words:** Fatigue, Measurement, Cerebral Palsy, Adolescents, Young Adults, Pain, Exercise, Physical Activity.

### **Co-Authorship Statement**

Chapter 2 - A Critical Review of Reports of Fatigue in Cerebral Palsy was co-authored with Dr. Charles Rice (School of Kinesiology, Canadian Centre for Activity and Aging, Western University, London, Ontario, Canada) and has been published in *Developmental Neurorehabilitation*, Volume 15 Issue 1 pages 54–62. Dr. Rice contributed to this chapter through review of the interpretations and editing of the writing.

Chapter 3 - A Review of Fatigue Measures Used in Neurological Conditions: Preparation for the Development of a Measure Specific to Cerebral Palsy was co-authored with Katharine Randall (Graduate Program in Health and Rehabilitation Sciences, Western University, London, Ontario, Canada) and Dr. Doreen Bartlett (School of Physical Therapy, Western University, London, Ontario, Canada). Katharine Randall contributed to this chapter through review and selection of the retrieved abstracts and editing of the writing. Dr. Bartlett contributed to this chapter through review of the interpretations of the findings and review and editing of the writing.

Chapter 4 - Understanding the Bodily Experience of Cerebral Palsy: A Phenomenology was co-authored with Dr. Doreen Bartlett (School of Physical Therapy, Western University, London, Ontario, Canada) and has been published in *Disability and Rehabilitation*, Volume 35 Issue 23 pages 1981-1990. Dr. Bartlett contributed to this chapter through secondary thematic analysis of the interview transcripts and review and editing of the writing.

Chapter 5 – Constructing a New Clinical Measure of Fatigue for Adolescents and Young Adults with Cerebral Palsy and Chapter 6 – Determining the Psychometric Properties of a New Clinical Measure of Fatigue for Adolescents and Young Adults with Cerebral Palsy: The Fatigue Impact and Severity Self-Assessment were written solely by this author. A manuscript combining these two chapters will be co-authored by Dr. Doreen Bartlett (School of Physical Therapy, Western University, London, Ontario, Canada) and Marilyn Wright (Department of Pediatrics, McMaster University, Hamilton, Ontario, Canada).



**Dedication:**

To my loving family and friends who have supported me throughout this endeavor, I am forever grateful.

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## Table of Contents

<b>Prefatory Pages.....</b>	<b>ii</b>
Abstract.....	ii
Co-Authorship Statement.....	iii
Dedication.....	iv
Acknowledgments.....	v
<b>Chapter 1: Introduction.....</b>	<b>1</b>
Cerebral Palsy.....	1
Fatigue, Pain and Physical Activity.....	5
Measurement Development.....	10
Focus of this Dissertation.....	12
Summary.....	14
<b>Chapter 2: A Critical Review of Reports of Fatigue in Cerebral Palsy.....</b>	<b>21</b>
Introduction.....	21
Reports of fatigue in cerebral palsy: The conflict.....	22
Central sites.....	25
Peripheral sites – Anatomical features.....	30
Peripheral sites – Physiological features.....	33
Summary and future directions.....	39
<b>Chapter 3: A Review of Fatigue Measures Used in Neurological Conditions: Preparation for the Development of a Measure Specific to Cerebral Palsy.....</b>	<b>45</b>
Introduction.....	45
Methods.....	46
Results.....	50
Discussion.....	61
Study Limitations.....	65
Conclusion.....	65
<b>Chapter 4: Understanding the Bodily Experience of Cerebral Palsy: A Phenomenology.....</b>	<b>70</b>
Introduction.....	70
Methodology.....	72
Methods.....	72
Results.....	76
Discussion.....	114
<b>Chapter 5: Constructing a New Clinical Measure of Fatigue for Adolescents and Young Adults with Cerebral Palsy.....</b>	<b>119</b>
Introduction.....	119
Methods.....	119
Results.....	123
Discussion.....	127

<b>Chapter 6: Determining the Psychometric Properties of a New Clinical Measure of Fatigue for Adolescents and Young Adults with Cerebral Palsy: The Fatigue Impact and Severity Self-Assessment.....</b>	<b>131</b>
Introduction .....	131
Methods.....	133
Analysis .....	136
Results .....	138
Discussion .....	144
Limitations.....	145
<b>Chapter 7: Summary, Clinical Implications and Future Research Directions</b>	<b>150</b>
Summary and Clinical Implications .....	150
Future Directions .....	155

## List of Tables

<i>Table 1-1 - Item Requirements by Test Purpose.....</i>	<i>11</i>
<i>Table 1-2 - Reliability and Validity Requirements for Discriminative Purposes .....</i>	<i>12</i>
<i>Table 2-1 - Motor Unit Activation in Individuals with Cerebral Palsy.....</i>	<i>29</i>
<i>Table 2-2 - Muscle Parameters in Individuals with Cerebral Palsy.....</i>	<i>32</i>
<i>Table 2-3 - Energy Expenditure During Walking in Individuals with Cerebral Palsy.....</i>	<i>35</i>
<i>Table 3-1- Definitions of Measurement Properties Assessed.....</i>	<i>49</i>
<i>Table 3-2 - Characteristics of the Identified Scales.....</i>	<i>51</i>
<i>Table 3-3 - Categorization and Rationale for Each Fatigue Measure.....</i>	<i>60</i>
<i>Table 4-1 - Individual Participant Characteristics.....</i>	<i>73</i>
<i>Table 4-2 - Characteristics of the Sample .....</i>	<i>76</i>
<i>Table 5-1 – Items Removed from First Draft of the Fatigue Impact and Severity Self-Assessment by Focus Group 1 .....</i>	<i>124</i>
<i>Table 5-2 - New Items Added to the Fatigue Impact and Severity Self-Assessment by Focus Group 1....</i>	<i>125</i>
<i>Table 5-3 - Items Removed from Second Draft of the Fatigue Impact and Severity Self-Assessment by Focus Group 2.....</i>	<i>125</i>
<i>Table 5-4 - New Items Added to the Fatigue Impact and Severity Self-Assessment by Focus Group 2....</i>	<i>125</i>
<i>Table 6-1 Participant Characteristics and Demographic Information of the Sample .....</i>	<i>139</i>
<i>Table 6-2 Summary of Exploratory Factor Analysis results for the Fatigue Severity and Impact Self Assessment.....</i>	<i>141</i>
<i>Table 6-3 Summary of Fatigue Scores for Known Groups Validity Testing.....</i>	<i>143</i>

## List of Figures

<i>Figure 2-1 - Factors Contributing to Conflicting Reports of Fatigability in Individuals with Cerebral Palsy.....</i>	<i>40</i>
<i>Figure 3-1 - Flowchart of Systematic Review Process.....</i>	<i>48</i>
<i>Figure 4-1 - Major Identified Themes.....</i>	<i>77</i>
<i>Figure 6-1 - Breakdown of Inclusion and Exclusion of Returned Surveys.....</i>	<i>138</i>

## List of Appendices

<i>Appendix 2-A – Copyright Permission From Developmental Neurorehabilitation to Reproduce “Fatigue in Cerebral Palsy: A Critical Review”</i>	160
<i>Appendix 3-A – Psychometric Properties of Identified Scales</i>	161
<i>Appendix 4-A Ethics Approval from Western University for Phenomenology</i>	174
<i>Amendment to Extend Study Deadline</i>	175
<i>Amendment to Change Assent Form</i>	176
<i>Appendix 4-B Ethics Approval from Thames Valley Children’s Centre for Phenomenology</i>	177
<i>Appendix 4-C Letter of Information and Consent Forms for Phenomenological Inquiry</i>	178
<i>Appendix 4-D Semi-Structured Interview Guide</i>	187
<i>Appendix 5-A Ethics Approval from Western University for Focus Groups and Feasibility Testing</i>	188
<i>Amendment to Ethics for Changes as a result of Thames Valley Children’s Centre Review Board Submission</i>	189
<i>Appendix 5-B Focus Groups and Feasibility Testing Approval from McMaster University</i>	190
<i>Appendix 5-C Ethics Approval from Thames Valley Children’s Centre for Focus Groups and Feasibility Testing</i>	191
<i>Appendix 5-D Feasibility Testing and Focus Groups Letter of Information and Consent Forms</i>	192
<i>Appendix 5-E Feasibility Questionnaire</i>	201
<i>Appendix 5-F Items for Consideration for the Fatigue Impact and Severity Self-Assessment</i>	202
<i>Appendix 5-G Initial Draft of the Fatigue Impact and Severity Self-Assessment for Focus Group 1</i>	205
<i>Appendix 5-H Second Draft of the Fatigue Impact and Severity Self-Assessment for Focus Group 2</i>	207
<i>Appendix 5-I Draft of the Fatigue Impact and Severity Self-Assessment for Feasibility Testing</i>	209
<i>Appendix 6-A Western University Ethical Approval</i>	213
<i>Appendix 6-B Letter of Information and Consent Forms for Validation Study</i>	217
<i>Appendix 6-C Recruitment Advertisement Flyer</i>	223
<i>Appendix 6-D Self-Report Measure of the Gross Motor Function Classification System Expanded and Revised Version</i>	224
<i>Appendix 6-E Demographic Questionnaire</i>	225
<i>Appendix 6-F Pain Questionnaire</i>	226
<i>Appendix 6-G Exercise Questionnaire</i>	228
<i>Appendix 6-H Correlation Matrix for All Fatigue Impact and Severity Self-Assessment Items</i>	230
<i>Appendix 6-I Scree Plot of Fatigue Impact and Severity Self-Assessment Factor Extraction Analysis</i>	231
<i>Appendix 6-J Final Fatigue Impact and Severity Self-Assessment Following Factor Analysis</i>	232
<i>Appendix 6-K Boxplots of Descriptive Results for Fatigue Score by Gross Motor Function Classification System Level, Pain and Physical Activity</i>	236
<i>Vita</i>	241

## **List of Abbreviation**

ASQME = Adolescent Study of Quality of Life, Mobility and Exercise  
CFCS = Communication Function Classification System  
CFS = Chalder Fatigue Scale  
ChFS = Chronic Fatigue Syndrome  
CP = Cerebral Palsy  
EFS = Empirical Fatigue Scale  
EMG = Electromyography  
FAI = Fatigue Assessment Inventory  
FIS = Fatigue Impact Scale  
FISSA = Fatigue Impact and Severity Self-Assessment  
FSMC = Fatigue Scale for Motor and Cognitive Functions  
FSS = Fatigue Severity Scale  
GMFCS = Gross Motor Function Classification System  
GMFCS-ER = Gross Motor Function Classification System Expanded and Revised Version  
ICC = Intraclass Correlation Coefficient  
MFI-20 = Multidimensional Fatigue Inventory  
MFIS = Modified Fatigue Impact Scale  
MFSS = Multiple Sclerosis Specific Fatigue Severity Scale  
MRI = Magnetic Resonance Imaging  
MS = Multiple Sclerosis  
MVC = Maximal Voluntary Contraction  
NFI = Neurological Fatigue Index  
NGT = Nominal Group Technique  
NMA = Neuromuscular Activation  
OFCP = Ontario Federation of Cerebral Palsy  
PD = Parkinson's Disease  
PFS = Parkinson Fatigue Scale  
r = Pearson's or Spearman's Correlation Coefficient  
SCPE = Surveillance of Cerebral Palsy in Europe  
SFQ = Short Fatigue Questionnaire  
SOFI = Swedish Occupational Fatigue Inventory  
TA = Tibialis Anterior  
UFIS = Unidimensional Fatigue Impact Scale  
VAS = Visual Analog Scale  
VO<sub>2</sub> = Volume of Oxygen per unit time  
 $\alpha$  = Cronbach's Alpha



## Chapter 1: Introduction

The objective of this dissertation is to gain a better understanding of the fatigue-pain-exercise complex associated with cerebral palsy (CP) described by many clinicians, researchers and individuals living with CP. This dissertation comprises a series of four studies that resulted in the development and validation of a measurement tool to assess the fatigue experienced by individuals with CP. Understanding how much fatigue individuals experience on a regular basis and the impact of this fatigue on their daily lives may help inform self-management and/or physical therapy interventions offered to these individuals to improve quality of life.

In this introduction I define cerebral palsy, elaborate on issues related to fatigue, pain and physical activity, identify the gaps in the present knowledge base (particularly around measuring fatigue) and propose a solution to the challenge associated with measuring fatigue.

### *Cerebral Palsy*

“Cerebral palsy describes a group of permanent disorders of the development of movement and posture, causing activity limitations, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of CP are often accompanied by disturbances of sensation, perception, cognition, communication, behaviour, by epilepsy and by *secondary musculoskeletal problems*” (Rosenbaum et al., 2007 p.9, italics by author). CP occurs in about 2 to 2.5 per 1,000 live births and represents a complex of symptoms more so than a specific disease path as a result of the highly individual nature of the lesion as well as secondary and tertiary conditions that can result from the primary disturbance (Stanley, Blair & Alberman, 2000). The mechanisms of injury are not fully understood; however, many factors that can contribute have been identified, including: extreme prematurity, infection and hypoxic-ischemic encephalopathy, ultimately resulting in damage to the immature brain (Johnston & Hagberg, 2007).

Although the injury to the brain is static, changes in functional status occur over time as the manifestations of the lesion are ever-changing (Sanger et al., 2003). Change in the appearance of CP over time can be caused by the development of the central nervous system, evolution of motor patterns at both the reflex and voluntary levels, by

motor learning or by therapy (Rosenbaum, Russell, Cadman, Gowland, Jarvis & Hardy, 1990) and by the natural history in terms of growth and development of secondary impairments to the musculoskeletal system. The foci of this work are these secondary impairments, specifically, fatigue and pain and how youth and young adults experience these impairments. As previously outlined in the definition of CP, the primary disability is a motor impairment. The movement disorder can include any or combinations of the following impairments: delay in movement onset, poor timing of force generation, poor force production, inability to maintain antigravity postural control, decreased speed of movement and increased co-contraction (Campbell, 1991). Therapists and caregivers have described a great deal of variability in the motor abilities of children and adolescents with CP. For example, children who walk securely may fall frequently or be unable to rise from the floor without assistance. This variability can be attributed to many reasons such as stress, illness and anxiety; it can also be ascribed to fatigue that occurs during activities of daily living including long distance ambulation or during periods of prolonged standing (Bjornson, Graubert, McLaughlin, Kerfeld & Clark, 1998).

The Gross Motor Function Classification System (GMFCS) was created to allow for communication within and across professions and for the purposes of intervention planning. The classification system is ordinal in nature and consists of five levels representing the functional spectrum observed (Palisano, Rosenbaum, Walter, Russell, Wood & Galuppi, 1997). The distinctions among categories of GMFCS level are based on clinically meaningful functional abilities and limitations experienced by individuals with CP. The five GMFCS levels define clinically meaningful subpopulations that are widely used by therapists in goal setting and planning of interventions (Hanna, Bartlett, Rivard & Russell, 2008). An individual classified as level I is able to perform all activities one would see in a person developing typically, although there may be difficulty in one or more of speed, balance and coordination of these activities. An individual classified as level V has difficulty controlling head and trunk postures and has very little voluntary control over his/her movements (Palisano et al., 1997). Although the GMFCS is age-dependent and was created for use in children, it has recently been expanded to include an adolescent age band from 12 to 18 years (Palisano, Rosenbaum, Bartlett & Livingston, 2008). The GMFCS has been shown to be a reliable and valid tool

for classifying children and adolescents with CP (Palisano et al., 1997) and further studies have shown the GMFCS level to remain stable over time (Palisano, Cameron, Rosenbaum, Walter & Russell, 2006; Wood & Rosenbaum, 2000).

Muscle tone is the term used to represent the force with which a muscle resists being stretched, or its stiffness. Muscle tone is conceptualized as a spectrum ranging from low to high (Bartlett & Palisano, 2000). Spasticity is a common term used in the CP literature; it is different from muscle tone in that it only represents the high end of the muscle tone spectrum (Sanger et al., 2003) and is defined as a velocity-dependent increase in the monosynaptic reflex within the muscle (Lance, 1980). Spasticity is the most common type of motor disorder associated with CP and is an important aspect to consider when describing experiences of activities of daily living in people with CP. Spastic CP is characterized by abnormal patterns of movement and/or posture, increased muscle tone and pathological reflexes which can include increased reflexes, hyperreflexia and/or pyramidal signs like the Babinski response (Surveillance of Cerebral Palsy in Europe (SCPE), 2000). Another common motor disorder is ataxia. Ataxic CP is described as abnormal patterns of posture and/or movement and the loss of orderly muscular contraction, which can result in movements performed with abnormal forces, rhythms and accuracy (SCPE, 2000). Finally, dyskinesia is a motor disorder associated with CP that is defined by abnormal patterns of movement and/or posture and involuntary, uncontrolled, recurring and occasionally stereotyped movements (SCPE, 2000). Dyskinetic CP can be subdefined into dystonic CP and choreo-athetotic CP (SCPE, 2000). Dystonic CP is characterized by reduced activity (hypokinesia) and an increase in tone (hypertonia) (SCPE, 2000). Choreo-athetotic CP is described as increased activity (hyperkinesia) and a decrease in tone (hypotonia) (SCPE, 2000).

Another common classification method used in clinical practice and research in CP is related to the distribution of involvement. Hemiplegia is used to describe individuals who have problems with gross motor function that are restricted to one side of the body. Generally, hand function is more affected than leg function and the person has near-normal control on the unaffected (opposite) side of the body. Often these individuals are classified as “relatively mild” (Bax, Flodmark & Tydeman, 2007). Diplegia is the term used to describe gross motor problems that exist primarily in the lower limbs while

the person demonstrates reasonably good fine motor functions in the upper limbs (Bax et al., 2007). Triplegia involves three extremities, usually both lower limbs and one upper limb. Quadriplegia is the term used to describe severe motor impairments that include all four extremities. Individuals who experience this form of CP are usually – but not always – classified as a GMFCS level IV or V and often have limited hand function at best.

Although the primary impairment of CP is a motor impairment, during growth and maturation the development of secondary impairments associated with CP can occur. These secondary impairments can include disturbances to the auditory and visual systems and disorders of the digestive and respiratory systems as well as musculoskeletal impairments such as fatigue, pain, and the development of deformities, which can lead to range of motion impairments (Hilberink, Roebroek, Nieuwstraten, Jalink, Verheijden & Stam, 2007) and further pain. This dissertation focuses on the impact of the secondary impairments to the musculoskeletal system, specifically fatigue and pain. Although the investigation of the development of musculoskeletal deformities is beyond the scope of this work, the musculoskeletal deformities observed in individuals with CP that may lead to pain can include: subluxations and dislocations of the hip, abnormalities of the foot, patella alta, scoliosis, pelvic obliquity and contractures (Gajdosik & Cicirello, 2001). Subluxations and dislocations of the hip can often lead to refractory pain or arthrosis and can require surgical intervention (Gajdosik & Cicirello, 2001). Abnormalities of the foot and knee such as patella alta (superior displacement of the patella) can also lead to pain (Gajdosik & Cicirello, 2001). Additionally, individuals with CP are at risk for deterioration of the scoliotic curve as a result of scoliosis from neuromuscular origins (Gajdosik & Cicirello, 2001). Osteoarthritis can be another cause of pain experienced by individuals with CP. Atypical, excessive or imbalanced muscle actions across a joint can lead to degeneration of articular cartilage and lead to bony deformities at certain joints (Gajdosik & Cicirello, 2001). Furthermore, contracture development can occur throughout the lifespan, this shortening of muscles or joints may be a result of the increase muscle tone associated with spasticity and can limit range of motion available for use in activities of daily living (Gajdosik & Cicirello, 2001). All of these conditions can develop during childhood and may not become intrusive or painful until adolescence or young adulthood. Further, it is important to study fatigue and pain in youth and young

adults as studies have shown that energy expenditure (and therefore, potentially fatigue) and pain increase with age in adults and children with CP (Waters & Mulroy, 1999; Opheim, Jahnsen, Olsson & Stanghelle, 2009). Many secondary impairments to the musculoskeletal system result in further pain. At the 2009 American Academy for Cerebral Palsy and Developmental Medicine conference, multiple plenary sessions alluded to a complex of fatigue and pain related to insufficient or excessive exercise or physical activity participation. Accordingly, this dissertation comprises an exploration of the complex created by fatigue, pain and exercise.

### *Fatigue, Pain and Physical Activity*

As previously described, the progression of the activity limitations in CP can occur through growth and development of secondary musculoskeletal conditions such as severe fatigue and chronic pain which can lead to further decreases in function that can affect independence in adult life (Tosi, Maher, Moore, Goldstein & Aisen, 2009). Several authors have described a cycle of deconditioning that can occur in CP. In this cycle, physical function decreases followed by a further decrease in physical activity, which can lead to a cascade of further functional decline (Tosi et al., 2009).

A recent study identified fatigue, pain and joint deformities as the top three CP-related impairments in adulthood that can impair activities of daily life (Hilberink et al., 2007). A few studies have reported that chronic pain and fatigue are more prevalent in adults with CP compared to the general population (Jahnsen, Villien, Stanghelle & Holm, 2003). Approximately 20% of physicians reported a noticeable functional deterioration in the adults with CP they treat (Hilberink et al., 2007) and some self-reported causes of this deterioration include spasticity, fatigue, pain and lack of physical training (Houlihan, 2009; Jones, 2009). It has also been demonstrated that muscle volume can be reduced by as much as 50% in individuals with CP, as well as the possibility of having less muscle reserve available for completing motor tasks compared to peers without disabilities matched for age and weight (Tosi et al., 2009). In addition, there have been reports that fatigue may contribute to physiological burnout in adults with CP, primarily through prolonged stress on the motor system combined with reduced muscle strength (Mockford & Caulton, 2010). Therefore, it is possible that deficits in muscle function, combined with the natural history of CP and the development of secondary conditions during

growth and maturation, may lead to functional deterioration and early loss of mobility (Tosi et al., 2009; Mockford & Caulton, 2010). Jones (2009) wrote a personal reflection about aging with CP and spoke of the need to understand the prevention of the development of secondary conditions such as musculoskeletal pain and fatigue.

Fatigue can be defined in a number of ways encompassing both mental and physical fatigue. For the purposes of the studies contained in this dissertation I have adopted Aaronson and colleagues' (1999) global definition of fatigue: "The awareness of a decreased capacity for physical and/or mental activity due to an imbalance in the availability, utilization, and/or restoration of resources needed to perform activity" (Aaronson, Teel, Cassmeyer, Neuberger, Pallikkathayil, Pierce et al., 1999, p. 46). Further, I have chosen to subdivide the overall concept of fatigue such that physical fatigue is defined as muscle fatigue that is a reduction in the force-generating capacity of the neuromuscular system that occurs during sustained activity (Bigland-Ritchie, Johansson, Lippold & Woods, 1983) and mental fatigue is thus defined as the failure to initiate or sustain cognitive tasks.

Adults with CP have reported significantly more physical fatigue than the general population (30% compared to 22% in a Norwegian sample) (Jahnsen et al., 2003). van der Slot and colleagues (2012) reported a 20% prevalence of fatigue within their sample of adults with CP; however, 41% of those individuals were classified as severely fatigued. In addition, of the individuals who reported severe fatigue (n=23) 83% also reported chronic pain and 44% reported depressive symptoms (van der Slot, Nieuwenhuijsen, van den Berg-Emons, Bergen, Hilberink, Stam et al., 2012). Individuals that reported severe fatigue tended to report more chronic pain with an odds ratio (OR) of 2.26 (95% CI = 1.08-4.72) and/or depressive symptoms (OR=3.38, 95% CI = 1.38-8.30) (van der Slot et al., 2012). Several studies have reported prevalence of muscle fatigue and predictors associated with fatigue in CP; however, objective measures of muscle fatigue have not been extensively used in the CP population as there are technical issues with spasticity and contractures interfering with testing positions and data collection procedures and with the measurement of fatigue during functional tasks (Brunton & Rice, 2012).

Pain however, has been studied expansively in the adult CP population and some literature is available on pain experiences of adolescents with CP. A high incidence of

chronic pain has been reported among children and adolescents (Engel, Petrina, Dudgeon & McKearnan, 2005); however, population-based studies on pain in adolescents and children with CP are needed to clarify prevalence of pain in this population. One study has shown prevalence of pain to be 62.5% in female adolescents and 49.2% in male adolescents living with CP (Doralp & Bartlett, 2010). The literature is inconsistent regarding the relationship between pain severity and functional status in CP. Two studies have shown a positive relationship between increased severity of pain and more severe motor impairment (Houlihan, O'Donnell, Conaway & Stevenson, 2004; Jahnsen, Villien, Aamodt, Stanghelle & Holm, 2003). However, in the study by Doralp and Bartlett (2010) and the study by Sandstrom and colleagues (2004), GMFCS level was not associated with prevalence or severity of pain, suggesting that children, adolescents and adults with CP, regardless of GMFCS level, could benefit from pain alleviation interventions (Doralp & Bartlett, 2010; Sandstrom et al., 2004). Pain has been noted to interfere with sleep, mobility and physical activities of daily living. It has also been suggested that there was a greater impact of pain when the adolescents were up all day without rest, which could reflect fatigue that is exacerbating pain (Engel et al., 2005). A review completed by Vogtle (2009) has suggested that pain in CP is much more complex than the typical musculoskeletal issues that can generate pain. Muscle weakness, fatigue and deterioration of functional status were highlighted as being contributors to the pain experienced by adults with CP (Vogtle, 2009); however, the relationships between these concepts have not yet been fully understood. Although there are quantitative data regarding incidence of pain and identification of painful sites, there is a dearth of information on the experience of pain and how adolescents understand their pain. One study has examined how adults cope with pain related to CP (Engel, Jensen & Schwartz, 2006); however, there is a need to understand how adolescents understand and experience pain in their everyday lives.

Children and adolescents with CP demonstrate lower levels of physical activity than their peers without disabilities (Pirpiris & Graham, 2004; Bjornson, Belza, Kartin, Logsdon & McLaughlin, 2007). They had significantly less uptime (Pirpiris & Graham, 2004), fewer daily step counts have been reported and daily walking activity decreased with functional ability (Bjornson et al., 2007). Similarly, young ambulatory children with CP were not participating in activities that provided enough intensity to reap the health

benefits of physical activity (van den Berg-Emons, Sarls, de Barbanson, Westerterp, Huson & van Baak, 1995). Also, activities chosen were of slower tempo compared to their peers without disabilities (Maher, Williams, Olds & Lane, 2007; Brown & Gordon, 1987). Others have identified a much larger proportion of sedentary participants with CP (29%) compared to an able-bodied comparison group (10%) (Longmuir & Bar-Or, 1994). As well, physical activity scores decrease on average during the adolescent years after a peak between the ages of 10 to 12 years, and adolescents adopt a more sedentary lifestyle during their second decade (Longmuir & Bar-Or, 1994). Studies conducted around the world have also demonstrated that adolescents with CP are less active than their peers (Stallings, Zemel, Davies, Cronk & Charney, 1996; Bandini, Schoeller, Fukagawa, Wykes & Dietz, 1991; Margalit, 1981; Maltais, Pierrynowski, Galea, Matsuzaka & Bar-Or, 2005; Longmuir & Bar-Or, 2000). Maher and colleagues (2007) investigated physical activity patterns of adolescents with CP aged 11 to 17 years. The least physically active response on a self-report measure was consistently reported and they reported lower average levels of physical activity compared to age-matched controls (Maher et al., 2007). They found a strong association between overall physical activity level and gross motor function, such that more physical activity is associated with higher levels of motor function. They also demonstrated a significant inverse relationship between physical activity and age (Maher et al., 2007).

Brunton & Bartlett (2010) provided rates of exercise participation among adolescents with CP across all GMFCS levels. In their study, participants with more gross motor function, regardless of gender, reported greater exercise participation than those with less motor function. In addition, exercise participation decreased over the four-year period of the study (Brunton & Bartlett, 2010). The overall participation rates were low; only 9.5% of males and 6.5% of females met the Health Canada recommendations for moderate activity and only 11.7% of males and 7.8% of females met the recommendations for vigorous exercise. The authors also found that participants in GMFCS Levels IV and V were less likely to engage in moderate activity than those in levels I to III (Brunton & Bartlett, 2010).

One study has outlined some recommendations for rehabilitation professionals around physical fatigue. The authors suggested the need to understand how fatigue is



impacting a client's life and ability to accomplish their activities of daily living (Svien, Berg & Stephenson, 2008); however, to date this has not been empirically studied. Jones (2009) has provided some personal reflections and recommendations to further study the impact of aging with CP, secondary conditions and the impact on participation in life events. In addition, the potential relationships between fatigue, pain and physical activity have not been fully explored in youth and young adults with CP.

One study demonstrated that physical activity was between 70 and 100% more frequently reported in respondents who experienced a low prevalence of secondary conditions, suggesting that physical activity may contribute to preventing fatigue, pain, and functional deterioration in adults with CP (Jahnsen, Villien, Aamodt et al., 2003). The relationship between fatigue and physical activity has been studied using a variety of methods. Maltais and colleagues (2005) hypothesized that the low physical activity levels observed in their study may have been a compensatory mechanism to prevent fatigue. The authors further demonstrated a relationship between low walking economy and low physical activity levels (Maltais, Pierrynowski, Galea & Bar-Or, 2005), accentuating a possible relationship between fatigue and physical activity level. Another group of authors hypothesized that adults with CP who had low physical activity levels (both objectively and subjectively measured) and low physical fitness would experience more fatigue (Nieuwenhuijsen, van der Slot, Dallmeijer, Janssens, Stam et al., 2011). Although the study determined that adults with CP had low physical fitness levels and were less physically active than their peers with at least 50% of their sample experienced fatigue, the only relationship between activity and fatigue was demonstrated in men with CP, in whom lower physical fitness was positively related to experiencing more fatigue. However, the study conducted by Nieuwenhuijsen and colleagues (2011) had a limited sample size (n=42, 29 men and 13 women), therefore further research is necessary. A second study also demonstrated a weak relationship between physical fitness and fatigue, and once again only for male participants (van der Slot et al., 2012). The relationship between pain and physical activity level has been explored in a study by Sandstrom and colleagues (2004) in which increased pain was often reported in conjunction with inactivity, further highlighting a potential relationship between pain and exercise or physical activity participation (Sandstrom et al., 2004).

Any potential relationship between fatigue and pain in adults with CP is yet not fully understood. One group demonstrated significant moderate correlations between the number of painful sites and both general and cognitive fatigue, along with similar correlations between the impact caused by pain and overall fatigue in non-ambulatory adults with CP (Malone & Vogtle, 2010). However, in ambulatory adults with CP no relationship between fatigue and pain were demonstrated (Malone & Vogtle, 2010). Another group demonstrated pain to be a significant predictor of fatigue for adults with CP (Jahnsen et al., 2003).

Finally, the relationship between fatigue and the severity of CP is also undetermined; some authors demonstrated that severity of CP was not associated with chronic fatigue (Jahnsen, Villien, Aamodt et al., 2003), suggesting that fatigue is an individual experience that may be unrelated to GMFCS level. Another group has demonstrated an increase in energy cost of walking with increasing GMFCS level (Johnston, Moore, Quinn & Smith, 2004) that suggests that the level of fatigue experienced may be dependent on the severity of the condition.

In summary, further research is necessary to fully understand the relationships between fatigue, pain and physical activity. However, in order to understand the relationships amongst and between these variables, a valid and reliable measure of fatigue is needed. In the next section I discuss the importance of measurement development and the requirements necessary to construct a valid and reliable measurement tool.

### *Measurement Development*

Measurement tools can be created for three different purposes. A discriminative index distinguishes between individuals with and without a particular characteristic or function (Rosenbaum et al., 1990; Kirshner & Guyatt, 1985). A predictive index classifies people into categories based on what is to be expected of their future status (Rosenbaum et al., 1990; Kirshner & Guyatt, 1985). Evaluative indices are designed to measure change over time and/or change in response to an intervention (Rosenbaum et al., 1990; Kirshner & Guyatt, 1985). The measurement tool created for this dissertation was constructed for both discriminative and predictive purposes. These purposes were considered when devising the items and determining how they would be scaled. The

scaling requirements for the items are contingent on the item wording and the purpose (discriminative or predictive) it is intended to serve. A summary of the item requirements for both underlying purposes of this project can be found in Table 1-1.

***Table 1-1 - Item Requirements by Test Purpose***

	Discriminative	Predictive
Item Selection	<ul style="list-style-type: none"> <li>- Items should represent all important components of the domain of interest</li> <li>- Items should apply to all possible respondents</li> <li>- Items must be stable over time</li> </ul>	<ul style="list-style-type: none"> <li>- Items should be statistically associated with a criterion measure</li> </ul>
Item Scaling	<ul style="list-style-type: none"> <li>- Response options should be short and have uniform interpretations</li> </ul>	<ul style="list-style-type: none"> <li>- Available response options should maximize correlations with the criterion measure</li> </ul>
Item Reduction	<ul style="list-style-type: none"> <li>- Through internal scaling or consistency</li> <li>- Must weigh the comprehensiveness of the scale versus respondent burden</li> </ul>	<ul style="list-style-type: none"> <li>- Must weigh power to predict versus respondent burden</li> </ul>

(Adapted from Kirshner & Guyatt, 1985).

Although the scale was created with both discriminative and predictive purposes in mind, validation of this scale for predictive purposes was not feasible within the scope of this dissertation, and will be an area of future study. Table 1-2 contains information about the psychometric properties that are important for the validation of a scale for discriminative purposes.

**Table 1-2 - Reliability and Validity Requirements for Discriminative Purposes**

	Discriminative Purpose
Reliability	- Large and stable inter-subject variation - Correlation between repeated measures
Validity	- Cross-sectional construct validity - Relationship between index and external measures at a single point in time

(Adapted from Kirshner & Guyatt, 1985; Portney & Watkins, 2000)

### *Focus of this Dissertation*

This dissertation is the first to explore fatigue from the perspective of the individual living with CP and consider it within the context of available empirical data available on fatigue in this population. The end result of this dissertation was the creation of a measurement tool to identify individuals with differing levels of fatigue associated with CP, as well as a preliminary exploration into the fatigue, pain and physical activity complex.

### *Review of the Mechanisms of Fatigue in CP*

The initial study in this program of research was a critical review of the mechanisms of fatigue in CP and an interpretation of the available evidence about individuals with CP and their fatigue experience and forms Chapter 2 of this dissertation. It was completed as one of the requirements for the comprehensive examination process. This chapter is intended to serve as a foundation to inform the subsequent studies. The focus of this dissertation is not on the mechanisms of fatigue; rather this review serves as background information to understand the challenge associated with the measurement of fatigue and with the exploration of the phenomenon of fatigue in youth and young adults with CP. The research questions answered in this chapter are:

- What factors may be responsible for the conflicting reports of fatigue in individuals with CP?
- At which sites in the neuromuscular system do variations exist in individuals with CP compared to their peers without disability?

### *Review of Existing Fatigue Measures*

The second study in this dissertation was a systematic review of all available measurement tools that characterize or measure fatigue in conditions of the nervous system that may be similar to CP in the experience and impact of fatigue. From this review of available tools, items relevant to the CP population were selected for inclusion in pilot testing of a new measurement tool. This review forms Chapter 3 of the dissertation and is one method used to generate items for the new fatigue measure. The research questions answered in this chapter are:

- What, if any, is the best scale to measure fatigue in a population with CP?
- Which fatigue scales have evidence of rigorous psychometric validation for use with populations with neurological conditions?

### *The Bodily Experience of CP*

The third study explored feelings of muscle fatigue and pain in adolescents and young adults with cerebral palsy. Specifically, this study was a phenomenological inquiry to understand and describe the bodily experience of CP. The aim was to understand the lived body, with a focus on fatigue and pain, as it was experienced by individuals with CP. This study is outlined fully in Chapter 4. The information gathered from the phenomenology was used as a client-centered method of generating or selecting items for the measurement scale. The research question answered in this chapter is:

- What is the bodily experience of living with CP?

### *Scale Creation and Pilot Testing*

Chapter 5 comprises the detailed creation of the new fatigue measure. After all items were added to the item bank (both from clients through the phenomenology and from the systematic review), focus groups were conducted with health care professionals who regularly worked with individuals with CP, to reduce the number of items contained in the scale. After the focus groups, pilot testing of the scale was completed with a small number of participants (n=5) to assess the feasibility of the scale in the population of interest. An advisory committee meeting was called to discuss the merits of different scaling options once the final item bank had been generated for this measure. The research questions answered in this chapter are:

- Which items should be removed from the preliminary fatigue scale?

- Are there any additional items to be added to the preliminary fatigue scale to reflect underrepresented constructs?
- Is the Fatigue Impact and Severity Self-Assessment feasible to complete by individuals with CP aged 14-25 years?

*Determining the Psychometric Properties of the Fatigue Impact and Severity Self-Assessment*

The fourth study (and Chapter 6) in this dissertation comprised a large sample of participants (n=130) who were contacted by mail with the final questionnaire to assess the discriminative validity and the reliability of the tool. The internal consistency of the new measurement scale was assessed using Cronbach's alpha, and test-retest reliability was assessed over an interval ranging from 2 to 4 weeks with a small subset of the participants (n=31) using an intraclass correlation coefficient. Tests of known-groups, convergent and divergent validity were used to establish the construct validity of the measure. Additionally, an exploratory factor analysis was performed to gather information about the factor structure of the newly developed questionnaire. The research questions answered in this chapter include:

- What is the factor structure of the Fatigue Impact and Severity Self-Assessment?
- Is the Fatigue Impact and Severity Self-Assessment a valid measure of fatigue for individuals with CP aged 14-30 years?
- Is the Fatigue Impact and Severity Self-Assessment a reliable measure of fatigue for individuals with CP aged 14-30 years?

*Summary*

Understanding the ways in which fatigue, pain, and physical activity impact the daily activities of adolescents and young adults living with CP may provide important information for clinicians and other care providers working with these individuals, especially in planning for transition from the pediatric to adult health care settings. The remaining chapters in this dissertation will outline, in detail, the studies aimed at understanding and assessing fatigue, pain and physical activity in adolescents and young adults with CP.

It should be noted that although they are related, each chapter within this dissertation is a separate study with specific inclusion and exclusion criteria as described in the method section of each particular chapter. The specific subset of the population of interest (particularly in regard to age and functional level) evolved over the course of the four studies contained within the dissertation. As such, it was decided to continue to change the inclusion and exclusion criteria as new information emerged and as new subject groups were interested in participating in this research program. I considered this emergent and pragmatic standpoint to be important in ensuring the maximum relevancy of the final outcome of this dissertation, the Fatigue Impact and Severity Self-Assessment. Given the exploratory nature of the studies contained in this dissertation I felt that including as many viewpoints as possible would provide the most comprehensive and useful version of the fatigue measure.

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## **Chapter 2: A Critical Review of Reports of Fatigue in Cerebral Palsy**

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### *Introduction*

This chapter comprises a critical review of the mechanisms of muscle fatigue in individuals with cerebral palsy (CP). Although the direct study of the anatomical and physiological aspects of fatigue are not the focus of this dissertation, this review establishes a basic understanding of the differences in the fatigue process between individuals with CP and their peers without disability. These differences may suggest a need to measure fatigue differently for individuals with and without CP, in addition to highlighting the specific challenge of measuring fatigue (in this case objectively) in individuals with CP.

One of the most common impairments experienced by adults with CP is fatigue (Hilberink, Roebroek, Nieuwstraten, Jalink, Verheijden & Stam, 2007). However, controversy exists in the available information about individuals with CP and the experience of fatigue. For example, recent laboratory studies have shown individuals with CP to be less fatigable than control participants (Moreau, Li, Geagan & Damiano, 2008; Stackhouse, Binder-Macleod & Lee, 2005), which is in conflict with reports that fatigue is a chronic and disabling symptom of CP (Hilberink et al., 2007). Locomotion is not usually a considerable cause of fatigue in the neuromuscular system, but for individuals with CP, fatigue becomes an important factor during ambulation over both short and long distances depending on the severity of the motor impairment and the terrain. Thus, in this context, adults with CP experience higher levels of fatigue than the general population (van der Slot et al., 2010; Opheim, Jahnsen, Olsson & Stanghelle, 2009) and with chronic pain it is estimated that this combination affects 34% of the population with CP (van der Slot et al., 2010). Indeed, a 7-year follow-up study reported that individuals whose walking ability deteriorated had increased levels of physical fatigue, but no significant differences existed between a sample with CP and the general population in terms of mental fatigue. These results indicate that the fatigue experienced

in CP is mainly of physiological origin (Opheim et al., 2009), but the factors responsible are unclear. Thus, the purpose of this review is to synthesize information to explain the conflicting nature of fatigue in CP (with a specific focus on spastic CP), and to describe the characteristics of CP as it relates to fatigue during functional tasks of daily living (Tsoi, Maher, Moore, Goldstein & Aisen, 2009). It should be noted that this is not a systematic review, but rather a critical examination to highlight gaps in our understanding to help design future studies towards a better understanding of the important aspects of fatigue in this population. Furthermore, this review also may help evaluate current techniques and therapies and to develop new methods to alleviate fatigue in individuals with CP.

In this review, fatigue is defined as a reduction in muscle force-generating capacity in the neuromuscular system that occurs during prolonged or ongoing activity (Bigland-Ritchie, Johansson, Lippold & Woods, 1983). The development and maintenance of a given level of force are dependent on the integration of the entire neuromuscular system; there are several points in the system that can fail and lead to fatigue and fatigue may occur at more than one site concurrently (McComas, Miller & Gandevia, 1995). Factors contributing to fatigue are organized into three sections: central factors, peripheral anatomical factors and peripheral physiological factors.

*Reports of fatigue in cerebral palsy: The conflict*

In a controlled isokinetic protocol, the knee flexors and extensors of individuals with CP were shown to be less fatigable compared to a group without disability. However, torque was normalized to the maximum peak torque and this normalized torque declined less in the individuals with CP than in the comparison group (Moreau et al., 2008). Additionally, individuals who were the least severely affected (as measured by the Gross Motor Function Classification System (Palisano, Rosenbaum, Bartlett & Livingston, 2008) (GMFCS)) and considered to have higher functional ability had greater rates of decline in their normalized peak force for the knee extensors (Moreau et al., 2008). One other study demonstrated a significant difference in the decline of normalized peak force after an electrically elicited fatiguing protocol. Children with CP experienced a 42% decline in normalized peak quadriceps femoris force compared to the decline of 52% in the control group (Stackhouse et al., 2005). In both of these studies, absolute

maximum torque was between 50 to 73% less in individuals with CP, compared to controls, and thus normalization for comparison between groups could be misleading. As a result of this lower overall strength compared to their peers without a disability (Stackhouse et al., 2005) normalizing the force reduction that occurs with fatigue to the child's own maximal force capacity removes the functional practicality from the situation. Thus, although children with CP have reduced fatigue compared to non-disabled peers, only part of the picture of fatigue is being recognized in this population. Despite that individuals with CP appear to fatigue less than their non-disabled peers, the measurement of fatigue in the laboratory (i.e. using dynamometry of isolated muscles instead of functional tasks) does not reflect the chronicity of the problem. It is also possible for individuals with CP to have greater resistance to fatigue in the laboratory than the general population but still experience a greater subjective feeling of fatigue, and a greater impact of fatigue on activity, as a result of the inefficient mechanics associated with gait in CP (Moreau et al., 2008). Therefore, studies comparing fatigue data between individuals with CP and their peers developing typically are confounded by several factors. Specifically, normalization to peak torque can be misleading without also including a comparison in absolute terms (i.e. the use of whole body weight). The normalized data from laboratory studies also may be affected by factors such as co-contraction and variability in the most effective order of activating muscles, both of which have been demonstrated in individuals with CP, but not in peers without CP, causing comparisons between the two groups to be misleading.

A study by Leunkeu and colleagues (2010) demonstrated increased fatigue in individuals with hemiplegic CP compared to controls by assessing the decline in the slope of the median frequency of a surface electromyography (EMG) recording of the vastus lateralis and rectus femoris muscles. In this study, participants with CP required higher levels of motor unit recruitment along with lower median frequency compared to control participants. This indicated increased skeletal muscle fatigue that has been linked to abnormal muscle function (Leunkeu, Keefer, Imed & Ahmaidi, 2010). The authors used a measure of muscle activity to observe the effects of the contraction without relying on an inference from a decline in force production to demonstrate increased fatigue in a sample with CP (Leunkeu et al., 2010). These results further highlight the need for

caution when using force normalization techniques that can lead to inaccurate conclusions of fatigue resistance in CP. Measures of muscle activity from EMG therefore may be a useful adjunct to measures of force to provide a more complete or accurate description of fatigue.

Further, it is troubling that those individuals who are already weaker than their peers are still experiencing a large reduction in muscle force during fatiguing tasks. This reduction in force has the potential to have a great impact on the capacity to continue performing activity and thus becomes a larger drain on the force reserve needed for continuing activity in individuals with CP compared to their peers. A decline of 42% of their peak torque producing capability with fatigue may have more of an impact on their ability to continue performing daily activities due to a reduced maximal absolute force generating capacity and higher energy expenditure demands. Individuals with CP have weakness that may be due to a variety of factors (discussed in the peripheral factors section), and the demands of walking require a greater percentage of the force generation capacity of the muscle (Moreau et al., 2008) and of the individuals' maximal oxygen consumption for individuals with CP (Leunkeu, Gayda, Nigam, Lecoutre & Ahmaidi, 2009). A study by Slaman and colleagues (2013) demonstrated that individuals with CP use a larger portion of their metabolic reserve during walking when compared to their peers. The increased demands on the muscle relative to its reduced overall capacity are reflected in reports of chronic fatigue in this population. In addition, several studies have documented an increased co-activation/co-contraction of agonist and antagonist muscles around the same joint in individuals with CP (Stackhouse et al., 2005; Unnithan, Dowling, Frost & Bar-Or, 1996; Burtner, Qualls & Woollacott, 1998). Small amounts of co-activation are normal; however, too much co-activation increases energy expenditure and as a result could lead to a faster rate of fatigue in both agonists and antagonists (Unnithan et al., 1996; Feltham, Ledebt, Deconinck & Savelsbergh, 2010). It should be noted that one laboratory study has demonstrated higher hamstring co-contraction, spasticity and reduced hamstring strength to be predictive of fatigue resistance in the hamstring muscles (Moreau, Li, Geaghan & Damiano, 2009). Additionally, higher quadriceps co-contraction and lower quadriceps strength were predictive of fatigue resistance in the quadriceps muscles (Moreau et al., 2009). The authors suggest that this



relationship may be a result of the muscle adapting to disordered neural inputs; that is, constant co-contraction with movement may act as a training stimulus (Moreau et al., 2009). It is unknown how this process may translate to functional tasks such as walking, but it should be explored further.

It is possible that during functional activities of daily living, such as walking over long distances, individuals with CP experience a decline in force production greater than that demonstrated in an isolated laboratory setting. Functional tasks require individuals to manage and negotiate their entire body weight, which stresses the neuromuscular system in a different manner than has previously been studied. One study has reported individualized accounts of persons with CP experiencing more fatigue during activities where managing their entire body weight was required compared to seated or water activities (Brunton & Bartlett, 2013). In addition, recovery from fatigue has not been studied in a population with CP and important differences may exist between individuals with CP and their non-disabled peers in recovery rate or time that may help explain the conflicting reports of fatigue in this population.

#### *Central sites*

The neural factors contributing to fatigue have not been studied extensively in CP, but some studies have indicated disorder in and damage to the corticospinal projections to the lower motor neurons in individuals with CP (Cheney, 1997; Rose & McGill, 2005; Brouwer & Ashby, 1990). Mechanisms of reciprocal excitation of antagonists in CP that contribute to co-activation of muscles around a joint have been proposed in a comprehensive review by Cheney (1997). The first mechanism suggests corticospinal disorganization or abnormal synaptic organization, in which corticospinal neurons co-facilitate the motoneurons of both flexor and extensor muscles around a joint (either monosynaptically or through interneurons) (Cheney, 1997). A second mechanism could be the result of abnormal synaptic organization at the spinal level, where spindle afferents from agonists excite both the agonist and antagonist muscles around a joint (Cheney, 1997). Cheney (1997) also described evidence from reflex studies demonstrating a reduction of presynaptic inhibitions acting on muscle spindle afferents as a contributing factor to spasticity in CP (Cheney, 1997). In addition, Heinen and colleagues (1999) demonstrated a lack of inhibitory control in the motor cortex of adolescents with CP. The

inability or loss of descending inhibition to antagonist or synergist muscles may contribute to reduced synchronization of motor unit firing and potentially contribute to observed muscle weakness and increased co-contraction in CP (Heinen, Kirschner, Fietzek, Glocker, Mall & Korinthenberg, 1999).

Evidence of corticospinal disorganization was shown by reduced synchronization of motor units in the tibialis anterior (TA) of individuals with CP, hypothesized to be due to a decrease in cortico-motoneuronal connections (Rose & McGill, 2005). Others (Brouwer & Ashby, 1990) have inferred abnormal development of projections from the motor cortex to spinal motoneurons that contribute to abnormal patterns of muscle activation in CP. This is manifested as the loss of specificity of the projections from the motor cortex to the motoneuron pools of the lower limb muscles and was demonstrated by similar activation of the TA and the soleus muscles in individuals with CP as a result of magnetic stimulation intended to produce activation of the TA only (Brouwer & Ashby, 1990).

Other evidence of corticospinal tract damage demonstrated that children with CP do not show the tonic suppression of H-reflexes during the stance phase in gait observed in children developing typically (Hodapp, Klisch, Mall, Vry, Berger & Faist, 2007). The suppression of the H-reflex happens as the corticospinal tract matures and it has been hypothesized that the immature pattern persists in children with CP due to corticospinal tract damage (Hodapp et al., 2007). In a laboratory setting, individuals with CP may appear less fatigable than the general population as a result of the inability of the descending tracts to fully transmit the signal to the muscles needed to produce a maximal contraction. However, functionally, a decreased efficiency of the descending signal from the motor cortex through damaged or abnormal projections to agonist and antagonist muscles around a joint can also lead to increased co-activation and increased energy expenditure, a potential cause of fatigue in individuals with CP (Unnithan et al., 1996), as discussed above.

In a study of activation and recruitment of motor units in individuals with CP, Stackhouse et al. (2005) demonstrated significantly lower voluntary muscle activation ratios and lower force production for children with CP compared to controls, and therefore less muscle fatigue because the muscle was insufficiently activated; this

information is summarized in Table 2-1. Rose & McGill (2005) demonstrated that maximal M-wave amplitudes were similar between a group with CP and a control group, suggesting that the total numbers of available motor units were not different (Rose & McGill, 2005) although maximum voluntary contraction (MVC) torque was less in the group with CP for both the TA and gastrocnemius muscles. Additionally, the surface EMG amplitudes during MVC and, therefore, the levels of neuromuscular activation (NMA) during MVC were significantly smaller in the group with CP. A decrease in NMA with the unaltered M-wave reflects the inability to fully activate all available motor units to sustain the required level of torque.

Elder et al. (2003) demonstrated lower mean amplitude of EMG activity from both the plantar flexors (50% lower) and dorsiflexors (40% lower) of children with CP. The reduction in mean amplitude is also thought to reflect incomplete muscle activation either through an inability to activate available motor units or due to the inability to recruit higher threshold motor units (Rose & McGill, 2005; Elder, Stewart, Cook, Weir, Marshall & Leahey, 2003). Rose & McGill (2005) demonstrated an intact relationship between recruitment and firing rate modulation at low to moderate levels of contraction evidenced by increased firing rates and recruitment with increasing voluntary activation of the muscles for both a group with CP and those without. However, the submaximal contractions required more voluntary effort for participants with CP as reflected by the highest target NMA levels corresponding to about 50% of the MVC NMA levels for individuals with CP and approximately 20% of the MVC NMA levels for the controls (Rose & McGill, 2005). A person with CP can produce an equivalent contraction in terms of recruitment and firing rates as a control, but may require full voluntary effort compared to a submaximal effort for the control. Controls can then increase contraction strength by increasing firing rate or recruitment while the person with CP cannot (Rose & McGill, 2005) and theoretically, those with CP would experience more fatigue. Rose & McGill (2005) also calculated a projected maximal firing rate for the TA and gastrocnemius for both groups (Table 2-1) to suggest that maximal firing rates in individuals with CP are reduced by approximately 50% compared to controls, potentially due to impairment of, or a decrease in, the number of cortico-motoneuronal connections

(Rose & McGill, 2005), which may be a contributing factor in reports that children with CP are less fatigable than their peers.

**Table 2-1 - Motor Unit Activation in Individuals with Cerebral Palsy**

<b>Study</b>	<b>Muscle(s) examined</b>	<b>Parameter measured</b>	<b>Difference between CP and controls</b>
Robertson et al. 1984	Tibialis anterior	Silencing motor unit activation	Children with CP had difficulty silencing motor units at both the gross and single motor unit level after activation.
Stackhouse et al. 2005	Quadriceps femoris	Voluntary muscle activation ratios <sup>†</sup>  Knee extension force	Children with CP had 33% less voluntary activation than control children  Lower voluntary activation corresponded with 56% less force production in the children with CP
Stackhouse et al. 2005	Triceps surae	Voluntary muscle activation ratios  Plantar flexion force	Children with CP had 49% less voluntary activation than control children  Lower voluntary activation corresponded with 73% less force production in the children with CP
Rose et al. 2005	Tibialis anterior  Gastrocnemius	Projected maximal firing rate  Projected maximal firing rate	Children with CP had a projected maximal firing rate of 16Hz compared to 31Hz for the control group  Children with CP had a projected maximal firing rate of 13Hz compared to 25Hz for the control group

<sup>†</sup>Voluntary activation ratios are calculated by stimulating the muscle during a maximal voluntary contraction and dividing the augmentation of force by the force of stimulation at baseline

Skeletal muscles are normally electrically silent when there is no movement (Roberston, Lee & Jacobs, 1984). Robertson et al. (1984) showed individuals with CP had difficulty in achieving electrical silence after performing a contraction of the TA and thus required more trials to learn to silence the muscle after activity. Individuals with CP also had difficulty silencing a single motor unit after achieving activation. This demonstrates an inability to inhibit the TA muscle at both the motor unit and gross motor level (Robertson et al., 1984). The delayed ability to silence motor units after activity may contribute to the co-activation around a joint during reciprocal movements. In the example of gait, the different phases require activation and inhibition of different muscles in a short span of time. Inability to silence motor units after activation during this task could contribute to increased energy expenditure reported in individuals with CP.

Overall it seems that although children with CP may be performing at or near maximal effort, there is also the possibility that not all of their motor units can be activated as a result of impaired motor pathways (Rose & McGill, 2005). It has also been hypothesized that in the case of incomplete activation, type I fibers are preferentially recruited (or there is an inability to recruit higher threshold motor units) (Rose & McGill, 2005; Elder et al., 2003) at lower firing rates, which may contribute to the observed greater fatigue resistance evidenced in the CP population in laboratory settings using normalized comparisons (Moreau et al., 2008). However, incomplete activation can result in individuals with CP using more effort to produce a contraction and maintain movement, which could result in increased, or early onset of, fatigue.

#### *Peripheral sites – Anatomical features*

There is limited information regarding the structure and function of the sites in the peripheral neuromuscular system that directly relate to fatigue in individuals with CP. The neuromuscular junction is the link between the central and peripheral aspects of the neuromuscular system. Some research has suggested that individuals with CP have extrajunctional acetylcholine receptors and other structural differences that can affect the depolarization and re-polarization of the muscle membrane following a signal from the descending motor pathways (Theroux et al., 2005; Theroux, Akins, Barone, Boyce, Miller & Dabney, 2002). A relationship has been demonstrated between increased

severity of impairment in motor function and increased abnormality of the neuromuscular junction (Theroux et al., 2005). The implications of these differences are not fully understood and increased fatigue during functional tasks in individuals with more severe motor impairments may be a result of decreased function of the neuromuscular junction in communicating the signal from the descending motor pathway.

Reports on muscle size and the resulting strength of individuals with CP has produced conflicting results and hypotheses related to fatigue in CP. Some authors attribute the lower mean torque values (and less relative fatigue) observed in individuals with CP to differences in muscle size because greater strength or muscle mass may contribute to higher levels of fatigue. Leg volumes, cross-sectional area, muscle thickness, fascicle length and pennation angle all have been shown to be reduced in people with CP (Elder et al., 2003; Moreau, Teefey & Damiano, 2009). A summary can be found in Table 2-2. Thus because smaller muscles generate less force and less force creates less fatigue, these features have the potential to contribute to the observed increased fatigue resistance in the laboratory testing of individuals with CP. It is not clear whether these observed anatomical changes are secondary to the damage in the central nervous system or a tertiary condition as a result of decreased activity.

An alternative hypothesis suggests that weakness as a result of lower muscle mass can increase levels of fatigue as a result of the need to recruit more motor units to achieve a given force level and due to a greater frequency of excitation required to perform a given task at an absolute force requirement (Lindstrom, Lexell, Gerdle & Downham, 1997). This is supported by reports of relatively more fatigue during functional tasks, such as ambulation, and warrants further investigation using test parameters that are not confounded by differences in skeletal muscle function. Reports that individuals with CP are more functionally fatigable may be related to a lower force-generating capacity that can lead to a lower force reserve in order to maintain constant activity as discussed earlier (Stackhouse et al., 2005). Similar to older adults, activities of daily living require individuals with CP to use a greater percentage of their maximal strength. Literature on aging has suggested that the rate of recovery from fatigue may also be impaired following these tasks (Allman & Rice, 2002), but this has not been tested in CP.

*Table 2-2 - Muscle Parameters in Individuals with Cerebral Palsy*

<b>Study</b>	<b>Muscle(s) examined</b>	<b>Parameter measured</b>	<b>Difference between CP and controls</b>
Elder et al. 2003	Anterior compartment: tibialis anterior, extensor digitorum longus, extensor hallucis and peroneus tertius	Muscle volume (MRI)	27% less for individuals with CP
Elder et al. 2003	Posterior compartment: soleus, gastrocnemius, plantaris, popliteus, tibialis posterior, flexors digitorum longus and hallucis longus	Muscle volume (MRI)	28% less for individuals with CP
Moreau et al. 2009	Rectus femoris	Cross-sectional area (Ultrasound)	48.5% lower in individuals with CP
Moreau et al. 2009	Rectus femoris Vastus lateralis	Muscle thickness	32% lower in individuals with CP 31% lower in individuals with CP
Moreau et al. 2009	Rectus femoris	Fascicle length	27% shorter in individuals with CP
Moreau et al. 2009	Vastus lateralis	Pennation angle	3 degrees less in individuals with CP

MRI = Magnetic Resonance Imaging

A potential contributor to the fatigue resistance observed in laboratory studies of individuals with CP may be a predominance of a specific fiber type (Moreau et al., 2008). Indeed the few available reports indicate evidence of increased area of type I muscle fibers, or atrophy of type II fibers in CP (Ito, Araki, Tanaka, Tasaki, Cho & Yamazaki, 1996; Marbini, Ferrari, Cioni, Bellanova, Fusco & Gemignani, 2002; Rose, Haskell, Gamble, Hamilton, Brown & Rinsky, 1994). Muscle biopsies provide evidence of variability in fiber size with reduced diameters of type I and II fibers, that is significantly more frequent in individuals with CP over 10 years of age, but no evidence of degeneration (Ito et al., 1996; Marbini et al., 2002; Rose et al., 1994).

Greater variation in fiber size was detected in the more severely affected side, indicating that the severity of CP may be a contributing factor to alterations in muscle composition (Ito et al., 1996). In addition, studies have shown varying degrees of



disorganization or disorientation of myofibrils in individuals with CP (Marbini et al., 2002; Rose et al., 1994). Together these reports indicate that changes in the motor cortex and the descending pathways influence the organization of motor units at the spinal level. A predominance of fatigue resistant muscle fibers in CP may explain the observation of reduced fatigue during laboratory testing, such that the muscle fiber composition is different between individuals with CP and their peers. This supports the concern about comparisons that are based only on normalized peak force rather than on absolute capacity. The disorganization of the myofibrils could also impair excitation-contraction coupling, further reducing the strength of each muscle contraction. A predominance of type I fibers could contribute to decreased strength in individuals with CP, having the potential to increase their fatigue during functional tasks requiring high forces or prolonged activity.

#### *Peripheral sites – Physiological features*

Sufficient blood flow is essential to maintain muscle force production in terms of oxygen delivery and removal of metabolic by-products created during contractions. Some have suggested that spastic muscles do not properly support venous return as a result of increased muscle tone associated with CP, resulting in inhibited muscle lactate and metabolite clearance during activity (Hoofwijk, Unnithan & Bar-Or, 1995; Lundberg, 1978). Decreased clearance of metabolic byproducts may contribute to lower maximal oxygen consumption and increased or earlier onset of fatigue in individuals with CP.

Many studies have documented increased energy expenditure in individuals with CP compared to their peers without disabilities; a summary can be found in Table 2-3 (Leunkeu et al., 2010; Leunkeu et al., 2009; Campbell & Ball, 1978; Duffy, Hill, Cosgrove, Corry & Graham, 1996; Johnston, Moore, Quinn & Smith, 2004; Bell & Davies, 2010; Rose, Haskell & Gamble, 1993; Keefer et al., 2004). Youth with CP have higher energy expenditure during locomotion than their peers at similar speeds and the rate of energy expenditure increases as children with CP age (Campbell & Ball, 1978). An increase in body weight and size as a child matures increases the demand for energy during locomotion and other activities, and requires greater physical exertion (Campbell & Ball, 1978). During gait, adolescents with CP work at a higher percentage of their maximal oxygen consumption compared to the youth developing typically (one study

reported values of 53.5% and 22.5% respectively) (Unnithan et al., 1996). Children with CP are both working at a higher percentage of their maximal aerobic power and working harder than their peers at a given speed (or submaximal load). This may cause them to fatigue more easily during prolonged exercise (Leunkeu et al., 2009; Rose et al., 1993). Interestingly, at a given submaximal level there appears to be no differences in the respiratory exchange ratio between individuals with CP and their peers without disabilities (Rose et al., 1993). This indicates similar cardiorespiratory responses to submaximal exercise in both groups demonstrating that the cardiorespiratory response is not a contributing factor to fatigue in CP (Rose et al., 1993). The type and severity of CP can also influence the energy expenditure during walking. Van den Hecke et al., (2007) investigated energy cost in individuals with hemiplegic CP and determined that the increase in energy cost of ambulation was due to an increased mechanical work and that the efficiency of work was similar between individuals with CP and controls when walking at the same speed (Van Den Hecke, Malghem, Renders, Detrembleur, Palumbo & Lejeune, 2007).

**Table 2-3 - Energy Expenditure During Walking in Individuals with Cerebral Palsy**

<b>Study</b>	<b>Task</b>	<b>Measure of energy expenditure</b>	<b>Differences observed</b>
Campbell & Ball 1978	Free walking at comfortable, self-selected pace	Energy cost (oxygen consumption $\text{VO}_2$ )	<p>Energy expenditure was greater in children with CP compared to controls at the same walking speed.</p> <p>Energy expenditure increased as children with CP aged.</p>
Rose et al. 1993	Treadmill walking at various speeds	Oxygen Uptake	<p>Children with CP had higher oxygen uptake than controls while walking the speeds of 21.5m/min and 37.6m/min.</p> <p>Children with diplegia had higher oxygen uptake than children with hemiplegia while walking at the speeds of 21.5m/min and 37.6m/min.</p>
Rose et al. 1993	Treadmill walking at various speeds	Oxygen Pulse	<p>Children with CP had higher oxygen pulse values compared to controls while walking at the speeds of 21.5m/min and 37.6m/min.</p> <p>Children with diplegia had higher oxygen pulse than children with hemiplegia while walking at the speed of 37.6m/min.</p>

<b>Study</b>	<b>Task</b>	<b>Measure of energy expenditure</b>	<b>Differences observed</b>
Rose et al. 1993	Treadmill walking	Most economical walking speed	<p>Most economical walking speed was slower for children with CP compared to controls.</p> <p>Oxygen uptake and oxygen pulse at most economical walking speed was higher for children with CP than controls</p> <p>Oxygen uptake at most economical walking speed was higher for children with diplegia compared to children with hemiplegia</p>
Duffy et al. 1996	Free walking at comfortable, self-selected pace	Oxygen uptake	<p>Children with diplegia had a higher rate of oxygen consumption per minute than a group with spina bifidia, a group with hemiplegia and controls.</p> <p>Children with CP (both hemiplegia and diplegia) had a higher energy cost for walking than the control group.</p>

Study	Task	Measure of energy expenditure	Differences observed
Unnithan et al. 1996	Treadmill walking	Energy cost (use of metabolic cart)	<p>Children with CP had higher values for absolute oxygen consumption, mass-relative oxygen consumption, net mass-relative oxygen consumption, percentage of maximal oxygen consumption, ventilation, heart rate, and net heart rate compared to controls while walking at the speed of 3km/h.</p> <p>When a relative intensity of 90% of the individuals fastest walking speed was used, differences between individuals with CP and controls were only observed in percentage of maximal oxygen consumption</p> <p>In individuals with CP co-contraction of the lower leg and thigh explained 42.8% and 51.4% of the variance in oxygen consumption respectively.</p>
Keefer et al. 2004	Treadmill walking	Energy expenditure (Oxygen consumption, VO <sub>2</sub> )	<p>No relationship was demonstrated between energy expenditure and thigh muscle co-contraction or quadriceps muscle strength in individuals with hemiplegia.</p>

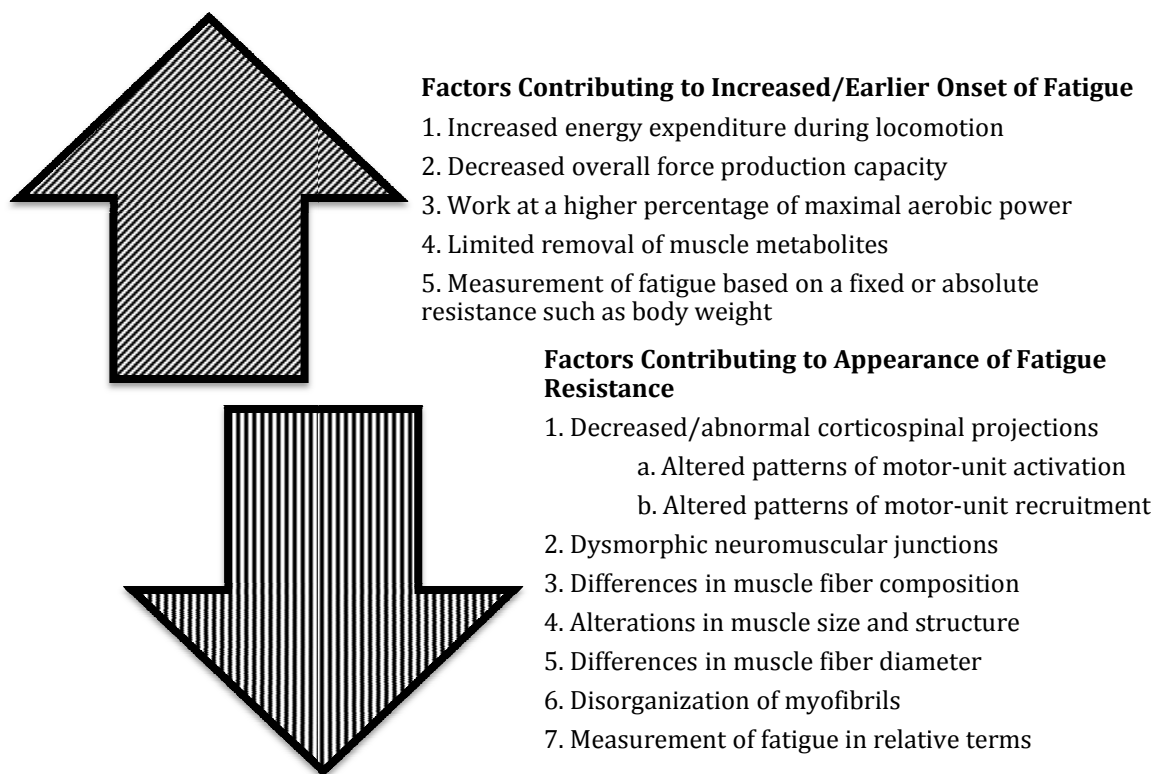
<b>Study</b>	<b>Task</b>	<b>Measure of energy expenditure</b>	<b>Differences observed</b>
Johnston et al. 2004	Walking at self-selected pace	Energy consumption (Volume of oxygen consumed per kilogram of body weight)	<p>Increasing energy cost of walking with increasing GMFCS level.</p> <p>Significant differences between each adjacent GMFCS level.</p> <p>Children with CP demonstrated a higher energy cost of walking than children with typical development.</p>
van den Hecke et al. 2007	Walking on a motor-driven treadmill	Energy cost (oxygen consumption $VO_2$ )	Mean energy cost value was 1.3 times greater for individuals with CP than control values.
Bell & Davies 2010	Free walking	Activity-related energy expenditure (indirect calorimetry)	<p>Children with CP expended more energy than controls.</p> <p>Children with diplegia expended more energy than children with hemiplegia.</p>

$VO_2$  = volume of oxygen consumption

### *Summary and future directions*

Controversy exists as to which factors contribute to fatigue in individuals with CP compared to peers without disability, largely dependent on how fatigue is defined and measured. Figure 1 is a graphic summary of the factors that may explain the resistance to fatigue demonstrated in isolated laboratory studies compared with the factors that may contribute to increased fatigue during functional tasks. It appears that in laboratory settings when strength is normalized, individuals with CP fatigue less than their non-disabled peers; however, comparing fatigue in an isolated manner instead of using a functional task that depends on an absolute load (i.e. body weight) does not reflect the chronicity or impact of fatigue for individuals with CP and can be confounded by several structural and functional differences in the muscles of individuals with CP. Laboratory studies are useful to understand specific sites of differences or potential limitations in the neuromuscular system of individuals with CP, but rehabilitation practitioners must recognize how to interpret these findings for functional tasks that require a certain level of absolute ability to be performed successfully.

It is clear from this critical review that neuromuscular fatigue is an important challenge of individuals with CP, but there is little consensus regarding the underlying mechanisms, and the functional impact. Part of this limitation is due to the few research studies and inconsistencies among these studies in how fatigue is defined and compared. Thus, one important direction is to understand the process of fatigue in individuals with CP, with a focus on functional tasks as they relate especially to locomotion and the use of absolute loads such as body weight. Specifically, a better understanding of the rate of the development and recovery from fatigue is also needed as this may help elucidate important factors underlying the chronic experience of fatigue in CP. Also, studies designed to assess the fatigability of individuals with CP, compared to a control population, need to account for differences in absolute strength and altered muscle function potentially highlighting the effect of a lower force reserve. Finally, it should be noted that the levels of fatigue experienced by individuals with CP may be related to the severity of the disability as measured by the GMFCS, therefore, fatigue should be examined across all GMFCS levels, and potentially, each level independently.



***Figure 2-1 - Factors Contributing to Conflicting Reports of Fatigability in Individuals with Cerebral Palsy***

*Relevance to Thesis*

There are many potential explanations for the physical fatigue experienced by individuals with CP, including the various alterations to the muscle mechanics as described above. As previously discussed, reviewing and/or studying the mechanistic differences of fatigue in individuals with CP is not the primary focus of this dissertation. However, this review was necessary to provide a better understanding of the challenge of measuring and understanding fatigue for these individuals.



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### **Chapter 3: A Review of Fatigue Measures Used in Neurological Conditions: Preparation for the Development of a Measure Specific to Cerebral Palsy**

#### *Introduction*

As described in the introductory chapter, several authors have described a cycle of deconditioning that can occur in cerebral palsy (CP). In this cycle, physical function decreases, followed by a further decrease in physical activity, which can lead to a cascade of further functional decline (Tsoi, Maher, Moore, Goldstein & Aisen, 2009).

Furthermore, as described in Chapter 2, individuals with CP experience deficits in the mechanisms and functions of their muscles and, when combined with the development of secondary conditions during growth and maturation, functional deterioration and early loss of mobility may be experienced (Tsoi et al., 2009).

Fatigue is a prevalent secondary condition experienced by individuals with CP (as previously described); however, there is no accepted measure to obtain information about fatigue in this population. A search was conducted to locate fatigue scales that have been validated for use with individuals with CP. Although several quality of life measures (such as the Short-form 36) may have one or more questions related to fatigue, there are currently no measures dedicated to assessing fatigue in CP nor are there any published psychometric information about any specific fatigue scale for use in CP. The purpose of this study was to conduct a systematic review of the fatigue scales that have been validated for use in other neurological populations that may have a similar experience of fatigue as an individual with CP. Other conditions of interest for this study included: Parkinson's Disease (PD), Multiple Sclerosis (MS), Chronic Fatigue Syndrome (ChFS) and Postpoliomyelitis. Although these four conditions may have different etiologies and manifestations of the primary impairment (than each other and different from CP), there may be similar experiences of fatigue across individuals and conditions because fatigue is a prevalent symptom with a large impact on daily activities (Berlly, Strauser, & Hall, 1991; Friedman & Friedman, 2011; Krupp, Alvarez, LaRocca and Scheinberg, 1988) or that the entire neuromuscular system may be involved in the experience of fatigue, as it is in CP. Furthermore, because the fatigue experience in CP is not well understood, including the conditions of MS and ChFS may provide insight about fatigue that may be

independent of exertion. Including the conditions of PD and Postpoliomyelitis will help gain information about fatigue that is related to exertion or physical activity (Aaronson, Teel, Cassmeyer, Neurberger, Pallikkathayil, Pierce et al., 1999). The results of this study will aid in determining which self-report fatigue scale (or which items) to use with a population of individuals with CP. Self-report scales were specifically chosen over laboratory based methods of determining fatigue as I was primarily interested in measuring the subjective experience of fatigue.

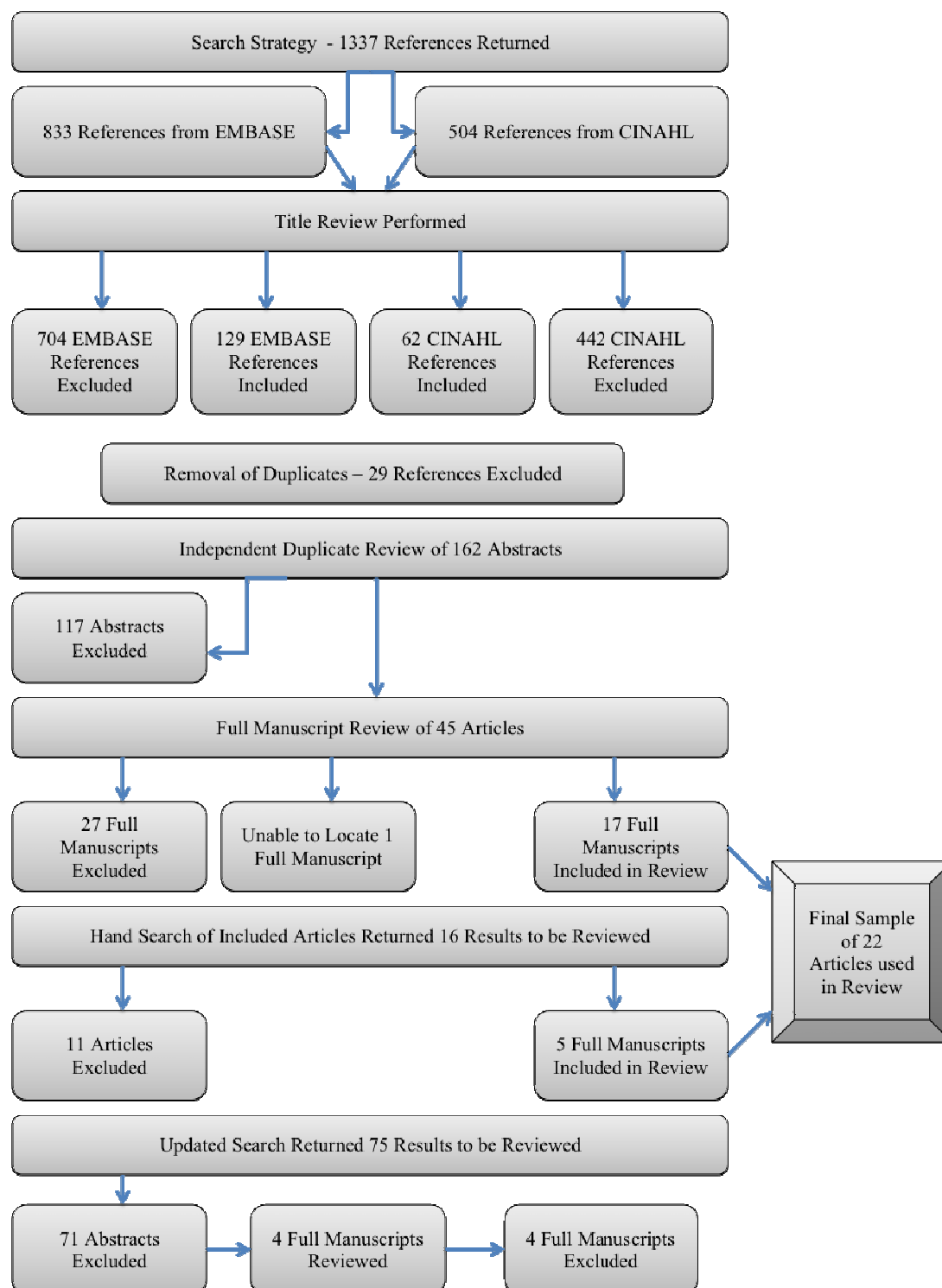
### *Methods*

A search was conducted in the CINAHL and EMBASE databases for relevant articles that assessed psychometric properties of fatigue scales. The search terms “chronic fatigue syndrome”, “muscle fatigue”, and “fatigue” were combined with “instrument validation”, “validation study”, “validation process” and “validation” to generate the initial search results on February 17, 2011 (EMBASE) and February 22, 2011 (CINAHL). The search was updated June 30, 2011 to capture any recent publications. Each search result underwent a title review by me before the abstract review process. During the title search, articles were excluded from the abstract review stage if they identified a non-neurological condition (anything other than PD, MS, ChFS or Postpoliomyelitis) or were not assessing the psychometric properties of a self-report fatigue scale. In cases in which the article title was vague, the article was included in the abstract review. In the next step, two researchers (myself and a research assistant) independently reviewed the abstracts of the remaining search results. Abstracts were included for full manuscript review if they assessed the psychometric properties of a fatigue scale in one of the conditions of interest, the sample included individuals over the age of 18 and the English version of the fatigue scale was used in the study.

A Kappa statistic was used to evaluate the agreement between reviewers over and above what would be due to chance alone (Guyatt, Rennie, Meade & Cook, 2008). A separate Kappa was calculated for the original abstract review and the abstract review as a result of the updated search. Any search result that did not have an abstract was scored as included in the full manuscript review stage by the reviewers. If reviewers disagreed on inclusion or exclusion of the abstract, a meeting was held to discuss and reach consensus to either include or exclude the abstract. I then read the full manuscripts;

articles not published in English and articles that were not primary sources were excluded at this stage, as well as other articles that did not fulfill any of the criteria of earlier review stages due to insufficient detail. Finally, the references of each included manuscript were hand searched for any relevant articles that the initial search did not identify. Figure 3-1 shows a graphic representation of the search and review process including where and when articles were excluded from the study. The final stage of the review included extracting information on reliability and validity from each primary source article. Table 3-1 provides definitions of the terms for validity and reliability that were employed in this review (Portney & Watkins, 2000).

It should be noted that assessing the responsiveness and sensitivity-to-change of the identified fatigue measures was not an objective of this review. The purpose of this review was to identify and assess the validity of available discriminative tools for assessing fatigue and responsiveness is not a necessary attribute for tools with a discriminatory purpose.



**Figure 3-1 - Flowchart of Systematic Review Process**



*Table 3-1- Definitions of Measurement Properties Assessed*

<b>Measurement Property</b>	<b>Definition</b>
Internal Consistency	Internal Consistency is a form of reliability that assesses the degree to which the items of an instrument all measure the same trait (Portney & Watkins, 2000).
Test-retest Reliability	Test-retest reliability is the degree to which an instrument is stable over repeated administrations of the instrument to the same individuals over an identified time interval (Portney & Watkins, 2000).
Construct Validity	Construct validity refers to the ability of an instrument to measure an abstract concept and the degree to which the instrument reflects the theoretical components the concept. Includes convergent and discriminant validity (Portney & Watkins, 2000).
Convergent Validity	Convergent validity indicates that two instruments measuring the same trait will produce similar results or will be highly correlated (Portney & Watkins, 2000).
Divergent Validity	Divergent validity indicates that different results or low correlations would be expected from instruments intended to measure different traits. Measures of different traits should not be highly correlated (Portney & Watkins, 2000).
Known Groups Validity	Known groups validity indicates that a measure can discriminate between individuals who are known to have the trait and those that do not (Portney & Watkins, 2000).

In the data extraction stage, measures were classified into one of three categories: adequate psychometric properties, inadequate psychometric properties and measures needing more validation efforts. To be allocated to the adequate category the measure needed to demonstrate moderate to high reliability (values of coefficients above 0.70) (Portney & Watkins, 2000) in concert with moderate to high correlations with other fatigue measures as evidence of convergent construct validity (values around or above 0.60) (Portney & Watkins, 2000) and evidence of discriminatory ability of the scale (i.e. known groups validity). Measures were classified into the inadequate category if they met any of the following criteria: low reliability (values of coefficients less than 0.70), low or non-significant correlations between the measure and other fatigue measures (values less than 0.50) (Portney & Watkins, 2000), inability to discriminate between

patient groups, inability to fit the assumptions of unidimensionality (for a unidimensional scale), or the only validation consisted of correlations with other health status measures not necessarily related to fatigue. Finally, measures were identified as needing more work if they demonstrated limited evidence of construct validity with other fatigue measures (i.e. only one fatigue measure, or in only one population) but other evidence of validity and reliability was strong (i.e. confirmed factor structure, ability to discriminate between groups). Additionally, the measures that did not have values reported for reliability, but had adequate validity, were identified as needing more evidence before use in a neurological condition.

### *Results*

A Kappa statistic of 0.64 was achieved for the first round of abstract reviews and a value of 0.40 was achieved for the second round of abstract review, suggesting moderate agreement between independent reviewers over and above the agreement that would be due to chance alone. In total, 241 abstracts were reviewed; the independent reviewers disagreed on 32 individual abstracts (approximately 13%), and disagreements were readily reconciled. The review resulted in 15 different self-report fatigue scales that had been employed in at least one of the conditions of interest, and two scales that had been altered to form a new version as a result of Rasch analysis or another test of unidimensionality. Table 3-2 contains a description of the characteristics of each identified scale.

**Table 3-2 - Characteristics of the Identified Scales**

<b>Scale Name</b>	<b>Number of Items</b>	<b>Scoring Range</b>	<b>Maximum Total Score</b>	<b>Scoring Direction (More Fatigue)</b>	<b>Subscales/Dimensions</b>	<b>Attribute Measured</b>	<b>Definition of Fatigue</b>	<b>Timeframe</b>
Fatigue Severity Scale	9	1-7 Likert	63	Higher Scores	None	Severity	None	Past week
Rasch Analyzed Fatigue Severity Scale	5	1-7 Likert	Not Stated	Higher Scores	None	Social Consequences of Fatigue	None	Past week
Fatigue Impact Scale	40	0-4 Likert	160	Higher Scores	Cognitive Functioning, Physical Functioning, Psychosocial Functioning	Impact	None	Past 4 weeks
Fatigue Assessment Instrument	29	1-7 Likert	7	Higher Scores	Severity, Situation Specific, Consequences of Fatigue, Responds to Sleep	Symptomology	Fatigue Defined Explicitly	Past 2 weeks
Multidimensional Fatigue Inventory	20	1-5 Likert	100	Higher Scores	General Fatigue, Physical Fatigue, Reduced Activity, Reduced Motivation, Mental Fatigue	Severity	None	"Lately"
Modified Fatigue Impact Scale	21	0-4 Likert	84	Higher Scores	Cognitive Functioning, Physical Functioning, Psychosocial Functioning	Impact	Fatigue Defined Explicitly	Past 4 weeks
Rasch Analyzed Modified Fatigue Impact Scale	13	0-4 Likert	Not Stated	Higher Scores	Physical Functioning, Cognitive Functioning	Impact	Fatigue Defined Explicitly	Past 4 weeks
Parkinson Fatigue Scale	16	1-5 Likert	5	Higher Scores	None	Impact	Unknown	Past 2 weeks
Chalder Fatigue Scale	14	4-point Likert	Not Stated	Not Stated	Cognitive Difficulties, Tiredness and Sleepiness, Strength and Endurance, Loss of Interest and Motivation	Severity	None	Unknown

Scale Name	Number of Items	Scoring Range	Maximum Total Score	Scoring Direction (More Fatigue)	Subscales/Dimensions	Attribute Measured	Definition of Fatigue	Timeframe
Empirical Fatigue Scale	3	1-6 Likert	18	Higher Scores	None	Not Stated	Unknown	Not Stated
Fatigue Scale for Motor and Cognitive Functions	20	5-point Likert	Not Stated	Not Stated	Cognitive Fatigue, Motor Fatigue	Trait Nature of Fatigue	Fatigue Defined Explicitly	"In General"
MS Specific Fatigue Severity Scale*	6	1-7 Likert	42	Higher Scores	None	Factors Influencing Fatigue	None	Not Stated
Neurological Fatigue Index	39	0-3 Likert	Not Stated	Higher Scores	Physical, Cognitive, Nocturnal Sleep, Diurnal Sleep	Not Stated	Fatigue Defined Explicitly	Past 2 weeks
Piper Fatigue Scale	22	0-10 Likert	22	Higher Scores	Behavioural/Severity, Affective Meaning, Sensory, Cognitive/Mood	Not Stated	Fatigue Defined Explicitly	Past 3 months
Short Fatigue Questionnaire	4	1-7 Likert	7	Not Stated	Not Stated	Not Stated	Unknown	Not Stated
Swedish Occupational Fatigue Inventory	20	0-6 Likert	Not Stated	Higher Scores	Lack of Energy, Physical Exertion, Physical Discomfort, Lack of Motivation, Sleepiness	Not Stated	None	Past 6 months
Unidimensional Fatigue Impact Scale	22	0-3 Likert	Not Stated	Higher Scores	None	Impact	Unknown	Past week

\*MS=Multiple Sclerosis

The Fatigue Severity Scale (FSS) was assessed most frequently with eight studies reporting psychometric data for the scale in three of the conditions of interest. Many studies reported moderate to high levels of internal consistency (range  $\alpha=0.80-0.95$ ) (Burger, Franchignoni, Puzic & Giordano, 2010; Grace, Mendelsohn & Friedman, 2007; Horemans, Nollet, Beelen & Lankhorst, 2004; Krupp, LaRocca, Muir-Nash & Steinberg, 1989) and two studies reported high levels of test-retest reliability (range intraclass correlation coefficient (ICC)=0.83-0.84) (Horemans et al. 2004; Krupp et al., 1989). Two studies applied Rasch analysis to the FSS (Burger et al., 2010; Mills, Young, Nicholas, Pallant & Tennant, 2009) (resulting in the FSS-5 and the FSS-8) and a third study assessed the unidimensionality of the FSS (Horemans et al., 2004) and determined two items should be removed from the FSS. The results of these three studies concluded that the 9-item original FSS is not a unidimensional scale. In addition, one group of authors (Burger et al., 2010) concluded that a 1 to 7 rating scale was inappropriate for the scale and reduced the scoring to a 1 to 3 scale. The FSS in general demonstrated moderate evidence of convergent validity in neurological conditions with significant but varying strength of correlations with visual analog scales (VAS) and multi-item scales used to measure fatigue (range 0.38-0.84) (Grace et al., 2007; Horemans et al., 2004; Burger et al., 2010; Vasconcelos, Prokhorenko, Kelley, Vo, Olsen, Dalakas et al., 2006; Flachenecker, Kumpfel, Gottschalk, Grauer, Rieckmann, Trenkwalder et al., 2002). The FSS also demonstrated significant moderate strength correlations with other measures of health status (range 0.50-0.74) (Horemans et al., 2004; Burger et al., 2010). One study demonstrated evidence of divergent validity of the FSS, such that fatigue was a separate construct from depression and excessive sleepiness as the result of non-significant low correlations with scales measuring these conditions (Vasconcelos et al., 2006). Several studies demonstrated the ability of the FSS to discriminate between patients in regards to fatigue. One study demonstrated the FSS was able to discriminate patients with disabling fatigue from those without (Vasconcelos et al., 2006). A second study demonstrated the ability of the FSS to discriminate patients with MS who experienced fatigue from those individuals with MS who did not experience fatigue and established a cut off value for determining fatigue of 4.6 on the FSS (Flachenecker et al., 2002). Finally, a third study

showed the FSS to be able to discriminate between individuals with MS and those without (Chipchase, Lincoln & Radford, 2003).

The Fatigue Impact Scale (FIS) was shown to have a high level of internal consistency ( $\alpha=0.98$ ) in one study (Fisk, Ritvo, Ross, Haase, Marrie & Schlech, 1994) and acceptable levels of test-retest reliability for the total score and subscale scores in another study (range ICC=0.68-0.76) (Mathiowetz, 2003). One study assessed the convergent validity of the FIS with other fatigue scales and demonstrated non-significant low correlations between the FIS and the FSS, as well as between the FIS and a VAS for fatigue (Vasconcelos et al., 2006). An additional study demonstrated a significant but low correlation between the FIS and the FSS ( $r=0.44$ ) (Mathiowetz, 2003). The FIS showed weak evidence of convergent validity with other health status measures; significant moderate correlations existed between the FIS and subscales of the Short Form-36 (range  $r=-0.54$  to  $-0.62$ ) (Mathiowetz, 2003). In addition, the FIS correlated moderately with the Sickness Impact Profile (range  $r=0.53-0.57$ ) (Fisk et al., 1994). One study revealed evidence of divergent validity of the FIS with non-significant low correlations between the FIS and measures of depression and excessive sleepiness (Vasconcelos et al., 2006). Finally, three studies attempted to establish known groups validity for the FIS; one study demonstrated the FIS was unable to detect differences between individuals with disabling fatigue and those without (Vasconcelos et al., 2006). The remaining two studies did show the evidence of the discriminatory ability of the FIS. Specifically, Fisk and colleagues (1994) demonstrated the FIS was able to discriminate between groups based on diagnosis and levels of fatigue associated with the diagnosis (ChFS, MS, and Hypertension). Chipchase and associates (2003) showed that the cognitive, physical and social dimensions of the FIS were able to discriminate between individuals who had MS and those who did not.

The psychometric properties of the Fatigue Assessment Inventory (FAI) have been assessed in a sample of individuals with MS and ChFS. The internal consistency of the FAI ranged from  $\alpha=0.70$  to  $0.92$  depending on the subscale and test-retest reliability values for the FAI ranged from  $r=0.29$  to  $0.69$  (Schwartz, Jandorf & Krupp, 1993). The psychological consequences and severity subscales of the FAI correlated moderately with the Rand Vitality Index ( $r=-0.41$  and  $r=-0.72$  respectively) providing some evidence of

convergent validity (Schwartz et al., 1993). Results from a factor analysis confirmed an underlying structure of four factors (Schwartz et al., 1993). In addition, two studies have assessed the known groups validity of the FAI; the scale was able to discriminate between individuals with chronic fatiguing syndromes from healthy individuals (Schwartz et al., 1993) and the severity subscale was able to discriminate between individuals with MS and those without (Chipchase et al., 2003).

The Multidimensional Fatigue Inventory (MFI-20) has been demonstrated to have high internal consistency in neurological samples (range  $\alpha=0.71$  to  $0.93$  for the total scale and individual subscales) (Lin, Brimmer, Maloney, Nyarko, BeLue & Reeves, 2009; Smets, Garssen, Bonke & De Haes, 1995). Test-retest reliability of the MFI-20 in a neurological sample has not been reported. One study has assessed the convergent validity of the MFI-20 with other health status measures and demonstrated moderate to high significant correlations (range  $r=0.50$  to  $-0.81$ ) (Lin et al., 2009). Convergent validity of the MFI-20 with other fatigue scales has not been established in a neurological sample. The discriminatory ability of the MFI-20 has been assessed; in one study, the MFI-20 was able to discriminate between individuals with ChFS and those who were considered chronically unwell or well (Lin et al., 2009). A second study demonstrated that the MFI-20 was able to discriminate between individuals who were chronically fatigued and those who were not (Smets et al., 1995). Finally, Smets and colleagues (1995) assessed the factor structure of the MFI-20 and confirmed a five-factor model, however they also tested two four factor models (combining general and physical fatigue or removing the general fatigue factor completely) and found an acceptable fit for the four factor models. A second group assessed the factor structure of the MFI-20 and had evidence to support the five-factor model. However, the authors noted that some factors within the scale were highly correlated and that several items would have loaded on more than one factor if they had not constrained the paths in the analysis (Lin et al., 2009).

The Modified Fatigue Impact Scale (MFIS) has been shown to have low correlations with other measures of fatigue (range  $r=0.47-0.56$ ) providing weak evidence of the convergent validity of the scale (Flachenecker et al., 2002). Additionally, the MFIS was able to discriminate between MS patients with and without fatigue, and a cut-off score of 38 on the MFIS has been established for determining fatigue in a sample with

MS (Flachenecker et al., 2002). However, one study tested the unidimensionality of the MFIS and demonstrated that the 21-item scale did not fit the Rasch model; as a result, the authors suggested removing three items from the physical subscale and five items from the cognitive subscale to improve the measure (Mills, Young, Pallant & Tennant, 2010a). There is currently no information available about the reliability of the MFIS in a neurological population.

The Parkinson Fatigue Scale (PFS) was shown to have high internal consistency values (range  $\alpha=0.97-0.98$ ) (Grace et al., 2007; Brown, Dittner, Findley & Wessely, 2005) and moderate levels of test-retest reliability (Spearman correlations between 0.52 and 0.72) (Brown et al., 2005). Two studies established moderate to high correlations between the PFS and other fatigue measures (range  $r=0.71-0.84$ ) (Grace et al., 2007; Brown et al., 2005). Evidence of the PFS's discriminatory ability was demonstrated by Brown and colleagues (2005), such that the PFS was able to discriminate between individuals who considered themselves to have fatigue and those who did not. Furthermore, within the fatigued group, the PFS was also able to discriminate between individuals who considered fatigue to be a problem and those for whom fatigue was not a problem (Brown et al., 2005). A confirmatory factor analysis performed on the PFS revealed a single factor that explained approximately 64% of the scale variance.

Morriss and associates (1998) evaluated the validity of the Chalder Fatigue Scale (CFS) in a sample with ChFS. The authors performed a factor analysis and identified four factors representing subscales of the CFS. Evidence of divergent validity of the subscales of the CFS comes from low to moderate correlations with other measures of health status including depression, function work capacity and grip strength (range  $r=0.22-0.69$ ) (Morriss, Wearden & Mullis, 1998); however, test-retest reliability has not yet been reported and evidence convergent validity with other fatigue measures is needed.

The Empirical Fatigue Scale (EFS) has been shown to be reliable in a population with ChFS (test-retest reliability  $r=0.87$  and  $r=0.91$ ) (Bailes, Libman, Baltzan, Amsel, Schondorf & Fichten, 2006); however, internal consistency has not been assessed. Evidence of convergent validity is limited to a low negative correlation with handgrip strength ( $r=-0.33$ ). Bailes and colleagues (2006) did provide evidence that fatigue and sleepiness were separate constructs as evidenced by a low negative correlation with the



multiple sleep latency test ( $r=-0.40$ ). The EFS was able to discriminate individuals with ChFS from those with narcolepsy or no health concerns (Bailes et al., 2006).

The Fatigue Scale for Motor and Cognitive Functions (FSMC) has been shown to be highly reliable in a population with MS with an internal consistency value that ranged from 0.91 to 0.95 and test-retest values over 0.85 (range  $r=0.85-0.87$ ) (Penner, Raselli, Stocklin, Opwis, Kappos & Calabrese, 2009). In addition, Penner and associates (2009) provided evidence of the convergent validity of the FSMC including significant strong correlations between the FSMC and other multi-item fatigue measures (range  $r=0.80-0.83$ ) as well as a moderate correlation with fatigue as assessed by neurologists ( $r=0.51$ ). Divergent validity of the FSMC was established by low to moderate correlations with measures of depression (range  $r=0.24-0.49$ ), indicating that fatigue and depression were separate constructs (Penner et al., 2009). Finally, the FSMC was able to discriminate between individuals with a diagnosis of MS and those without (Penner et al., 2009).

Flachenecker and colleagues (2002) have shown limited evidence of validity for the MS Specific Fatigue Severity Scale (MFSS) through a low correlation with the FSS ( $r=0.44$ ). However, the MFSS was not significantly correlated with a VAS for fatigue and only demonstrated a very low correlation with the MFIS (Flachenecker et al., 2002). The MFSS was able to discriminate between people with MS-related fatigue and those not experiencing fatigue. However, the FSS and the MFIS were better at discriminating between people with MS who experienced fatigue and those who did not when compared to the MFSS (Flachenecker et al., 2002). The reliability of the MFSS has not been assessed in a study within the parameters of this review.

The Neurological Fatigue Index (NFI) was demonstrated to have a high level of stability with test-retest reliability values above 0.79 for all subscales (range  $r=0.79-0.86$ ) (Mills, Young, Pallant & Tennant, 2010b). Evidence of the NFI's convergent validity has been reported as moderate correlations between the NFI other multi-item fatigue scales as well as a VAS for fatigue (range  $r=0.58-0.71$ ) (Mills et al., 2010b). In addition, each subscale of the NFI met the requirements of unidimensionality and fit the assumptions of the Rasch model providing further evidence of validity of the NFI (Mills et al. 2010b).

The Piper Fatigue Scale has been shown to have excellent reliability in a population with postpoliomyelitis with high values for both internal consistency ( $\alpha=0.98$ )

and test-retest reliability (ICC=0.98) (Strohschein, Kelly, Clarke, Westbury, Shuaib & Chan, 2003). The Piper Fatigue Scale also correlated highly with the CFS ( $r=0.80$ ), weak evidence of convergent validity, as the validity of the CFS has not been established (Strohschein et al., 2003). The Piper Fatigue Scale scores were significantly different between individuals with postpoliomyelitis and individuals without a neurological condition, providing evidence of known groups validity (Strohschein et al., 2003). Further evidence of construct validity is needed before the use of the Piper Fatigue Scale in a neurological condition.

The Short Fatigue Questionnaire (SFQ) has been shown to have high levels of reliability in a sample of individuals with postpoliomyelitis (Horemans et al., 2004). Internal consistency values ranged from  $\alpha=0.77$  to  $0.79$  and test-retest reliability values of ICC=0.73-0.90 established acceptable stability of the scale. Horemans and colleagues (2004) have established the convergent validity of the SFQ by demonstrating moderate significant correlations with other health status measures (range  $r=0.67$ - $0.68$ ) and the FSS ( $r=0.47$ ); however, further evidence of convergent validity with other multi-item fatigue measures would be beneficial.

Johansson and associates (2008) assessed the psychometric properties of the Swedish Occupational Fatigue Inventory (SOFI) and demonstrated moderate to high levels of internal consistency, depending on the subscale (range  $\alpha=0.68$ - $0.92$ ). Other than a moderate correlation between the Lack of Energy Subscale and the FSS (range  $r=0.53$ - $0.61$ ) across the three time points, the correlations between the SOFI and the FSS were low. In addition, a factor analysis demonstrated a five factor model with some items cross loading on more than one factor suggesting indistinct belonging (Johansson, Ytterberg, Back, Homqvist & von Koch, 2008). Test-retest reliability of the SOFI has not been assessed in a neurological sample.

The Unidimensional Fatigue Impact Scale (UFIS) has been shown to have excellent reliability both in terms of internal consistency ( $\alpha=0.96$ ) and test-retest reliability ( $r=0.86$ ) (Meads, Doward, McKenna, Fisk, Twiss & Eckert, 2009). The 22-item UFIS is truly a unidimensional measure of fatigue, demonstrating fit to the assumptions of the Rasch model (Meads et al., 2009). Evidence of convergent validity of the UFIS was demonstrated by moderate correlations with other health status measures

(range  $r=0.47-0.64$ ) and by moderate correlations with anxiety and depression (range  $r=0.46-0.60$ ) (Meads et al., 2009). No evidence of convergent validity with other fatigue measures has been reported for the UFIS in a neurological sample. The UFIS was shown to discriminate between individuals with different levels of self-perceived severity of MS and general health. As well the UFIS demonstrated the ability to discriminate between individuals experiencing a flare-up of symptoms related to MS and those who were not (Meads et al., 2009).

A supplementary file (Appendix 3-A) provides detailed information about reliability and validity obtained from the studies examined in the review. Each measure was classified into one of three categories based on the reported psychometric properties in a neurological condition. Table 3-3 contains the information regarding the specific rationale for allocation of each measure to a specific category.

*Table 3-3 - Categorization and Rationale for Each Fatigue Measure*

<b>Measure</b>	<b>Rationale</b>
<b>Adequate Psychometric Properties</b>	
Parkinson Fatigue Scale	Moderate to high reliability and validity. Ability to discriminate between fatigued groups. Confirmatory factor analysis verified a singular construct and appropriate use of a summary score.
Fatigue Scale for Motor and Cognitive Functions	Evidence of high reliability and convergent validity. Evidence of divergent validity and known groups validity.
Neurological Fatigue Index	Evidence of moderate levels of construct validity and moderate test-retest reliability. Each subscale fit the assumptions of unidimensionality.
<b>Inadequate Psychometric Properties</b>	
Fatigue Severity Scale	Did not meet the assumptions of unidimensionality, therefore the use of a summary score is inappropriate. In addition, the scaling of the measure was called into question.
Fatigue Impact Scale	Non-significant low correlations with other fatigue measures. Conflicting reports about discriminatory ability of the measure.
Modified Fatigue Impact Scale	Inability to meet the assumptions of Rasch analysis. Weak evidence of convergent validity with other fatigue measures.
Empirical Fatigue Scale	No evidence of convergent validity with other fatigue measures. The scale contains a limited number of items and has only been correlated with handgrip strength.
Multiple Sclerosis Specific Fatigue Severity Scale	Low or non-significant correlations with other fatigue measures and no evidence of reliability reported.
Swedish Occupational Fatigue Inventory	Low correlations with other fatigue measures and factor analysis demonstrated evidence of items loading on multiple factors.
Fatigue Assessment Inventory	Poor test-retest reliability of the subscales and lack of evidence of convergent validity with other fatigue measures.
<b>Needs More Validation</b>	
Multidimensional Fatigue Inventory	High levels of internal consistency, and evidence of discriminatory ability of the scale. Five-factor structure of the scale confirmed, however, two separate four-factor models also achieved acceptable fit, more work is needed to determine the factor structure of the scale. In addition, information about the test-retest reliability of the scale and convergent validity with other fatigue measures is needed.
Unidimensional Fatigue Impact Scale	Evidence of high reliability values and discriminatory ability of the scale. Information on the convergent validity with other fatigue measures is needed.

Measure	Rationale
Chalder Fatigue Scale	Some evidence of validity demonstrated by low to moderate correlations with health status measures and a four-factor structure has been confirmed. No evidence of the reliability of the measure or convergent validity with other fatigue scales.
Short Fatigue Questionnaire	Evidence of high reliability and moderate correlations with health status measures. Low to moderate correlation with the fatigue severity scale requires further investigation.
Piper Fatigue Scale	Evidence of high reliability and discriminatory ability of the scale. Only evidence of convergent validity is with the Chalder Fatigue Scale, which has not been correlated with other fatigue measures.

### *Discussion*

Fifteen fatigue measures were reviewed in detail and categorized into three groups: adequate psychometric data, inadequate psychometric data and scales requiring further investigation. Three scales were considered acceptable for use and all were designed for a specific neurological condition. Seven of the identified scales were deemed inadequate for use in neurological conditions for various reasons including insufficient reliability or convergent validity. Many scales showed some degree of evidence to support their use in a neurological condition; one or more important element was missing leading to the suggestion that further validation efforts are necessary.

The PFS, FSMC and the NFI were the only fatigue measures that had satisfactory data supporting their use in a neurological condition. These three scales had adequate reliability, convergent validity with other scales measuring fatigue and evidence of the ability of the scale to discriminate between groups based on fatigue. Additionally, a confirmatory factor analysis verified that the PFS was measuring a single construct and the use of a summary score was supported and the NFI met the assumptions of unidimensionality for each subscale. These scales were developed for a specific neurological population and have not been assessed outside the condition of interest; therefore, prior to being used in another condition validation would be required.

The FSS, FIS, MFIS, EFS, MFSS, SOFI and FAI were all considered to be inadequate for use in a sample of people with a neurological condition. The FSS demonstrated satisfactory convergent validity with varying strength of significant correlations with other measures of fatigue and health status; however, the original 9-item

FSS did not withstand tests of unidimensionality, illustrating that calculating a summary score of the nine items is inappropriate in neurological conditions. Also, the validity of the 7-point scoring was been called into question in one study (Burger et al., 2010). Although the FIS demonstrated excellent internal consistency and acceptable test-retest reliability, it has not been shown to have adequate convergent validity with other fatigue measures, only with related health status measures. In addition, studies have shown conflicting results in regards to the discriminatory ability of the FIS. The MFIS demonstrated low correlations with other measures of fatigue and did not fit the Rasch model test of unidimensionality indicating that the measure may be compromised by items that do not reflect the construct of fatigue.

The EFS was shown to be reliable between assessments; however, the evidence of convergent validity was inadequate. The authors chose to correlate the fatigue scale solely with handgrip strength (Bailes et al., 2006) and as a result the validity of the EFS as a fatigue measure remains to be demonstrated. Additionally the limited number of items contained in the EFS limits the information that can be obtained from a single assessment. The MFSS correlated weakly with other fatigue and did not perform as well as other fatigue tools previously used in a sample of people with MS. Additionally, no information was available about the reliability of this scale in a neurological population. The SOFI was shown to have inadequate validity as a result of low correlations between the SOFI subscales and the FSS. Although the internal consistency of the FAI was moderate, test-retest reliability values of individual FAI subscales were low to moderate suggesting instability of the scale between assessments. Additionally, evidence of the convergent validity of the FAI has only been established based on relationships with other health status measures.

Five of the identified scales had some evidence of validity for use in a neurological condition; however, more work is necessary to strengthen the results or determine other psychometric properties of the measure. Evidence of convergent validity of the MFI-20 with other health status measures such as the Short Form-36 has been established. The MFI-20 has displayed the ability to discriminate based on patient groups or diagnosis related to fatigue. The factor structure of the five-dimension scale was confirmed by Smets and colleagues (1995), however, two separate four factor models

also achieved acceptable fit and suggest that the dimension of general fatigue may not be a necessary factor within the scale. A second study also generated doubt in the factor structure of the MFI-20 through indistinct loadings of several items (Lin et al., 2009). Future studies should report the internal consistency value obtained for the MFI-20 and relationships with other multi-item fatigue measures to provide further evidence of reliability and convergent validity.

Horemans and colleagues (2004) demonstrated adequate reliability and moderate convergent validity for the SFQ. Additionally, the Piper Fatigue Scale has been shown to be very reliable in a sample of individuals with postpoliomyelitis and some evidence of convergent validity has been provided by Strohschein and associates (2003). Future work demonstrating relationships between both the SFQ and the Piper Fatigue Scale with other multi-item measures of fatigue would provide verification of the construct validity of these measures.

A factor analysis revealed a four-factor solution to the CFS, this along with low to moderate correlations with other health status measures provide some evidence of the validity of the CFS. Further information about the performance of the fatigue scale is needed before recommending its use in a population with a neurological condition, specifically information about the reliability and relationships with other established fatigue scales.

The evidence of the reliability of the UFIS was sound; however, convergent validity of the UFIS was only established with other measures of health status. Future studies should demonstrate the relationship of the UFIS to other fatigue measures.

It should be noted that several of the studies included in this review used a Pearson's correlation and one study used a Spearman's correlation to assess test-retest reliability. The use of an ICC accounts for the agreement between the scores obtained, whereas correlations only provide information about how the scores vary together and do not account for potential systematic errors in the measurement of fatigue (Portney & Watkins, 2000). In a clinical population, test-retest reliability is important for clinicians to know they are obtaining stable estimates to have confidence in the measurement of fatigue. In this review, categorization of measures was not based on the statistics used; however, studies that reported test-retest reliability using a Pearson's or Spearman's

correlation should be interpreted with caution. The use of 95% confidence intervals around estimates of reliability and validity were scarce in this investigation. Future validation efforts should report the confidence intervals around the point estimate to provide users with more accurate information about the performance of an individual scale.

Many of the scales reviewed here neither included a definition of “fatigue” nor identified the specific attribute of interest that the measure was capturing in regards to fatigue (i.e. impact or severity of fatigue). Without a definition of fatigue the results obtained from a measure are difficult to analyze and interpret. Also missing from many studies was a specific purpose for the scale (discrimination, prediction or evaluation), and because the intended use of a scale dictates the types of validity most important to the scale this is an identified weakness for this area. Future work should consider defining fatigue, the attribute of fatigue that is to be measured and be explicit about the intended use of the scale to allow potential users to choose the appropriate measure for their purposes. It should be noted that evidence of sensitivity-to-change or responsiveness of these measures was scarce, and since the primary interest for identifying psychometric properties of the available scales was related use for discriminative and predictive purposes, information regarding sensitivity to change or responsiveness is not included in this report. Finally, the timeframe of each measure varied considerably (for example “during the last four weeks” or “the past two weeks”), and potential users should consider the timeframe when choosing a scale for use.

The fatigue scales reviewed in this report have varying levels of evidence to support their use various populations with disorders of the nervous system. From this review, it appears that several of the diagnosis-specific fatigue scales have strong psychometric properties (Brown et al., 2005; Penner et al., 2009; Mills et al. 2010b). However, it is unknown whether a diagnosis-specific scale would perform similarly in a sample with a neurological condition other than what it was designed for (but with similar fatigue profiles, such as CP); therefore, a scale should be validated within the population of interest before it is employed. However, as all three scales that demonstrated adequate psychometric properties were created with a specific clinical population in mind, the relevance of the items to the fatigue experience of individuals



with CP may be limited. This review highlights the advantages of a diagnosis-specific fatigue scale and it may be beneficial to create a fatigue scale tailored to the specific experience of individuals with CP.

#### *Study Limitations*

The search strategy and key search terms employed in this study returned a large number of irrelevant studies for the title and abstract review stages. Further, as a result of the high exclusion rate, the Kappa statistic employed, as evidence of agreement between reviewers, was not as robust as the authors anticipated (0.64 and 0.40 for the initial and updated abstract reviews respectively). As a distribution becomes more extreme there is a higher level of chance agreement, which makes obtaining high agreement above chance difficult (Guyatt et al., 2008). This was especially the case in the updated search abstract review as many more of the abstracts were excluded than included. However, the reviewers agreed on 87% of the abstracts in this review and all disagreements were discussed until a consensus was achieved between the reviewers to ensure that any relevant article was included in the study.

#### *Conclusion*

This review highlights three diagnosis-specific fatigue scales (one for PD and two for MS) with adequate psychometric data for use in clinical practice or research with individuals with a neurological condition. Seven fatigue scales were considered inadequate for use and five scales require further validation before a recommendation for use could be made. Currently, potential users of fatigue scales should consider using the PFS if interested in measuring fatigue in individuals with PD, and the FSMC or the NFI if interested in measuring fatigue in individuals with MS. Further validation efforts are needed before using any of the fatigue measures identified by this review for individuals with ChFS or Postpoliomyelitis.

#### *Relevance to Thesis*

It appears that no single fatigue scale will be best employed in a population with CP; however, this review has provided a starting point for the creation of a new scale from items and scoring similar to those included in scales with established validity and reliability in neurological conditions similar to CP in the experience of fatigue. Validation of a newly created scale for use in CP will require a definition of fatigue,

specification of the attributes of fatigue, and the timeframe of response, assessment of internal consistency, test-retest reliability, as well as an assessment of construct validity related to the purpose of the measure.

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#### **Chapter 4: Understanding the Bodily Experience of Cerebral Palsy: A Phenomenology**

(An abbreviated version of this paper is published as Brunton, L. K., & Bartlett, D. J. (2013). Understanding the bodily experience of cerebral palsy: A journey to self-awareness. *Disability and Rehabilitation*, 35, 1981-1990).

##### *Introduction*

Rehabilitation practitioners and caregivers have described a great deal of variability in the motor abilities of children and adolescents with cerebral palsy (CP). This variability can be attributed to many reasons such as stress, illness or anxiety; it can also be ascribed to fatigue that occurs during activities of daily living including long distance ambulation or during periods of prolonged standing (Bjornson, Graubert, McLaughlin, Kerfeld, & Clark, 1998).

In a recent study, fatigue, pain and joint deformities were identified as the top three CP-related impairments in adulthood that can interfere with activities of daily life (Hilberink, Roebroek, Nieuwstraten, Jalink, Verheijden & Stam, 2007). Furthermore there are some reports that fatigue may be a contributor to early loss of functional abilities in adolescents and young adults with CP (Houlihan, 2009). Although it is known that adults with CP experience more fatigue than their peers without disabilities, fatigue remains largely unexplored in a population with CP, because there are technical issues with spasticity and contractures interfering with testing positions and data collection procedures, as well as a lack of focus on functional tasks (Brunton & Rice, 2012). Pain however, has been studied fairly extensively in the adult CP population and some literature is available on adolescents' pain experience. A high prevalence of chronic pain has been reported among children and adolescents (Doralp & Bartlett, 2010; Engel, Petrina, Dudgeon & McKearnan, 2005). Pain interferes with sleep, mobility and physical activities of daily living. It has also been suggested that there were greater impacts of pain when the adolescents were up all day without rest, which could reflect fatigue that is exacerbating pain (Engel et al., 2005). Although there are quantitative data regarding prevalence of pain and identification of painful sites, there is a dearth of information on the experience of pain and how adolescents understand their pain. One study has examined how adults cope with pain related to CP (Engel, Jensen & Schwartz, 2006);

however, there is a need to understand how adolescents understand and experience pain in their everyday lives.

Furthermore, very little information is available on how people with CP experience their bodies. One study has explored the lived body in adults with CP and found trends of a *dys-appearing body*, a *not-appearing body* as well as feelings of being different and having restricted autonomy, among other themes (Sandstrom, 2007). Jones (2009) wrote a personal reflection about aging with CP and highlighted the need to understand the lived experience of aging with CP and focus medical care on preventing secondary conditions of fatigue and pain. Another study was conducted to explore the experience of mobility in adolescents with CP (Palisano, Shimmell, Stewart, Lawless, Rosenbaum & Russell, 2009), yet there is no information available on how adolescents or young adults with CP experience their bodies. However, the literature suggests that low levels of physical activity may adversely affect adolescents living with CP as a result of higher energy costs (Maltais, Pierrynowski, Galea & Bar-Or, 2005), potentially leading to more fatigue experienced by these individuals. Therefore, it is important to understand how these factors may shape the way adolescents and young adults experience their bodies in their everyday world.

This is the first study to explore the experience of living with CP and perceptions about muscle fatigue and pain directly from adolescents and young adults with CP. Kembhavi and colleagues' (2011) performed a mapping review of outcomes studied in adults with CP and noted that the study of fatigue in CP has really only emerged within the last 10 years. At the time of the review there were no studies that explored the impact of fatigue on the activity and participation level variables for these adults. Therefore, the purpose of this study was to have a client-centered account of fatigue and pain that is experienced by adolescents and young adults living with CP using phenomenology (van Manen, 1997). This stage is particularly important to understand the experience of fatigue, and its relation to pain and physical activity, as a result of growth of the individual and negotiation of new body management techniques and to identify factors that shape the experience.

### *Methodology*

The aim of the study is to describe, in detail, the lived experience and the essence of the lived body in CP. Phenomenology is useful for studying the essence of an experience and understanding how meaning is lived (van Manen, 1997). The goal of a phenomenological study is to create an evocative text comprised of behaviours, actions and meanings in the lifeworld, to understand the phenomena of interest (van Manen, 1997). van Manen (1997), writes about objectivity of the researcher as remaining completely focused and true to the object and recognizes that objectivity and subjectivity are not mutually exclusive. Subjectivity is the attention to being perceptive and astute in order to capture the full depth of the object or experience (van Manen, 1997). For the purposes of this study bracketing (a method by where the author acknowledges and sets aside their preconceived notions) was not used, rather I acknowledge that my preconceived notions and knowledge about CP have contributed to the findings of the study, particularly in the interpretation of the results. Phenomenology can be either descriptive or description that is interpretive; van Manen (1997) posits that the description can never be separated from interpretation and that everything is interpreted from the questions participants are asked to how their answers are understood. As a result, I acknowledge that the methods employed in this study are interpretative in nature.

### *Methods*

#### *Participants*

Ethical approval was obtained from the Ethics Review Board at Western University (Appendix 4-A) and the Research Advisory Council at Thames Valley Children's Centre (Appendix 4-B). Adolescents and young adults aged 14 to 25 years who were classified as levels I to III on the Gross Motor Function Classification System (GMFCS) (Palisano, Rosenbaum, Bartlett & Livingston, 2008) and as level I or II on the Communication Function Classification System (CFCS) (Hidecker, Paneth, Rosenbaum, Kent, Lillie, Eulenberg et al., 2011) were invited to participate from two children's treatment centres in southwestern Ontario. Each participant reviewed a letter of information and provided signed consent (Appendix 4-C). Additionally, advertisements were placed in the newspaper at Western University and in fitness centres around the city



of London, Ontario. Finally, snowball sampling was used to recruit interested adolescents and young adults through previous participants. Descriptive information for each participant, including their chosen pseudonyms, age, gender, employment or educational status, GMFCS and CFCS level can be found in Table 4-1.

**Table 4-1 - Individual Participant Characteristics**

Participant	Distribution of Involvement	GMFCS Level	CFCS Level	Sex	Educational/ Employment Status	Age
Wade Wilson	Diplegia	II	I	Male	Full-time Student	20
Chris Bosh	Diplegia	II	I	Male	Full-time Student	16
Goofy	Diplegia	II	I	Male	Full-time Student	24
Ariel	Quadriplegia	II	I	Female	Unemployed	20
Dori	Triplegia	II	I	Female	Full-time Student	19
Travis	Diplegia	II	I	Male	Full-time Student	19
Nemo	Diplegia	III	I	Male	Full-time Student	19
Hillary Duff	Diplegia	III	II	Female	Full-time Student	15
Jasmine	Diplegia	III	I	Female	Full-time Student	21
Mickey Mouse	Diplegia	I	I	Male	Full-time Student	19

#### *Data Collection Method*

One semi-structured interview was conducted with each participant in the study. The interviews were approximately one hour in length and conducted in person by me at a location deemed convenient for the participant. During the interview participants were asked to describe a typical day, a day in which they experienced fatigue and/or pain and more general questions about living in a body with CP (a copy of the interview guide can be found in Appendix 4-D). Probing was used to get a deeper understanding of these concepts during the interview.

#### *Data Collection Procedures*

Demographic information (as described earlier) was collected by self-report. Interviews were audiotaped and transcribed verbatim. The researcher also used field notes to identify areas to probe for further information during the interviews to ensure participants' stories were fully completed and explored to the extent the individual wished to share.

### *Analysis*

There is no commonly agreed upon method underlying phenomenology; however, van Manen (1997) describes six methodological themes or activities to follow. van Manen's approach to phenomenology was chosen as the methodological framework for this study to ensure the results would be clinically relevant to clinicians and care providers who work with individuals with CP and as a result of his perspectives on objectivity, subjectivity and interpretation. Specifically, van Manen recognizes that objectivity and subjectivity are not mutually exclusive and that the description can never be separated from interpretation (van Manen, 1997). As a result, the author acknowledges that the methods employed in this study are interpretative in nature and that my preconceived notions and knowledge about CP have contributed to the findings of the study.

Specifically, this study employed a hermeneutical approach to data analysis in trying to understand the description and interpretation provided by the participants. Line-by-line coding was performed in the NVivo 9 (QSR International, 2011) computer software program to identify any relevant text about the lived body within each transcript. Subsequently, a thematic analysis approach was used, in that elements that were continually apparent in the text were examined and interpreted as themes that allowed the researcher to understand the meanings contained in those themes (van Manen, 1997). The researcher returned to the field notes taken during the interviews at the time of analysis to reflect on and add context to each interview, as the analysis was performed after all 10 interviews were completed. Aspects of the interview that the participants put particular emphasis on (either by repetition of the topic or through body language and/or intonation documented in the researchers field notes) were considered to be meaningful to the participant and captured as a code related to their lived bodies. Within each transcript the researcher generated unique codes related to their experiences. The researcher then began to combine codes and look for recurrence of similar codes across participants. Codes that related to similar experiences of the lived body across participants were then aggregated into larger themes. Themes were then graphically depicted to examine the inter-relationships between themes related to the lived body.

The analysis was guided by the 6 essential research activities described by van Manen (1997) to ensure the researcher remained committed to the text and that the analysis was a reflection of the important aspects of the phenomena of interest. The first essential activity is to commit to the phenomena of interest and understanding that the result of the phenomenology is one single interpretation (van Manen, 1997). The second activity is to investigate the experience as it is lived and remaining attuned to the lived experience and in the case of this study, the lived body (van Manen, 1997). The third activity is identified as reflecting on the essential themes, which really speaks to understanding the special significance of each theme that emerges from the data (van Manen, 1997). The fourth essential activity is the art of writing and rewriting and understanding that language and thinking are difficult to separate and that phenomenology is the art of applying language to a phenomenon (van Manen, 1997). The fifth idea that van Manen posits is maintaining a strong and oriented relation. This relates to the researcher remaining completely committed and attuned to the original notion and fundamental question being researched (van Manen, 1997). The final activity is balancing the research context by considering parts and the whole; this is where the idiographic and nomethic elements come into action (idiographic analysis involves looking at individual's own meanings alone and nomethic analysis looks for convergence of meaning across all participants). One must constantly step back from the parts and understand the whole context and understand how the parts relate to each other (van Manen, 1997). During the analysis the researcher continually shifted focus by examining each individual participant's own meanings and looking for convergence of meaning across all participants.

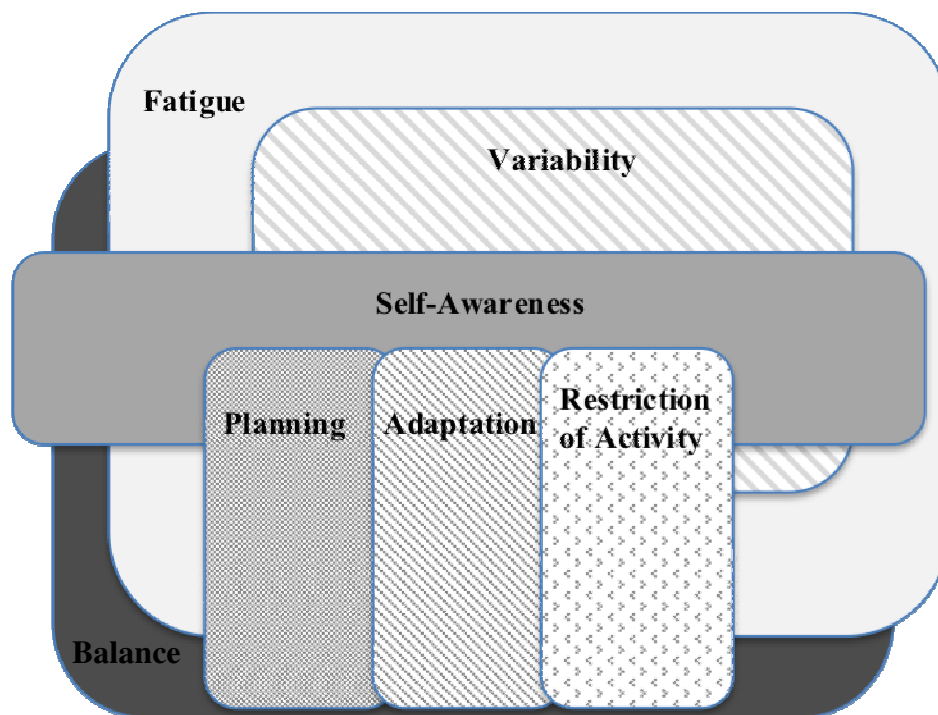
A second researcher (DJB) carried out an independent coding and thematic analysis to confirm the relevance of the identified themes. Participants were provided a written summary; a graphic depiction of the commonly used terms and definitions of each theme identified in the analysis and were invited to provide their thoughts and comments regarding the analysis and themes described.

### *Results*

Ten individuals with CP participated in this study, the sample characteristics of the participants can be found in Table 4-2. Many of the themes were inter-related and the experience of living with CP was shaped by several components at once. The major themes identified by the analysis can be seen in Figure 4-1. Each theme is described and explored in further detail next.

***Table 4-2 - Characteristics of the Sample***

Characteristic	
Participants	n=10
Age in years (SD)	19.2 (2.49) Range= 15-24
Distribution of Involvement	Diplegia n=8 Triplegia n=1 Quadriplegia n=1
GMFCS Level	Level I n=1 Level II n=6 Level III n=3
CFCS Level	Level I n=9 Level II n=1
Sex	Female n=4 Male n=6
Educational or Employment Status	Unemployed n=1 Full-time Student n=9



*Figure 4-1 – Major Identified Themes*

The bodily experience of CP centered on issues of fatigue and pain as a feeling of muscle soreness. Pain that was different from muscle pain was not as frequently discussed despite being asked by the researcher. A few participants explained what fatigue feels like to them.

*“I guess for me, it’s just a really sort of sore feeling, almost like a muscle cramp, but not to that extent. And to the point where if I try running or walking on them [my legs], it’s not that I completely collapse, but the sort of energy and strength that...that part of my body has is pretty well gone and so I can’t really do any, like, strenuous activity. I can still walk on them but even that is somewhat sore to do and then running or anything else is nearly impossible at that point.” – Mickey Mouse*

*“Well, [pause] well if...if you tie a knot in the shoelace and then pull it and you can feel it getting tighter, that’s what my muscles feel like if...when they’re tired and in pain.” – Dori*

A few participants mentioned that they fatigued less when their full body weight was not required for the activity, for example, walking in water or swimming; one participant in particular discussed this in great depth.

*“Yeah, I would definitely have to say that [managing my whole body weight] does have an impact [on fatigue] ‘cause if I’m swimming, I’ll...I won’t really fatigue as quickly as I would if I was say running or just having a long day at school. Mainly because the...that pressure is off of my body and off of, sort of, my legs and my joints so that I don’t reach that point of fatigue as quickly. So I would definitely say that being in a position where I don’t need to fully manage my body weight and sort of maintain the movement of my whole system is...is much easier on me in terms of fatigue and pain.” – Mickey Mouse*

*“Yes. I do find that as well in that not only just with the squats and the whole balance aspect because with a leg press you’re seated, so you don’t actually have to worry about that, and it’s just the one movement of your hips and your knees. Whereas with a squat it’s your whole body and your whole body movement, I find that for me squats are a lot more difficult than a leg press is. And I think that is in part because of the balance [needed], but also in part simply because there’s more of me to move around and so it has much more of a strain on my system and it’s a lot tougher for me to actually do than, say, a seated leg press.” – Mickey Mouse*

An overwhelming amount of the discussion on fatigue emphasized the fatigue that occurs with walking and that the experience of fatigue primarily existed in the muscles of the legs. This was prevalent throughout all of the conversations and was mentioned throughout the discussion of other related topics. One example comes from Travis as he spoke about when he was most tired during an average day.

*“Usually I feel...usually I feel the most tired...at period four just because I’ve been doing...I’ve been doing a lot of walking just because the classes I have are on the...on the main floor, and then I have a class on the first and the fourth floor. So there’s a lot of walking for me to do, and by then in the day my legs are pretty tired because of the amount of walking that they...that they’ve had to do.” – Travis.*

Many other participants reinforced the issue of fatigue with walking and prolonged activity.

*“I would say more so distance and more on foot stuff would fatigue me more than necessarily swimming or other physical activity.” – Mickey Mouse.*

*“Most likely physically tired ‘cause of...well, my muscles and...like walking from...like, from here to the fitness room isn’t...isn’t a long walk but it’s a long walk for me.” – Dori.*

*“Probably running, [causes the most amount of fatigue], I would think that is the one major one. Or, just in general, like moving over long distances, so like running or walking. In terms of anything else, like going up stairs, moving around the house, that doesn’t really generate as much fatigue, but when I run or walk for longer periods of time then that is probably what generates the most fatigue for me.” – Mickey Mouse.*

Many individuals talked about fatigue in a way that conveyed a finality of fatigue, speaking about it in terms and phrases like “my legs are done”, “I’ll collapse” and other analogies that symbolized the intensity and large impact of fatigue for these individuals.

*“Usually...usually my legs would get tired more so...I just find that...and I find that I’d be out of breath, I’d be short of breath too.” – Ariel*

*“They [muscles] kind...they feel...they kind of feel like jelly just because you’ve been using all the muscles in your leg, so it’s like...they feel like jelly.” – Travis*

*“It would usually tingle for some reason and then my legs will just jelly and I will collapse, sometimes.” – Jasmine*

Fatigue was mentioned as a major consequence of activity, many participants talked about feeling fatigued and sore from muscle pain after a challenging day or a day that contained a great deal of physical activity.

*“I find that, like, school trips...I, in the middle school, I went to a camp and I was really excited to go, and then after...after I came back from the camp I was...I was ill because I was so tired. Like, just...walking...a lot of activities throughout the day, like, my brain...well my brain couldn’t really keep up, so after that trip I...my brain kinda said what...”we’re done” so we’re...so it kinda...didn’t know what to do, so that’s...I think that’s why I got ill after.” – Dori*

Some participants talked about pushing themselves too hard with their physical activities.

*“Over longer periods of time, especially if I’m really really active and I remember the fall of this year I actually ended up playing for our senior football team, and I remember after a couple of weeks the practices would go for about two and a half, three hours everyday after school for five days a week. So that’s relatively strenuous activity. And I found after a week or two I could still continue, but I was often more inclined to sort of take it easy because I would be fatigued more quickly and...not necessarily pain but just, like muscle fatigue would set in a lot more quickly than it would have at the beginning of the season. I was probably pushing myself a little too hard, but yeah, that was the one thing that I did notice but with time it sort of progressively got a little bit worse.” – Mickey Mouse*



*“As far as the feeling goes, it...if you have, like some...there were some days where after a practice my back would hurt so much where I’d try to get out of bed and I’d have trouble sitting up. It’s like...the muscles get restrictive at that point and they kind of respond saying “You haven’t had enough time yet. You shouldn’t be going anywhere. Stop.” And that’s...the typical response is they tighten up and you feel it, and your range of movement is severely restricted at that point.” – Nemo*

Although fatigue was a consequence of activity, participants did not always view physical activity as something that solely created fatigue. Many of the participants talked about the benefits of being physically active on their endurance, energy level and even potential preventative action against fatigue.

*“Oh yeah. Obviously...like, being more active and stuff although it might fatigue you the day of and the day after, overall you’re getting stronger, right? So it’s...it gets better and you get more energy as you keep doing stuff like that, so yeah, I think it helps.” – Chris Bosh*

*“Well...I can’t, like, I...with cerebral palsy I can’t really walk that far without getting tired. Like, with fitness I, like I walk around, like, kind of around the...the fitness area and the first time I did it I walked...I could walk half of it and then have to take a break because I was winded. But now...but since I’ve started fitness I can walk three or four times around.” – Dori*

*“I think [skiing] helps me in school just because going down the hill it helps you with your endurance just because the...the speed you have is also...really helps you when you’re...when you’re in school because when you have to go to class and use that to keep up...to keep up pace so that you’re not late, and also really make sure that you’re keeping your body fit so that if the time arises that your class is outside, to make sure that you...that you have enough energy so that you can go, so that you can be with your class outside.” – Travis*

*“First year of my Master’s, no exercise, it was just like...I was just like, all right, bookwork, you know, eating late at night, no exercise, no stretching, no nothing. And I gained, like 20 pounds in like three months, it was...it was horrible, I could barely function. I could barely bend over to tie my shoes, like even the little things like that I just notice, like...I was at a friend’s house and I just put my shoes on and I actually had to, like...sit down, and it took me, like, a couple of attempts to actually get the flexibility to, like, reach over my knees and to tie my shoes properly and that’s...that was my wake up call was, wow, I can’t even tie my shoes right now. You know what I mean? And it was like “I need to do something about this.” So even like little things like that it wasn’t, like, oh I can’t...whatever, like I can’t walk, like...five blocks, it was no, I can’t even tie my shoes.” – Goofy*

The time of year and/or the weather was a factor that many participants discussed in relation to the fatigue they experienced. Many participants expressed that winter weather compounded issues of fatigue and talked about other factors related weather, such as pressure changes and temperature.

*“[There is] definitely more fatigue in the winter and pain I would say ‘cause it’s a lot harder to move.” – Jasmine*

*“The two biggest factors [for fatigue] I guess, would be, like, how physically active I am that specific day, and the weather. The weather’s a big...so if it’s snowy, like, I find like, obviously, the ground’s not level, I’m sinking in the snow, it’s slushy, my shoes and/or socks get wet, which means my orthotics get wet. Or if it’s very humid, it affects like the plates...like I have plates and pins in my body, so, like, those are affected by changes in pressure. So they expand and contract accordingly. And...that causes bone pain. So it can depend.” – Goofy*

*“And I guess temperature’s a big thing because...you noticed when I was in here I’m a friggin’ hot pocket, right? So like...the thing about summer is “okay there’s no slipperiness, I can reach and I can pull” but I sweat like a dog, right? Whereas winter*

*it's just like there's more obstacles but my body temperature is lower and I feel...like, I go out in winter in t-shirts and stuff 'cause I burn so much heat and so much energy that it's just like...but I can keep warm, but at the same time I'm...you know, I'm going over, like...there's no 2-foot snow drifts in the middle of July, right?" – Wade Wilson*

*"The problems mainly arise when, like, it's 70 on Monday and then it's, you know, 45, 35 on Tuesday or, like, it's 100. You know, when there's extreme kind of changes in a pressure and temperature, that's where I kinda notice, like, I have a lot more muscle ache, my tendons are like stiffer...you know, and just general areas where I have, like, where I've had modifications hurt a little more, like not...well, hurts a strong word...there's discomfort would be more accurate." – Goofy*

One participant spoke about fatigue and the weather while relating it to his physical activity level.

*"In the winter time, I would definitely say I am more fatigued, I'm a lot sorer, I'm in a lot more sort of minor pain when I do physical activity. I think that has sort of a part to play in the fact that because it's winter most people are very immobile, you know, they're not going out and running five, six kilometers in freezing cold weather. So I think for me as well, you know, I don't...I'm not as active during the winter months as I am during the summer, so I think that definitely has a part to play, because in the summer time I feel more energized, I'm around, I'm moving, I'm swimming, I'm going for runs." – Mickey Mouse*

Although fatigue was discussed by all the participants with CP, the theme of self-awareness emerged from the analysis as the most essential theme related to the bodily experience of CP. One component of this theme was becoming self-aware of the limits of their physical bodies. In this theme, participants spoke about knowing (or not knowing) the limits of their bodies. One participant spoke about the experience of learning his own limits of his body:

*“But...you know, I was kinda telling myself, like, I would say things to myself like ‘What, are you soft?’ blah-blah-blah, to use whatever lingo I want, but ‘I can go for another 10 minutes.’ Then you’d reach that 10 minute mark and I’d feel like I had a little bit more left in the tank, ‘I could go for another 10 minutes’. Well I ended up going for about, like, 42 minutes or whatever, I couldn’t get off the treadmill. Like I couldn’t...once I stopped and then my muscles had, like, had a chance to just kinda settle down or whatever, I couldn’t move. I had to be, like...carried off the treadmill and, like, out of the gym and then picked up, like I couldn’t...so...and for me it’s not like...I don’t feel myself, like progress...getting progressively tired more so, like if I’m walking throughout the day and whatever, then yes, but if I’m at the gym and doing something intensive, I think that, like I don’t really...I...it...it just...like I can coast then it just hits me all at once. Like, oh my god, like okay now I have no energy, kinda...So I’m...I don’t get like progressively fatigued, but I know that, like, I know that about myself, so I...and through trial and error, years and years of whatever, I’ve been able to figure that out.” – Goofy.*

The same participant spoke about learning how to be aware of his surroundings and how to negotiate his body to compensate for difficulties with his balance as a result of CP.

*“Getting older I kind of learn the tricks, kinda wa...know how to watch my footing, I’m always kind of aware that I’m around a wall or something to grab onto just in case I happen to, like, lose my balance or whatever, so...I don’t fall as often now that I’m older, well...getting older, ‘cause I have a better awareness of kind of my surroundings and my footing, whereas when I was younger, I just wanted to get out there and do things...” – Goofy.*

During the comparison across participants, the level of self-awareness that each individual had achieved varied greatly. The level of self-awareness showed some relation to age, such that the older participants in the study talked openly about knowing their limitations and the consequences of activity, yet in some of the younger participants this

self-awareness was only starting to emerge. For example, Nemo who was 19 years old speaks about not realizing his fatigue level until after activity is over.

*“So there’s, I think...I feel it a lot more when I’m finished the day, when I actually realize how much I’ve done and then I go sit down and it’s like “I’m going to be feeling that later as opposed to now.” – Nemo*

Interestingly, when asked about strategies to combat fatigue or to stop activity before it reaches the point of having a consequence, this was something Nemo had not yet considered.

*“As far as strategies go...I’m not really sure I have one.” – Nemo*

Travis who was also 19 years of age recognizes the fatigue in his body sometimes only when it is too late and speaks about what can be done in the future, but has not yet learned how to predict his fatigue.

*“My legs are...my legs are done. ‘Cause after...after a good bike ride with my family, I can definitely tell my legs are done because...there are times where I’ve had...where I’ve had to stop and actually had...had to have one of my parents push me home because my legs were totally done.” – Travis*

Travis was becoming aware of his limits and potential ways to mediate the effect of fatigue on his body.

*“[Using ice cream as motivation] it doesn’t mean that my legs aren’t going to be tired, what it means is that even if my legs are getting...are tired, I’ll still get there, even if it means that my legs will be angry at me when I get...when I wake up the next day.” – Travis*

Mickey Mouse was 18 years old but has had a lot of exposure to training and is very physically active. This has been helpful to develop his sense of the limits of his body and how to learn to recognize the signs of fatigue. He notes that there are times he did not recognize that he had gone beyond the physical limits of his body, but that now he has found a method or plan for ensuring that does not happen often.

*“I find that if I don’t keep a regular sort of regimen in terms of physical activity, I will fatigue gradually, just in terms of normal fatigue. But, another thing too is if I am travelling long distances on foot, I will actually fatigue more quickly than those without cerebral palsy.” – Mickey Mouse*

*“How do I put this? Really, when you reach that point where your body is...has completely exhausted all of it’s energy, and I know this has happened to some of my able-bodied friends as well who push themselves way too hard...you really...you have no energy left the morning after. You literally get up and realize you have no, really life in you at all, and so movement is very lethargic, you know you’re not very active the day after, most of the day I would probably either spend sitting downstairs, or lying in bed or lying on the couch because I’m so tired and the tiredness and the pain are still there from the day before or two days before depending on how hard I pushed myself. So it is that sort of...gradual sort of recovery period where you have to take the time off to reach that neutral point again. But I’ve probably only reached that point a couple of times, not very very often. ‘Cause again I try to...the best I can if I can reach a sort of closer point I’ll try to take it a bit easier.” – Mickey Mouse*

Wade Wilson was 20 years old and he talked a great deal about being aware that he cannot do things the way someone would expect him to, as well as ensuring that his own comfort is important factor in determining the choices he makes.

*“You know what I mean, like you’ve got to find your own way to, I guess...like...yeah compensate and to...to deal with it, I guess.” – Wade Wilson*

*“So...yes. I guess it just comes down to adaptability and finding a different way to do things, but it’s about comfort too, you want to be comfortable, right.” – Wade Wilson*

Jasmine, who was 21 years of age has become aware of the limits of her body. Although she did not think she was directly planning her activities as a result, she understood how to predict the occurrence of fatigue and techniques to manage this in her daily life.

*“Sometimes I can tell, sometimes I can tell the day before because of what I’ve done that day. I know there are consequences of what I’ve done, so I can sometimes predict, you know, because I did this today, this is what I’m going to feel like tomorrow, and I can get ready for it, right?” – Jasmine*

*“Well like...when I went for that [3 hour] walk, I knew that my trunk was going to hurt the next day and the day after. So then I kinda was able to plan my days, or...not plan my days ‘cause I’m not a big planner, but, like...like you know, sorta say well this is what it’s going to feel like and this...prepare myself more mentally, I guess, for it.” – Jasmine*

Goofy was 24 years old and the participant with the most developed sense of self and his limits. He talked a great deal about knowing his body and knowing his surroundings and merging the two in order to enhance his functioning within his environment.

*“So even though, like my energy level and balance is...is...well I wouldn’t say my balance is worse, but my energy level is definitely lower, my pain threshold is a little lower, recovery time is a little lower....but I have more of an awareness now, so it kinda balances each other out, ‘cause now I know what not to do, how not to step, how to kinda plan a route...and the route can be you know, if there are many obstacles, I’m like how am I going to get around them without falling over or kicking something over? So that...it’s kinda more of a preplanning and an awareness of my own ability in conjunction with each other to get from point A to point B without falling over.” – Goofy*

*“You know, I could probably walk the five blocks, but then the next day I wouldn’t be able to walk two feet. You know, that’s the thing. And you’ll find, well, for me personally and I think for a lot of other people, is your body...everybody’s body adapts, so...when I’m working out, I can feel certain muscles cheating. And it’s not because, it’s just that’s how my body....my body has always adapted in that way so...so, like, I can always feel certain muscles in my back firing that shouldn’t be firing. But it’s just it’s just to help me accommodate the weight I’m lifting. So then I know to tell myself I need to reduce the weight so that muscle doesn’t kick in to cheat.” – Goofy*

Goofy spoke about creating strategies for dealing with specific issues related to balance or fatigue associated with CP.

*“I always question kinda in my mind, like...okay...if I’m carrying two things of kind of equal or similar weight, so it could be a gym bag in one hand and textbooks in the other, whatever it may be...do I carry both and try to, like, balance out the weight, or do I want to have one hand free ‘cause then if I slip, I have, you know, something to protect myself so I’m not...going face first into the concrete. Usually I opt for option B.” – Goofy*

*“So it’s a lot of, like, preparation and knowing like...but that speaks to balance ‘cause then I find myself, well how am I going to open the door if I want to hold these three [things], if I can’t put them down?” – Goofy*

Although not all participants were fully self-aware, many participants spoke understanding the need to continue physical activity in relation to maintaining their ability to ambulate.

*“And plus I wanna...and plus I wanna...practice walking.” – Hillary Duff*



*“Just to maintain my muscles in my legs. Because I used my chair non-stop probably for a year, like I...I walked around the house but any time I went outside the house I used my chair and the same with school, like anytime I left my room, then I would use my chair and I noticed that my pants were too big in the back because I had lost so much muscle and I didn’t, like, that was...I didn’t even realize that I...that happened, right? I don’t want to do that again sorta thing.” – Jasmine*

*“I will try and walk as much as I can ‘cause don’t want to lose my strength, right? So...like, yeah, I walked for three hours the other day and I was...I noticed that I was tired, but I didn’t have to stop.” – Jasmine*

*“Not most of my life, but in the last couple years, I’ve really sort of begun to understand the importance of it [physical activity]. When I was very young I really didn’t have any mobility at all, I was confined to a wheelchair, you know, someone had to push me around. From there it gradually progressed to having ankle foot orthotics and a walker. And then, earlier in high school I eventually developed the strength to just become totally independent. So it was really at that point that I realized that if I didn’t become physically active as part of my daily routine, I would lose everything or at least backslide, so I realized sort of how important that was and began sort of taking that on.” – Mickey Mouse*

Ariel spoke in her interview about not being very physical active, and relating this to her experience of fatigue and her ability to walk now versus a time where she was more active.

*“Well I find now I kinda need to improve my walking, I don’t find I can use my walker as much as I used to. I find I get tired a lot more easily, probably because I don’t use it as often as I should, but...I use the wheelchair more, it’s a lot faster, but I do find it’s inconvenient because I have to...my parents have to take it to take it and set it up, so that’s what I do find inconvenient, but I wouldn’t...I wouldn’t be able to lift it up myself.” – Ariel*

There were several themes that were related to self-awareness. Planning was a theme that was evident in most of the participants' responses; however, in some cases the participants did not realize they were planning and making adaptations to their activities or daily lives to manage with fatigue and/or pain that they experienced as a result of CP.

*“So I think it [fatigue] does play a part when I am planning the week, but more subconsciously, like I’m not really fully thinking about it, I just sort of tell myself when I get up, okay I’m pretty tired today, so let’s take it easy. But I really don’t think about that if it makes any sense.” – Mickey Mouse*

*“If I’m going on a trip or...like, if there’s something out of the norm, then I try to, like, prepare myself, so like...for example if I’m going to the mall or whatever, like, I kinda plan out my route to...kinda figure out what stores I need to hit, and where I’m going to be, kinda, standing the longest, or like, if there’s some...where I could sit, or like a wall I can lean against.” – Goofy*

*“Sometimes it is, just...well, for example, I went to Toronto with another friend who has CP and we were both in our chairs, and I had to do a lot of planning for that, just in terms of how I was going to get around and like...I knew I was going to take my chair again, you know, how much can I do in one day...and that sort of thing, and just making sure that I could get where I needed to go in the right amount of time.” - Jasmine*

When asked about having a routine, Ariel spoke about needing to plan more in her life to help her manage her fatigue and overall tiredness.

*“I think it...I think I wouldn’t be as tired [if I had more of a routine], and maybe would be more active. I find that even...like, I just...I think it has, over the years I think I kind of...I kind of decreased...I’m not as healthy as I maybe used to be.” – Ariel*

Participants often spoke about making adaptations to their activities as a part of their everyday lives.

*“And I mean you might not, you know, move or do things the same way as most people but I think you find a way.” – Wade Wilson*

*“You know, kids with CP or any kind of physical disability I guess would be...you know, we all...kinda want independence, we all kinda wanna, you know, be able to do our own thing and...find a way to get things done and...I find, like...a lot of it comes down to, you know, adaptability and there’s always a way to do something I think it just...like you can’t really do it, you know, in the typical way, the way people are used to doing it.” – Wade Wilson*

Both Chris Bosh and Goofy talked about finding new approaches to every day activities, activities that would not normally require a plan for someone without CP.

*“You think of more than one way to do things, that’s because you gotta find the easier way and most effective way, so it’s not...too difficult and then doesn’t take too much time.” – Chris Bosh*

*“So you know, to carry three...you know, you’re out to here and it’s very cumbersome for me and I find, like okay, well, yeah, I can either hold these files or I can open the door in front of me ‘cause I need to get through that door eventually, so I would have to put two files down, hold one, kick the door open, put a stop, stop there and grab all three, go through, put things down and then shut the door ‘cause it’s a secure room.”  
- Goofy*

Participants in the study employed many different strategies to adapt their activities to manage fatigue. Dori explained that she often relied on others to assist her with tasks that were unavoidable and generated fatigue.

*“But I tend to have scribes for me, like a lot of writing, like exams...like exams. I...I’ve had a...I tend to write them, but once...if my hand gets tired I get...I tend to get a scribe.” – Dori*

Dori also spoke about selectively choosing to use the least impaired limb for tasks that required endurance, because of fatigue.

*“Yeah, it’s...that’s part of the...that’s most likely the reason because...well since I haven’t used it [left arm] for many things, I’ve, like...I can go maybe a minute with doing something with my left arm and then I have to take a rest because it’s tir[ed]...like, the muscles are so tense I can’t...like I can move it but not...not as much as I wanted. So...I tend to use my right arm for everything.” – Dori*

Several of the participants in the study expressed that they chose a different method of mobility than they either usually employed or would like to use, in order to manage fatigue, pain or other difficulties they experienced with their activities.

*“I don’t really, like, want to use the chair more than I have to. So if it’s small enough that I know I can walk around, then I will. But yeah, if it comes to a decision of, you know, am I too tired for this, then yes I will use it more.” – Jasmine*

*“Usually I will take my chair on a trip like that and I know that, like, I don’t have to think about it, like...if I’m going somewhere that I know I’m gone for the day and I need to keep up with people then I will take my chair because it’s easier to get help that way too.” – Jasmine*

*“[I] started using two [crutches], ‘cause you know, I would hit like an ice patch and like “Uhhh” you know, I still had one more to try and like...and so I guess it’s just like...and then I, you know, could carry the bag and I could last the whole day, you know what I mean? So I guess it just depends on where you...kinda like, I guess, divert*

*your energy, right, 'cause my knees probably wouldn't last that long, but like...I guess you find, like a different way of doing things. If that sorta makes sense.” – Wade Wilson*

*“In advance, I would either...if I knew I had a lot of walking to do, I would either pack...tell my mom to, or go myself to get the walker or the, like, my...I have...I have canes, a cane too, so...if...if I want to, like, if I want to walk, but not far, I use the cane or the walker, most likely the walker, but, again, for a long, a long walk it would be either the wheelchair.” – Dori*

*“Then I go down to the cafeteria for dinner. And I usually take my chair there because I can't carry food by myself without dropping it, but if it's on my lap then it's not going to go anywhere.” – Jasmine*

*“There are various things I have to consider such as...transcripts of my notes and if I'm getting a lot on a certain day opposed to just recording or listening to a lecture. And getting from class to class can be difficult on some days, like, some days I use my walker, and some days...I have a motorized scooter at home, which, has been a huge help even though I was highly opposed to the idea originally. I...I'm very attached to my independence so...the idea of power mobility, I didn't want to become too self-reliant on that sort of thing.” – Nemo*

*“If I'm in my chair I can last a lot longer, like I don't get tired very easily, like, I can go a full day without getting tired. But if I'm walking without my crutches which I'm not supposed to do, but it's easier sometimes...then I can walk probably less than a block before I get tired.” – Jasmine*

Two participants talked about using activity or stretching to keep muscles from becoming too tight or from fatiguing from maintaining a static position.

*“Like I’d try and stretch and stuff, and...like in a sense that pain is a good thing ‘cause you’re...you’re trying to...like...’cause I feel like the muscles that can’t build, I think, are because they’re so tight and stuff, so the more you keep them stretched out the more you can activate them and the more...the more that you can activate them, the more they get stronger, and the less they tighten up, right, like the more you use them.” – Wade Wilson*

*“Because if I keep my muscles moving, they’re prone to sort of get used to that movement, and at least give my muscles a change to stretch out. Because I think along with that tightness that we mentioned, oh I think that’s more prone if I do stand or sit for longer periods of time, that that’s going to begin to tighten up and become more [fatigued].” – Mickey Mouse*

Mickey Mouse talked about pacing his activity in order to manage his fatigue while running a race.

*“A couple of years ago me and Mom ran a 5 kilometre run back in the fall, and I would run for part of it and then walk for part of it. So I would run for say, two to three minutes, maybe less, and then I would walk for say a minute or two minutes. Now I did that for probably close to two and a half, three kilometres, and then I reached a point where to run, physically, the strength in my legs to push myself at that speed just wasn’t there. And I was, I was too tired, I had no energy in terms of my actual leg energy to continue at that pace. So at that point I just...I walked and sort of slowly jogged the rest of the way.” - Mickey Mouse*

Nemo spoke about researching his disability to be prepared for and to understand his symptoms such as tremors.

*“The more I know about my disability, or the more I’ve researched it, it was a real...probably a real concern to me when I was...younger, just because I knew that probably shouldn’t be happening, like I knew like...when I get tired, I didn’t know*

*then I couldn't control my shaking, that's just a natu....as I said before a natural byproduct of fatigue, but now I know that there's no use in trying to combat it that just because it's natural and I know it's going to happen, it's not as big a concern or me anymore.” – Nemo*

One specific adaptation that was discussed by a large number of participants of this study was restricting activity as a result of fatigue. Many participants, and some of their family members, talked about having to restrict activity as a result of pain or fatigue.

*“So activities stop earlier because of fatigue than you might want them to.” – Travis’s Mom*

*“And then I can still remember one time in camp where everybody was going out for a scavenger hunt and I stayed back with the teachers because I was so...I was so tired...I was so tired, like I couldn't sleep because my muscles were so sore from the previous day.” – Dori*

*“If you do things for a day, by supper time...you can't even think about doing something else, where other kids go golfing or whatever.” – Mickey Mouse’s Mom*

*“As far as just day to day, like...like going out and...like play, like...I know it's different with my age now too, but yeah I used to go out in the winter time and, say go tobogganing or something but I wouldn't even try to attempt that anymore. I was a lot lighter and my Dad could carry me around a lot more when I was smaller, but, to do all that by myself it would be just too physically tiring, and I just couldn't...I couldn't do it unless...like, I'd be done for the day after an hour of doing that kind of thing.” – Chris Bosh*

*“I think everybody when they were younger thought they were invincible and couldn't be...couldn't be touched, but now, not only has physically become a factor, but people possess the brain power and the common sense to say some activities on certain days*

*you can just step back and say “I shouldn’t be doing that today” or “maybe I should put that off for a while” because fatigue or soreness or pain or any of that stuff.*

*Whereas when you were younger everything was “I have to do this right now” and “this can’t be put off till later, I don’t care how tired I am, this has to be done right now” and that’s kinda...mentally that’s changed, and physically that’s changed too.”*

*– Nemo*

*“So I think, especially in my case, you know, wanting to do all these sports and wanting to be physically active, knowing that I’m going to tire out quicker sort of almost hinders me to do those things. So I think that could be one of the most worrying symptoms, to me, at least. Just because there is that fear that if I do get so fearful of just tiring out, tiring out and tiring out, that I eventually just abandon the whole idea entirely and then become sedentary, which I know is not going to help me at all.” –*

*Mickey Mouse*

Interestingly, restriction of activity did not simply mean stopping or not pursuing an activity because of fatigue or pain, Mickey Mouse spoke about reducing the intensity of an activity to manage fatigue.

*“If I’m planning to work out Monday, Wednesday, Friday, and say Monday I have a great day and...but Tuesday I’m really tired, I might say, well, okay, let’s take it easy on Wednesday because I don’t want to wear myself out, you know middle of the week. So...that doesn’t mean that I’m not going to go in for a workout as to not over, you know strenuate myself and then eventually no energy by Thursday or Friday.” -*

*Mickey Mouse*

*“With me if I reach a point of fatigue where I realize, okay, if I go any harder I’m probably not going to be able to get up stairs to sit down and actually recover. So at that point, you know, I’ll pace myself, so I’ll still keep working but not at the intensity I was, so if I’m in a workout, I can at least finish the workout, but not overwork myself to the point of not being able to recover afterwards.” – Mickey Mouse*



Similarly, Dori spoke about changing her activities to compensate for pain.

*“Well if, like, my back’s hurting, I kind of avoid doing anything with it. I just kind of like stretch it out. So it’s...yeah, I kind of stretch out muscles that are hurting, I just kinda, like stay away from, for fitness I kinda stay away from working out muscles that hurt, I just stretch them out instead of working them out. And that seems to be....seems to be better than having them hurt.” - Dori*

The subtheme of rest as an adaptation made to accommodate fatigue or pain during activity was prevalent and discussed by almost all of the participants. This discussion often included talking about building in rest breaks, taking the chance to sit down or to lean on something or someone to relieve fatigue and/or pain and to continue with their planned activity. Strong language was used by the participants to talk about the need for rest, including words like “recharge”, “recuperate” and “removes pressure” conveying the overwhelming impact of fatigue on these individuals.

*“I just sit down on my walker and take a rest.” – Hillary Duff*

*“After school, after school I...hmmm...I generally just...go home, go home and...take a load off my feet because at the end of my day...well my left foot and mostly is painful, and I just take...I just take it easy...and then whenever I have to get up, I get up and walk...and walk around a little to loosen it...loosen it up, but...I can’t do most, like, a lot of walking once my foot starts hurting.” – Dori*

*“Just like...And like...you know, I was pretty done when I got here [to the interview], but like now that I’ve been sitting down I’m just like, you know, I’m starting to unwind a little bit, like, I’m still tired, but I could go for a little while, you know what I mean? I’m starting to recharge my batteries I guess.” – Wade Wilson*

*“Oh the biggest one is right when I stop, right when I get to the door, and like I’ve put them down...like I’ve put them down and it’s just like “Okay, I need to sit” you know what I mean? And then, like, after I sit down for a while, like...you know, I start...like...like okay I can get up and I can walk a little bit now and like I’m starting to get back to my...like norm or equilibrium or something.” – Wade Wilson*

*“I don’t know, it’s like...sit down and put your feet up and like...give yourself a chance to recuperate.” – Wade Wilson*

*“Yeah, so, like, things like that [dancing] are pretty strenuous too ‘cause again, you’re like...you’re moving in repetitive motions and your...certain muscles aren’t getting a break. So like I said...and it’s funny, but I mean, I find that I...I look for areas to cheat, so if I’m out dancing, like, okay, closer to a wall perhaps, or like a chair so I can kinda...every two minutes or whatever just lean up against it, recharge the battery real quick and go, you know, go again.” – Goofy*

*“So I would lean up against Mom for support as well, so I think that is...that is also sort of a factor I need to remember is that when I do reach that point of tiredness, I often lean on objects, like chairs or walls or other people, just to sort of support myself because I understand that my posture and balance was off.” – Mickey Mouse*

*“It’s the whole idea of bringing the pressure off of myself because for me to just say walk down a curb or just to get through the house, is...it’s still putting pressure on me whether it’s balance or fatigue, so just to regain that balance and sort of regain sort of a neutral position so that I can recover is sort of my point for leaning, and my reasoning.” – Mickey Mouse*

*“And sitting helps, like even sitting for, like, two minutes...really recharges the battery I guess, like if I’m exhausted I can sit for like, two or three minutes. That plays like a huge role and I can usually go for about another hour.” – Goofy*

*“Whereas you have...as far as daily activities go, you have time to kind of...yeah, recuperate, recover when you’re sitting down.” – Mickey Mouse*

*“There’s times when...there’s times when after gym class...when...when I’ve been using my leg muscles that I’ll...I’ll take a breather just because the muscles in my legs are tired from the...from the extent that I was using them.” – Travis*

*“I remember when I was with my...when I was with my family on a trip, and we were walking and there was...we were taking a track all the way to the lake and I had to stop because the walk there was so long and my legs wouldn’t...were getting to the point that they want...that they wanted to stop, so I sat...I sat on the bench because I needed to get my legs...I needed to get my legs...I needed to get the blood in my legs moving again so I just sat...I just sat down and then I continued.” – Travis*

*“[He needs a rest] every hour, hour and a half.” - Travis’s Mom*

*“[I sit for] 15 to 20 minutes and I would take to just sort of...to let my leg muscles relax.” – Travis*

*“Well, usually if I’m noticing that it’s hurting then I’ll stop...I usually stop every hour and take a break because then I start to notice it.” – Jasmine*

*“So I basically just, like, squat and sit there for, like a few seconds and then I’ll get back up and then I’ll be okay to move again.” – Jasmine*

The participants also spoke about planning rest into their weeks, taking into account all of their daily activities put together. Several participants spoke about using the weekends for rest and the need to consider the additive action of fatigue over the week.

*“Yeah, and...well, with...with the weekend, I kinda just...it’s kind of just...it’s kind of just my lay back and chill days because of being on my foot all week and...it’s kind of*

*what my brain, what my body wants, to just sit down and take weight off my foot and...and just chill.” – Dori*

*“It depends on what I’ve done that week. ‘Cause definitely, like if I’m going and going and going and then I stop, then I feel it a lot more because I actually haven’t given my body a chance to recuperate.” – Jasmine*

*“On the weekend it gives me freedom to relax and...either get together with somebody outside of school, or...take the time to kind of recharge for the next Monday, I guess.” – Nemo*

One participant spoke about not wanting to delegate “days for rest” and really thought that including rest on a daily basis was important.

*“Not really, like I don’t say like okay this is like a rest day, I mean, I’ll take like an hour or 20 minutes, or whatever I need.” – Goofy*

Goofy also talked about the reverse effect of too much rest or being too sedentary and how that affected his functioning.

*“So sitting is a big thing even if it’s just minimal. But that also has a reverse effect, so if I’m sitting for like two hours, I...you know, then I’m an old man and I can’t like...it takes me about, you know, 20 minutes to get up.” – Goofy*

Many of the themes were inter-related and the experience of living with CP was shaped by several components at once. Wade Wilson talked about the weather affecting his fatigue but also the combined effects of weather and fatigue on his selection of activities.

*“I still go out in my wheelchair a couple times a week maybe, like, in the summer and stuff. But like, if you’re in the winter, obviously there’s no point and...like, you know,*

*winter versus summer it changes the way, like, the way you doing things.” – Wade Wilson*

The theme of emerging self-awareness was also linked to restriction of activity. Nemo and Travis were two participants who were just beginning to understand their own limits and talked about these limits in terms of their activities.

*“There’s definitely some weeks where I have to consider am I going to be up for this by the end of...the week, or...am I going to have to put this off for another week to where I’m not as busy on the Monday or Tuesday, and I can handle that on my schedule.” – Nemo*

*“As I said before there’s kind of a time delay on it where sometimes you feel like you’ve got lots of energy and lots of reserve and all of a sudden it hits you and it’s like ‘Oh man, I shouldn’t have done all the stuff I did today’ or ‘I’m glad I got all this stuff done, but now I’m not going to be able to do anything tomorrow because I’m so worn down from yesterday’ type thing. So, plan of action, a lot of times productivity suffers as a result.” – Nemo*

*“It’s...it’s muscle soreness just because after a long walk I can feel...I can feel my legs because at times when I’m on a bike ride that’s long and my legs’ll cramp, like, they actually hurt me because they’re done. And when that happens...and when that happens I know that I’ve over extended it a little bit, so...there’s when I’m doing exercises when I have to understand that there are exercises I can do, but there’s times when I have to stop because I don’t want to overextend it to...for my legs to...quit on me.” – Travis*

Balance was an additional concern that many of the participants spoke about in the interviews. Balance was described in the study as something with a very fine line, the participants used very descriptive words like “takes a swing” and “hanging by a thread” reflecting the precarious nature of balance in CP and the variability of this attribute.

*“I don’t know, I’m very wobbly, like...I need...ask my brother, he...like I can’t stand still for more than three seconds.” – Jasmine*

*“I remember when I was younger, my balance would be very much sort of hanging by a thread, so I’d be walking, and if one single thing deterred me at all, I would trip and fall over. Whereas after the surgery, I have become more prone to if I start to lose my balance, being able to recover to a neutral position, so I think that is more predominant, now than it was, say, eight or nine years ago.” – Mickey Mouse*

*“I can go [standing] for a little while, problem is I have trouble like...like, I’ve literally been like...you know, standing in front of the mirror for a little bit each morning trying to like, just stand, like...you know just trying to balance and stuff, ‘cause that’s a lot of my issue.” – Wade Wilson*

*“But the biggest impediment for me would definitely be balance, I still have difficulty, like, going down stairs without the assistance of, like, a wall or...so I can’t do...or I shouldn’t say can’t, but...I have difficulty doing step...going down steps, two or more steps without assistance, like I...I would have to, like grab on to something or, like...and I’m not saying, like fully grab on, like it could be something as simple as just, like, making contact with, like the person next to me to, like kind of re-establish my balance and then go. But then I’m bumping into everybody and every...and that’s not the greatest thing either.” – Goofy*

There were clear relationships between balance and self-awareness, noticeable in the way individuals talked about their balance in relation to their bodies.

*“I don’t know, I think I just know my body more now, I think and I can recognize the signs and then prevent a fall, and I’m just a lot better at catching my balance now I think.” – Jasmine*

Further relationships were found between the subthemes of self-awareness in the way the participants planned, adapted and restricted their activities as a result of difficulties with balance. Specifically, participants spoke about impairments in balance being associated with their choice to participate in certain activities.

*“But it...again, it depends on what I’m carrying, what I’m doing. Even simple things like...I hate baking ‘cause I don’t like bending over the hot...and sticking things in a hot oven. But a stove or a barbeque no problem, I don’t mind that at all ‘cause it’s kind of higher up and...and it’s different. And, like, bending over, I hate the oven I hate trying to bend over and putting in things and, like, I just...when I’m bent over I don’t have my balance, obviously isn’t as great, so I’m always paranoid I’m going to fall in the oven or...whatever else.” – Goofy*

*“Well...I guess for me it’s balance, so...yeah, as long as I’m holding onto something it’s usually alright. Yeah I guess...it’s shaped who I am in a sense like in terms of what activities I do, I mean, like kayaking versus hopscotch or tap dancing obviously I’m not going to be a ballet dancer – it was one of my dreams when I was younger, so...I’m still you know, kinda emotionally distressed over that. I looked really good in a tutu.” – Wade Wilson*

*“In ’09 I had a...my left foot reconstructed to help me balance and...but it didn’t...it didn’t really work out, I’m still trying...I’m still a little hesitant on balance. So anything that people ask me for with balance issues, I have to say no. Even though if I want to do them.” – Dori*

Balance and fatigue were integrally linked in this study. Several participants discussed fatiguing more when required to simply stand still or maintain their balance in a static position, than they did during low-level activity.

*“Well, if I’m standing still...if I’m standing still for any long period of time, I...I’ll start to fatigue more so than other people, so...with me, my hips will actually rotate*

*and my knees will touch together, my back will slouch...so that does have an influence on my balance as well because, you know, my core is not activated to hold me upright, so I'm more prone to falling over if I were to, say, get bumped.” – Mickey Mouse*

*“Because [standing still] I...require more concentration, in like...actually knowing what my body is doing kind of...and to actually have, like, ‘cause...I think I would tighten up way too much and being tight tires me out no matter what I’m doing.” – Jasmine*

*“With being in one place for a long time, my legs will start to get tired, and when that happens...it’s a good...it’s a good thing to have a chair nearby just because if I’m standing for a long period of time my...my legs start...I can...my legs get sore.” – Travis*

*“I get the most fatigued when I’m standing still. Like when I’m...I feel like I get tired, like, for example, like when I was in school, like, elementary school, I would be...I would feel more fatigued standing for the national anthem and morning announcements, than I would for, like, outside running around at recess. ‘Cause it’s that constant kinda like pressure on your muscles and joints for an extended period of time with no movement. Even in the movements themselves, by shifting the weight and whatever, you’re giving other...other muscles kind of a breather.” – Goofy*

*“Well when we were...when I was in choir we had to stand up a lot because it’s better for your...your breathing kind of stuff, to stand up so you’re not all cramped. And so we were standing for, like two hours, which was a lot for me even at that point, ‘cause I’ve only been using crutches for six or seven years now. I used to stand up on my own and everything like that, so...But that was...that was my breaking point.” – Jasmine*

*“Right. But and...but the one place I noticed that I experience a lot of fatigue if I’m sitting at the piano. Because I sit in the same position with my back arched and it’s just...it’s a lot of strain on my lower back again. Yeah, and like...I don’t have a*



*chance to move, 'cause I have to sit with the pedal and...like my hands are moving, but other than that I can't move." – Jasmine*

Many participants also talked about balance being more compromised when they were more fatigued.

*"I notice I have more trouble with my balance just because it's a longer walk and I find myself getting tired easily just because that...just because of how long it takes me to get back up and going back home with my family. It takes a lot of energy to get up [from a fall] than it does to get down." – Travis*

*"My balance is definitely affected. More so when I'm...when I've reached that point of fatigue than just normally. I mean normally my balance is always affected, but it's more predominant when I'm tired. Partially I think because my posture and my gait become more irregular which leads me to having an off balance step. So I'm more prone to losing my balance at that point. But yes, my balance is definitely a major symptom as well." - Mickey Mouse*

*"I would say in terms of my balance, my balance is usually affected by the amount of activity I do, whether that's at home, or at the gym...you know, just in general. If I do work more, I become more off balance, I think I just because I've...I think my balance and my fatigue are very much linked, so the more I work, the more tired I get, the more tired I get the more my balance is affected." – Mickey Mouse*

The effect of the environment and weather were related to the challenges experienced with balance as well.

*"No, it's just the fact that nothing is clear [in the winter] and I always have to walk through snowbanks and over ice and stuff like that. And it's just a lot harder to keep my balance, especially with the crutches with the cork on the bottom, it doesn't catch as easily, so...like I know that when I walk with my crutches I use my arms more to*

*pull myself along than my legs, they're just kind of following. But I can't get enough grip on them to pull hard enough in the winter, so it's just I don't move as far for as much effort as I put in, right, so..." – Jasmine*

Variability was a theme many of the participants in CP discussed in a number of ways. The participants in this study talked about not being able to predict the behavior of their own muscles and many participants continued to state and re-state the unpredictability of their own bodies.

*"I think one of the things that...one of the things with CP is my balance just because for me I don't...like for me in different situations I'm doing with my family, I don't...I don't know when...I don't know when my balance is going to go – let's say if I'm on a walk and it's been long, I can tell that my balance is going to take a swing, because after a long period of time my legs get tired and that's usually when...that's usually when my balance kicks in and I either have to hold on to a tree or I have to just tell my family that I need to sit down because I'm going to lose my balance." – Travis*

Travis in particular spoke about how the unpredictable nature of fatigue further restricted the activities he participated in.

*"It's hard just because there are things that I can't do because if my...if my youth group is going on a hike, and...on a hike, I can't do it just because with my disability it's hard because I have no clue...I have no clue when my legs are going to give out on me and they...I don't want to...slow them down because they...I understand that they're on a hike so I don't...I don't...that's...that's also fatigue and pain." - Travis*

Some of the participants spoke about the amount of fatigue they experienced being variable depending on their day and the activities in which they participated.

*“Depends what I’m doing, like, if I’m walking a lot during the day then my body will fatigue faster, but just everyday...like...like I use a wheelchair in school so everyday it’s not too bad. I do get tired towards the end of the day though.” – Chris Bosh*

*“It’s [fatigue] kind of dependent on the day, so I can’t really give you a standardized answer.” – Goofy*

Goofy related some of the variability of fatigue with activity level to maintaining his balance.

*“Again, I’m always cautious to put, like, a time on it. ‘Cause I...it’s just level of activity I guess, so if I’m doing, you know like...I could be at a fatigue level at Monday at one o’clock that I am today at 5. It just depends on what I’m doing all day. But yeah, fatigue is definitely a factor just because...my muscles are tired and although, you know, it might take me...if I was, you know, kinda less fatigued then I would be more apt to trying, to catch myself, where...sometimes it’s just like no this isn’t happening I’m going down so it’s more of a protect your head, protect your chest and...get up after.” – Goofy*

Many participants talked about good and bad days, and that the experience of their bodies differed depending on their day.

*“Well, I’ll have good days and bad CP days, and on bad CP days I will try and not walk as much because usually my muscles are really tight and it’s just impossible to actually get my feet to move the way I want them to. I’ll drag them more which means I trip over my shoes more and then I fall more and then...I don’t know, it’s just a snowball of bad stuff.” – Jasmine*

*“I think part of it ‘cause some days I feel really, you know, some days I wake up and I feel like “goodness, I want to fly” you know what I mean? And some days it’s like I feel kind of lethargic and kind of you know...I still feel kinda lethargic at times, and I*

*still feel like...you know...sometimes it's like "Okay, c'mon pick it up, lets go, I'm late" or like "Damn it" you know "I'm slow today" like some days I feel good like, you know what I mean...but I like to...but I like to think that, you know if you keep at it I'm eventually going to get faster and stronger, hopefully, that's like...kinda the plan, so kinda you know...I guess compensate a little bit would be the word." – Wade Wilson*

Factors such as sleep, fatigue, activity level, balance and spasticity all played a role in determining good days from bad days. Participants also spoke about knowing that after a day where they felt good and maybe pushed themselves too hard, they had to incorporate rest and recovery into the days that followed.

*"Definitely my level of sleep the night before [determines whether it's a good day or a bad day] because it just takes a lot more effort to do things if I'm tired. And then I'm straining myself, I think, which causes more pain then." – Jasmine*

*"If I have a...like an energetic day, lets say I have a whole bunch of friends over and, you know I'm trying to entertain them, trying to move around the house at 100 miles an hour, then if that wears me out then I'll probably take it easy for the next couple of days just to sort of recover and reach that sort of neutral point again before I do anything really high energy." – Mickey Mouse*

Although participants were asked to focus on the physical aspects of their bodies, many participants could not fully separate the physical fatigue from mental fatigue and many felt it was an important associated factor that needed to be discussed.

*"Probably for me the one symptom that I noticed is probably just the fatigue and sort of the wearing down in itself. Because not only does that have an effect on you physically, in terms that you can't, you know, do long periods of activity, whether it be walking or if it influences someone differently depending on how they're affected by cerebral palsy, but not only does that affect people physically, it also affects after*

*doing it, it's going to have a negative effect somewhat on how you approach that situation in the future. You're not going to want to get back to that thing as readily as you were...if you weren't fatiguing every time you did it." – Mickey Mouse*

*"There's definitely some days I can tell that I've worked...had to work a little harder than others just because at the end of the day there's definitely more of a wear – either it be with stress mentally or just exertion physically, you can definitely...there are definitely some days that I'm more tired than others after I've finished the day." – Nemo*

*"But along with that comes a lot of other pressures, which people don't fully see. For me I know, it was a lot of psychological and mental stuff being...wanting to be at the same...not necessarily level, but sort of be at par with everyone else. And I think for me that was the biggest thing because it wasn't like I was confined to a wheelchair, you know, and sort of...I don't want to say so different but in such an extreme that it...I sort of understood where I was at and sort of that I couldn't really change it. I think that did have a lot of, you know, mental and psychological impact on me in the sense that I felt like I was always almost there, but there was always that one sort of step that I couldn't really overcome. So I think has, along with the physical aspect, has been a big part of sort of my 'walk' with CP as it were." – Mickey Mouse*

Many participants talked about physical fatigue affecting mental functioning, mostly in terms of schoolwork, but also in an overall a sense that physical fatigue can have an impact on their ability to perform mental tasks. As a result some participants used methods to limit fatigue in order to continue their academic pursuits to their fullest capability.

*"It's just because...I didn't use a wheelchair very much in elementary school, but moving to high school, like, there's a lot more moving around and stuff, and I didn't think I could focus, like...as much if I was tired from walking and stuff. And the halls are crowded and dangerous too." – Chris Bosh*

*“There are times that I get tired and that my brain just goes off, and...there’s times that I don’t want it to, but at the same time, with my disability, there will be times where my brain turns off and then...[my language capabilities are] a little off centre, so it’s like...when my brain’s off I tend to get a little bit hard.” – Travis*

*“Yeah. Well, I have some...with this cerebral palsy I have good days and bad days. And...bad days my, like, I can...if I have a test on a good day I could probably ace it. But if I have it on a bad day I could most likely fail it. And then...with studying it’s kind of a hassle because it’s...like information goes in one way and comes out like 20 other ways. So it’s not really...not really good.” – Dori*

*“I think I definitely don’t take as much, so like I may be looking like I’m paying attention. But because it...it took so much out of me to get there, I don’t...like I may look the same, but I don’t...it’s not...it’s going in one ear and coming out the other basically, right?” – Jasmine*

On the other hand some participants spoke about being able to overcome or withstand fatigue with motivation or if the activity causing the fatigue was fun and enjoyable.

*“So, for a while I remember for the first two or three practices I really thought, you know, why am I going to all the trouble to come out here for, you know, three hours at a time after school every day when I can be relaxing at home or working on an assignment or whatever. And then I sort of reached this point where I realized that it was for me to prove to myself that I could do these things, not necessarily to prove to other people that I was, say, good enough to do them, but more so just to tell myself, like, hey, why are you sitting around not doing anything, you can obviously put...play football and keep up with the other guys. So it was almost like a turning point for me is to understand that I can do these things and the reason why I haven’t been doing them, like, why I haven’t been doing them for years is really beyond me and that I should have been starting much earlier.” – Mickey Mouse*

*“It’s like you have to kind of look at everything in a positive kinda can do attitude. There are many different kinds of systems of thought with people with disabilities, some think that the world owes them a favour, you know, that it is all hopeless, and if they don’t have kinda that drive to kinda better themselves and know...kind of know or look to know what they need to do, then...yeah, they’re not going to follow thorough with any of it. The follow through is the biggest part. Or motivation is the biggest part, and then the follow through comes after motivation.” – Goofy*

Participants spoke about the benefits of exercise to their mental state as well, in spite of how fatiguing the activity might be. Participants also spoke about the motivation to participate in sports or physical activity allows them to deal with fatigue differently than fatigue they experience during daily activities.

*“I enjoy [fitness]. I feel that if I get...if I come into it tired, by the end of it I’m not tired anymore.” – Dori*

*“Pretty good actually. [Playing wheelchair baseball]...it’s giving me...it’s giving me energy to...it’s giving me energy to do what I want to do, kind of. Not actually getting me tired, it’s getting me pretty good in the baseball spirit.” – Hillary Duff*

*“Well...I guess moving around at school and studying I get tired and bored faster than...like, physically fatiguing at sports – don’t notice it as much as actually moving around and studying at school.” – Chris Bosh*

Some of the participants spoke about spasticity and tightness they experienced in their muscles and relationships between spasticity and fatigue or balance.

*“Like, when my muscles get sore, an example of kayaking or sledge or anything really – after I...after I run, or even after I wheel when I’m not using my legs, or if I go and play a game and do an hour of sledge, or...by the...like by the end of my kayak, right?”*

*Like you are...you'd expect because you are using all your arms that your arms would be the most sore, right? And like my arms are tired, but after a kayak, the thing that hurts the most is my knees, and my lower back and my legs because if I...like, you can't tell right now, but that's the whole idea behind spasticity and I'm fairly mild, again, so I'm...you know, I can...but if I'm rowing rowing rowing rowing rowing, and I'm like, I'm kicking, kicking kicking kicking kicking, so...like when my hands are firing, my legs are firing also, right? – Wade Wilson*

*“So I don't have the capability to, like...I guess discern...I don't know if that makes sense, but do you understand what I'm saying, so...so like...by the end it's like...I'm working out my knees and all of a sudden my shoulders are tired, but like...I could walk for a little...I could walk for, like a few hundred feet, like...you know, when I'm not tired, but like after the kayak even though I haven't used my legs per se, like I'm pushing against the pedals and I'm...I'm kicking constantly and it's like they're tight and they're pushing, right, so...by the end it's...the legs are more tired and like...you know, I'm still depending on...like I have to depend on my arms, like, more or...do you see what I mean, because...it's the legs that are tired. So, I mean in that sense I guess it keeps me lean, but...so I guess that's how it affects the muscles differently.” – Wade Wilson*

*“I prefer to be moving now because I know I will lose my balance if I'm stationary. So...I don't know, like I'd had surgery when I was 12 to lengthen the tendons in my ankles, my hamstrings and my hips. And I can tell you that before, like, in the...in the, like, two months leading up to that, it was horrible, like, I couldn't keep my balance no matter what I did, just because of how tight I was constantly and like...yeah. So I couldn't keep my balance no matter what I did and that was when I was about 12. And I think now that I'm tightening up more again, it's gotten worse again 'cause I know, like, 13 and 14 were good years because I was relatively loose in the, like...and so I could stand in one spot, I could walk without walking backwards more than forwards.” – Jasmine*



Finally, participants spoke about the increased energy expenditure required of them when participating in activities and the increased time required of them to complete activities compared to their peers. Both of these factors were talked about by the participants as “factual information” and were linked to fatigue. Particularly, individuals in the study talked about slowing down their pace of activities to manage fatigue and being accepting of the “fact” that activities required more time for successful completion.

*“It’s definitely...difficult because I think that cerebral palsy has this trait that it definitely takes more energy out of you to do the same sort of physical exertion as it would another person.” – Nemo*

*“I find it takes me longer to complete things as far as....mainly as far as walking takes me longer and I find that just getting around.” – Ariel*

*“Probably the...the slowness in the sense that when I’m walking I tend to be slower than other people when they’re walking ‘cause they’re...for other people, they can walk quickly, but for me I tend to walk slowly just because it’s...it’s not that easy for me to keep up pace without getting tired.” – Travis*

*“Yeah it [walking faster] does [have consequences] because I’m pushing myself more, and moving faster, I get tired faster and I have to stop more and stuff like that.” – Jasmine*

*“Like I can work as hard as I want and I’ll never, you know what I mean, I’ll never catch the slowest able bodied guy just on my feet.” – Wade Wilson*

Nemo spoke about the need to balance his desire to maintain his ability to walk and his need for a mobility aid to facilitate his transfer between classes at college, because of the energy expenditure and time that these transfers required of him.

*“Just because I feel that if I become too self-reliant on it [powered mobility], that’s all I’m going to use and I’ll be stuck to a chair for the majority of my adult life. And if it weren’t for surgery and various means of therapy I think I’d already be there right now, but I’ve been lucky enough to retain some of my...function in my legs and...I’ve been pushed in the past, I had been pushed for many years to switch to a wheelchair or powered mobility and I just originally didn’t like the idea, but I admit getting around at school it would take much much longer and it would be much more tiring if I didn’t have that means of transportation.” – Nemo*

### *Discussion*

It is clear from this phenomenological inquiry that fatigue is a major concern for many individuals living with CP. It is also clear that the experience of living with CP is complicated and multifaceted. Many individuals experienced some common elements, yet the variability in the experience of living with CP should be highlighted. Variability is a widely used term in all areas of research in CP both in how the neurological deficit is expressed (distribution of involvement, type of CP, secondary conditions that develop etc.) as well as in the everyday experience of each individual. In order to manage fatigue experienced on a daily basis, many of the individuals in this study found methods to adapt or alter their activities, while other participants had not yet reached this point on their journey to self-awareness.

The themes raised in this phenomenology have been echoed in written and published personal accounts described by authors with CP. Gwyn Jones (Jones, 2009) wrote about her personal journey of aging with CP and other co-occurring disabilities and presented many reflections that are consistent with the themes discussed by the participants in this study. Jones (2009) spoke of need to balance the effects of fatigue and pain through the use of creativity and innovation, describing the action of seeking new adaptations to activities of daily living as making the difference between living and existing with a disability. She also spoke of needing to prioritize, segmenting tasks into manageable pieces and sometimes restricting her activities (Jones, 2009), as did the participants in the current study.

In an ethnographic vignette of his own experience of living with CP, David Howe (Howe, 2009) discussed very similar concepts around fatigue, balance and notably the effect of weather on these factors. More interestingly though, Howe describes being intensely aware and feeling his every movement; it is this self-awareness that most resonates with the experience described in the current study. Interestingly, although a potential limitation to this study is a lack of participants with hemiplegia, the major theme and several subthemes resonates with Howe, who has hemiplegia. Furthermore, a former colleague at Western University, who also has hemiplegia, affirmed the themes of muscle soreness, balance and falling, along with an interaction of balance and fatigue and an acute self-awareness of his body and movements.

Typically, adolescents and young adults are not required to plan their days or weeks to combat physical symptoms such as fatigue; this is something that happens much later in life for the general population. Notably, for individuals with CP gait efficiency, gross motor function and performance have all been shown to slowly decline during adolescence (Kerr, McDowell, Parkes, Stevenson & Cosgrove, 2011). Even though many individuals understand the importance of maintaining participation in their daily activities, targeted therapy services for individuals with disabilities are often limited during adolescence. Many of these factors and more may combine and influence the experience of living with CP and the impact of many aspects of the condition on the lives of these youth. When preparing for transition from pediatric care, service providers typically encourage youth with CP to learn to manage their own health care needs (Gall, Kingsnorth & Healy, 2006). Self-awareness of their bodies and of the impact of fatigue should be fostered by service providers and included in clinical conversations about managing their health care needs for the present and the future.

#### *Relevance to Thesis*

The rich descriptions of fatigue provided by the participants in this study is a further affirmation of the importance of understanding fatigue for individuals with CP. Specifically, the personal accounts provided evidence that fatigue is complicated, multifaceted and variable. In addition to understanding fatigue, the descriptions provided by the participants revealed other key aspects of the bodily experience of CP that require further study: the developmental course to self-awareness and the need to plan. The

results of this project were used in addition to the systematic review (Chapter 3) to generate items for a new measurement tool to assist with assessing fatigue for youth and young adults with CP. A detailed description of how the new fatigue measure was created is provided in the next chapter (Chapter 5); specific questions regarding the key elements of the bodily experience (including awareness and the need to plan) were included as a result of this study. Once validated, this new measure may facilitate clinical conversations about managing the impact and severity of fatigue. Jones (2009) made several recommendations to facilitate healthy aging with CP, including forging collaborative doctor-patient partnerships. Self-advocacy is an important life skill to develop in order to participate in these partnerships necessary for navigating the adult healthcare world, and becoming self-aware is an important step to being able to initiate and participate in conversations about their healthcare needs. Some authors have indicated fatigue and inefficiency of gait as key factors contributing to loss of ambulation for individuals with CP (Mockford & Caulton, 2010); therefore, learning to manage fatigue earlier may prevent some loss of ambulatory skills for these individuals.

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## **Chapter 5: Constructing a New Clinical Measure of Fatigue for Adolescents and Young Adults with Cerebral Palsy**

### *Introduction*

There are numerous scales available to measure fatigue; however, as described in Chapter 3, all of these scales have been developed or validated in other clinical populations such as multiple sclerosis or Parkinson's disease and have not been validated for use in a population of individuals with cerebral palsy (CP). A well-constructed, valid and reliable tool to assess fatigue in CP would assist rehabilitation therapists and clients in collaborative goal setting and intervention planning throughout the lifespan, along with enhancing self-awareness and self-management of the condition. The review reported in Chapter 3 highlighted both strengths and weaknesses in the existing fatigue measures and served as a preliminary step for item generation to create a new fatigue measure. In addition, the interviews with youth and young adults who have CP described in Chapter 4 provided a client perspective to enhance item selection and generation for a new measure. The purpose of this chapter is to describe the creation of the Fatigue Impact and Severity Self-Assessment (FISSA). Specifically, this involved collecting multiple perspectives including information from the literature, clients and service providers to generate and refine items for this scale. Furthermore, feasibility testing was conducted to ensure acceptability by the final users of the scale, youth and young adults with CP.

### *Methods*

#### *Phase 1 – Initial Item Generation*

A copy of each fatigue scale that was identified in the initial database search conducted in the review was retrieved and examined for items relevant to individuals with CP. Item relevance was determined by two methods. First, I related the items to themes in the existing literature (for example: items asking about fatigue interfering with physical activities were selected because some literature has suggested fatigue as a potential cause of walking cessation). The second method of determining item relevance included relating items to the themes discussed by individuals in the phenomenological inquiry described in Chapter 4. Items that were deemed relevant to individuals with CP were rephrased to enhance relevance and to simplify the language and collated into a new

questionnaire. In addition to items identified from previous scales, new items were generated from the major themes related to the bodily experience as identified in Chapter 4 and added to the questionnaire.

#### *Phase 2 – Item Reduction and Refinement*

Two focus groups were held with health care providers (from a variety of professions) who normally interact with individuals with CP to reduce the number of items to a small, relevant subset of questions and to ensure content validity for the measure. Content validity is a psychometric property of a measure that contributes to the overall construct validity. Specifically, it assesses the degree of representativeness of all domains and elements of the targeted construct (in this case fatigue), within the scale (Haynes, Richard & Kubany, 1995). Content validity is important because it affects the clinical inferences that will be drawn as a result of use of the questionnaire (Haynes et al., 1995); involving expert clinicians in the refinement of this measure is a method of enhancing the content validity.

Ethical approval was obtained from Western University (Appendix 5-A), McMaster University (Appendix 5-B) and Thames Valley Children's Centre (Appendix 5-C). Each participant reviewed a letter of information and provided signed consent (Appendix 5-D). The focus groups followed a modified nominal group technique (NGT) (Delbecq, VanderVen & Gustafson, 1986) to reduce the items from the scale, as well as to reflect on any additional items that were needed to address issues that had been overlooked in the previous item generation phases. The use of both phases (reduction and addition of new items) ensures balance between retaining a high level of content validity and creating a feasible questionnaire and limiting respondent burden. The focus groups were audio-taped to allow for comprehensive documentation by myself as the focus group leader while allowing my participation in the session. Focus group participants were asked to review the questionnaire items prior to the meeting and make note of any thoughts or opinions they had regarding of any of the items on the scale.

At the focus group, the first step involved asking the participants, in turn, to select items they felt should be removed from the scale. However, it should be noted that rationale for removing the item was not to be provided at this stage. The objective of this step was to create a list of items that could potentially be removed from the scale, if the



same item was mentioned by more than one participant a count was created for that item in order to fully reflect the opinions of all group members (Delbecq et al., 1986). The next step in the NGT process was to have a discussion of each idea (question to be removed) mentioned by the group, again in turn. In this step, the rationale for the removal of each item was discussed and documented; at this time all participants were asked to discuss each item and whether they felt it should remain or be removed (Delbecq et al., 1986). The final step in the NGT process was a vote of the item importance for deletion. During this step, participants were given 5 separate index cards and were instructed to select the five most pressing items to be removed from the scale and write each one on a separate index card (Delbecq et al., 1986). Participants were first asked to decide which of their selection of items was the most important item to be deleted; they were then instructed to write the number 5 in the lower right-hand corner of the card and underline it three times. Participants were instructed to flip that card over, and choose the least important item to be deleted from the remaining four cards, write the number 1 in the lower right-hand corner and underline it three times (Delbecq et al., 1986). With the remaining three cards, the group was instructed to choose the most important item to be deleted and rank that card number 4, followed by the least important of the remaining cards and rank that card number 2, finally leaving the last card to be ranked 3 (Delbecq et al., 1986). A demonstration was given by the leader of the focus group to ensure each participant accurately completed the process. Participants were then given time to re-examine their ranking before passing the cards to the leader of the focus group. At this time, the leader shuffled the cards to preserve anonymity and calculated the results of the vote (Delbecq et al., 1986).

In the second part of the focus group, participants were asked to generate ideas about concepts that were underrepresented in the scale to ensure all possible concepts related to fatigue were covered by the measurement tool and the voting process occurred again for items to be added to the scale ((Delbecq et al., 1986). For the new questions to be added to the questionnaire, again the most important questions (as determined by the vote) were added the questionnaire.

After the initial focus group, which was held at the Children's Developmental Rehabilitation Program in Hamilton, the suggested changes were made to the measure

prior to conducting the second focus group, which was held at the Thames Valley Children's Centre in London. This allowed for a greater refinement to the measure and avoided redundancies between the two focus groups. Participants in the focus groups were given a record of the changes made to the fatigue scale, as per their recommendations, and were asked to review the document for accuracy or any further suggestions. After completion of both focus groups, a summary of the results of both focus group sessions and a copy of the final scale was provided to all participants. The NGT process was integral to the creation of the new fatigue scale as it incorporated the perspectives of service providers who work with individuals with CP. These perspectives are particularly important, as they will be a potential user group of the new fatigue measure.

#### *Phase 3 – Drafting of the Scale*

It should be noted that Phase 2 and 3 occurred semi-concurrently. Once a subset of items were established, the items were grouped into subscales to create separate profiles based on the attribute that each item was intended to measure (Impact, Severity and Management). These three attributes were selected to ensure that the entire fatigue experience was captured by the measure. As emphasized in Chapter 3, the newly created measure should specify not only the attribute(s) the scale is to measure but also provide a definition of fatigue to ensure all individuals completing the questionnaire understand the construct in a similar manner. A definition of fatigue was added to the questionnaire and specifically distinguished fatigue from the separate construct of pain. At the time of reorganization of the questionnaire into profiles, the scaling of the measure was also set. The full measure was circulated as a final review to focus group participants to allow for the expert healthcare professionals to review the content validity of these additional aspects of the scale.

#### *Phase 4 - Feasibility Testing*

To assess the feasibility of the questionnaire in the population of its intended use, 10 questionnaires were distributed to individuals with CP between the ages of 14-18 years who are classified as level I or II on the Communication Function Classification System (CFCS) (Hidecker, Paneth, Rosenbaum, Kent, Lillie, Eulenberg et al., 2011) as well as classified as levels I to IV on the Gross Motor Function Classification System

(GMFCS) (Palisano, Rosenbaum, Bartlett & Livingston, 2008). Ethics approval was obtained from Western University (Appendix 5-A), McMaster University (Appendix 5-B) and Thames Valley Children's Centre (Appendix 5-C). Participants were asked to read the new fatigue questionnaire (developed in the first 3 phases) and completed a feasibility questionnaire (Appendix 5-E) to ensure that the items were comprehensible to the population of interest, as well as unambiguous and asking a single question. I considered all responses from participants as a separate assessment of the content validity of the scale.

## *Results*

### *Phase 1 – Initial Item Generation*

The initial draft of the scale comprised 50 items. Forty-four items were selected from 9 different fatigue scales identified in the review. Furthermore, 6 items were created as a result of the interviews conducted with youth and young adults with CP. Appendix 5-F contains a copy of the items considered for inclusion from various scales and Appendix 5-G contains the initial draft of the Fatigue Impact and Severity Self-Assessment.

### *Phase 2 – Item Reduction and Refinement*

A total of 11 healthcare professionals (5 Physical Therapists, 3 Occupational Therapists, 1 Recreation Therapist/Kinesiologist, 1 Nurse and 1 Psychiatrist) participated in one of two focus groups. The initial focus group deviated slightly from the NGT process in that they reached consensus on 11 items to be removed from the scale due to redundancies, unimportant questions or multiple items asking the same question in different ways. Items that were removed in the initial focus group can be found in Table 5-1.

**Table 5-1 – Items Removed from First Draft of the Fatigue Impact and Severity Self-Assessment by Focus Group 1**

<b>Item Number</b>	<b>Item Descriptor</b>	<b>Reason for Removal</b>
13	I feel more isolated from my social contacts because of fatigue	Redundant and potentially a leading question
24	Fatigue makes normal day-to-day events stressful	Redundant with questions 32 and 41
25	My muscles feel weak due to fatigue	Redundant with questions 33 and 34
26	Fatigue makes me feel physically uncomfortable	Redundant with questions 33 and 34
28	Fatigue makes it harder for me to meet the demands other people place on me	Redundant with question 14 and too much variability with “other persons”
29	Fatigue makes me less capable of completing tasks that require physical effort	Redundant with question 10
32	Fatigue makes minor difficulties seem like major difficulties	Convolutd question, not clear exactly what is being asked
35	I need help doing my usual activities because of fatigue	Redundant with question 15
36	Exercise or physical activity brings on fatigue	Participants asked the question “If they don’t have fatigue why are they answering this scale?”
38	My motivation to do other tasks is lower when I am fatigued	Redundant with questions 21 and 22
39	Fatigue is among three of my most disabling symptoms	Not important, impact questions give much more information

In addition, the participants of the initial focus group concentrated on rephrasing the remaining items for consistency and clarity, and added 4 new items to the questionnaire, one of the questions added reflected the content of 5 separate questions from the initial draft. The new questions added to the scale can be found in Table 5-2. Participants in the second focus group received a copy of the FISSA with the changes made as a result of the previous focus group (Appendix 5-H). The second focus group followed the NGT guidelines and the vote resulted in 3 items to be removed from the questionnaire, these can be found in Table 5-3. The participants in the second focus group also added 4 items to the questionnaire, as described in Table 5-4, and provided input related to the definition of fatigue and the timeframe to be included in the introduction to the questionnaire.

**Table 5-2 - New Items Added to the Fatigue Impact and Severity Self-Assessment by Focus Group 1**

<b>Item Added</b>	<b>Reason for Addition</b>
A two-part question to include questions 43-47 of the original scale; specifically “Does your level of fatigue change depending on the time of day” then if yes answer, “What time of day is your fatigue worse?”	Participants felt more information was needed on when fatigue occurred if it was variable
Fatigue interferes with my ability to move around in my community	Participants felt that moving in the home and in the community were two different items and wanted that reflected as they could be impacted differently by fatigue
Fatigue increases my stress	Participants liked question 41 “Stress increases my fatigue” but were interested to know if the opposite was true as well
What could other people do to help reduce your fatigue	Participants wanted to know if there were things other people could do to reduce fatigue

**Table 5-3 - Items Removed from Second Draft of the Fatigue Impact and Severity Self-Assessment by Focus Group 2**

<b>Item Number</b>	<b>Item Descriptor</b>	<b>Reason for Removal</b>
11	Fatigue interferes with my ability to do things I would like to do	Redundant with questions 11 and 12 and all questions ask about “doing things”
24	Fatigue interferes with my ability to deal with unexpected things.	Participants felt the question was hard to understand or explain and would be hard for users to answer
28	I feel weak when I am fatigued	Participants felt that this question may be redundant with a definition of fatigue likely including weakness

**Table 5-4 - New Items Added to the Fatigue Impact and Severity Self-Assessment by Focus Group 2**

<b>Item Added</b>	<b>Reason for Addition</b>
A two-part question; specifically “Does your level of fatigue change depending on the day of the week” then if yes answer, “On which day of the week is your fatigue the greatest?”	Participants wanted to tease out differences in fatigue on weekdays versus weekends
Fatigue interferes with my ability to take care of myself (examples: Dressing, eating, bathing, brushing my teeth/hair, toileting etc)	Participants felt self-care was underrepresented in the measure
I use adaptive equipment to manage my fatigue (examples: a walker, manual wheelchair, power wheelchair etc)	Participants felt that adaptive equipment was often used as a result of or to combat fatigue experienced and that this should be included in the measure
What else could you do to reduce or manage your fatigue?	Participants wanted this measure to stimulate some thought about other methods that individuals could use to reduce or manage their fatigue

*Phase 3 – Drafting of Scale*

The participants in the second focus group suggested the including examples of fatigue and separating fatigue from pain in the definition for the scale. The final definition was drafted from this input and was set as:

“Please answer the following questions about your experience with fatigue. For the purposes of this questionnaire we would like you to think about fatigue in terms:

- physical tiredness,
- muscle soreness
- exhaustion of your muscles and body
- or any related feeling

When answering the questions, please try to focus on fatigue as it is defined above and not pain you may experience that is different from muscle soreness.”

At this time, the response options were set to a 1 to 5 Likert scale for the Impact and Management Profiles, allowing for a neutral option and anchors provided on each number from “Completely Disagree” to “Completely Agree”. This scale was chosen to align with the scaling requirements for a discriminative measure, specifically, a short distribution of choices that have a uniform interpretation to all users. Lozano and colleagues (2008) have demonstrated that the optimal scale has between 4 and 7 points to maximize both validity and reliability. Furthermore, Weng (2004) demonstrated that anchoring each response option resulted in higher test-retest reliability scores. The Severity Profile was given a variety of scaling options as related to individual questions where appropriate. Finally, a timeframe of 7 days was added to the questionnaire to orient users to consider the same timeframe when completing the questionnaire. The final scale (Appendix 5-I) that was circulated for expert review and comment after the focus groups contained the organization and specification of the profiles and attributes measured in each profile, the definition of fatigue, and the 1 to 5 Likert scale.

#### *Phase 4 - Feasibility Testing*

Five feasibility questionnaires were returned completed; 2 individuals withdrew from the project - one due to the time required to complete the questionnaire and one due to feeling overwhelmed by the impact of fatigue on their daily life as a result of reading the questionnaire. The remaining 3 questionnaires were not returned for unknown reasons. In response to a question asking about the ease with which the questionnaire was answered, 3/5 responded that it was easy or not too difficult, with one participant needing help understanding the meaning of some words. The final participant found the impact section of the questionnaire very confusing. When asked to specify which questions were confusing, this participant elaborated by stating he or she felt that the questions in the impact section were only relevant to individuals who were ambulatory. Four of the 5 participants indicated that the response options were appropriate given the questions that were being asked. The participant who found the questionnaire confusing felt the options were limited and this may have resulted in the confusion for the impact questionnaire. This participant felt ‘moderately agree’ was not the same as ‘somewhat agree’ and this change would have made the questionnaire more applicable to them. When asked to make any additional comments on the questionnaire, the participant who had trouble completing the questionnaire reiterated that the severity profile, management scale and additional questions section were easily understood and helpful; another participant reported their belief that many teens with CP will benefit from this questionnaire. As a result of these findings, no changes were made to the FISSA. It was decided that the comment regarding the scaling options of “moderately” and “somewhat” was purely a semantics difference that was not likely to change how the majority of participants answered the questionnaire. It was also felt that there was not enough information provided about what specifically was confusing within the impact section in order to make any changes.

#### *Discussion*

A new fatigue scale, the FISSA, was created using a variety of methods to ensure relevance to the users of the scale, intended to be both individuals with CP and their healthcare providers. The review of fatigue measures allowed for inclusion of items known to be related to fatigue in CP and other conditions that may have a similar

experience of fatigue, such as multiple sclerosis or Parkinson's disease. The interviews described in Chapter 4 provided a client-centered approach to item generation and selection to enhance the applicability of the questionnaire to individuals (even more specifically, youth and young adults) with CP. The healthcare providers who participated in the focus groups offered an expertise-based method of reducing items from and adding items to the scale to balance respondent burden while still retaining a complete and relevant set of items. After the refinement of the items comprising the scale, a final version of the measure was drafted, including a definition of fatigue, specification of the attributes being measured (and organization of items into profiles specific to these attributes), specification of a timeframe for response and setting of the final scaling for item responses. Finally, it was important to pilot test the FISSA in a sample of youth and young adults with CP to ensure the questions were easily understood and completed by potential users of the scale. One participant posed the question "Why do my muscles ache when I am fatigued?" to her doctor as a result of reading and completing the questionnaire; this provides some evidence that the FISSA is prompting individuals to think about fatigue and the consequences of activity, which may contribute to the development of self-awareness.

The results from the feasibility questionnaire did not result any changes to the FISSA, although one individual did find the Impact Profile confusing to complete, particularly as it related to ambulatory status. The FISSA was designed with consideration given to ambulatory status; it is likely that fatigue is experienced differently and has varying impact on individuals with different ambulatory status. Specifically, the FISSA was designed to measure fatigue in individuals with some ambulatory ability (GMFCS level I-IV), which may explain why this individual had difficulty in completing this questionnaire. The questionnaires were intended to be distributed only to those individuals classified as GMFCS Level I-IV; however, it appears in this case that one of the participants who received the questionnaire was classified as level V. After the validation of the FISSA, the measure may be useful in defining profiles of fatigue for individuals classified as different GMFCS levels as it is likely that fatigue impacts individuals with different functional abilities in distinctive ways.



*Relevance to Thesis*

The use of three separate sources of item generation (i.e. literature on measurement of fatigue in other neurological populations, the voices of youth and young adults with CP and the healthcare providers who serve these individuals) enhances the legitimacy of the FISSA for the population with CP. Furthermore, the scaling of the items were tailored to the requirements for the intended purposes of the scale (as outlined in Chapter 1, and specifically Table 1-1) which adds to the robustness of the measure. The use of healthcare professionals in reducing the number of items ensured that the FISSA retained enough items to still have a high degree of content validity; specifically, the goal was that the measure would be representative and capture all the elements of the construct of interest, fatigue among individuals with CP. The feasibility testing was, in itself, another part of assessing the content validity, also aiding in understanding the burden associated with responding to the questionnaire.

A well-constructed, client-centered scale, such as the FISSA, to measure fatigue in a population living with CP, may contribute to a better understanding of an individual's experience of fatigue, to collaborative goal setting and intervention planning by clinicians and their clients. Furthermore, this measure has the potential to enhance the development of self-awareness and self-management processes that will be necessary for navigation of adult healthcare services. The next chapter describes the testing of the psychometric properties of the FISSA for use with youth and young adults (aged 14 to 31 years) with CP.

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## **Chapter 6: Determining the Psychometric Properties of a New Clinical Measure of Fatigue for Adolescents and Young Adults with Cerebral Palsy: The Fatigue Impact and Severity Self-Assessment**

### *Introduction*

The Fatigue Impact and Severity Self-Assessment (FISSA) is a newly constructed self-report questionnaire designed to measure fatigue in individuals with cerebral palsy (CP). The content and construction of the measure was based on a literature review, consultation with healthcare professionals and interviews with youth and young adults with CP. A detailed description of the construction of the questionnaire can be found in Chapter 5 of this dissertation.

Before a new scale can be used to measure a variable of interest, validity and reliability assessments must be made to ensure the scale is appropriate for use as it is intended. Validity is a scientific property that can be used to establish how well a tool measures the variable it is intended to measure (Nunnally & Bernstein, 1994) and reliability can be defined as the ability of a measure to give consistent scores on repeated assessments in the absence of change to the characteristic being studied (Portney & Watkins, 2000). The purpose of a measurement tool is integral to its construction and validation (Kirshner & Guyatt, 1985). The FISSA was developed primarily as a discriminative tool and as such the validation of the measure will include an assessment of the tool's factor structure and discriminant validity.

The underlying structure of a tool (or factor structure) is important to determine prior to use, as a means of understanding how many constructs of interest are represented within the scale. A principal component analysis can be used to identify complex interrelationships among items of the scale and group those items into factors that represent unified concepts within a scale. The factors then represent distinct aspects of the overall construct of interest, which is fatigue in this case. In order to determine the discriminative validity (or "known groups" validity) of the new tool it is administered to groups expected to differ based on other existing measures and evaluate the ability of the measure to detect differences in those groups (Kirshner & Guyatt, 1985). Specifically, differences in levels of fatigue in groups varying in level of severity of CP, pain and physical activity will be used to determine the validity of the FISSA. The Gross Motor

Function Classification System (GMFCS) levels represent distinctly different functional capabilities and limitations for initiating and achieving movement (Palisano, Rosenbaum, Walter, Russell, Wood & Galuppi, 1997). It is possible that fatigue severity and impact may be different depending on the activities someone is able to perform and the difficulty they experience in execution of their daily activities. In addition, there is also some evidence that fatigue can provoke pain in adults with CP (Schwartz, Engel & Jensen, 1999). Finally, a weak relationship between fatigue and physical activity has been demonstrated (Nieuwenhuijsen, van der Slot, Dallmeijer, Janssens, Stam, Roebroek et al., 2011; van der Slot, Nieuwenhuijsen, van den Berg-Emons, Bergen, Hilberink, Stam et al., 2012). Together this literature has informed the specific hypotheses that were used to determine the discriminate validity of the FISSA.

In regards to establishing reliability; a discriminative measure should show large but stable intersubject variation (Kirshner & Guyatt 1985) to be able to discriminate between individuals yet remain stable in the absence of change. The internal consistency of a tool is important to assess, as it is a measure of how the items on test represent various aspects of the same characteristic and nothing else. Internal consistency coefficients range from 0 to 1 with higher values representing higher levels of internal consistency. Additionally, evaluating the test-retest reliability is important when determining the psychometric properties of a scale. Test-retest reliability is a measure of the stability of the scale over time and is based on assessments with the same measurement tool made on different occasions when change is not expected (Finch, Brooks, Stratford & Mayo, 2002). Similarly test-retest reliability coefficient values can range from 0 to 1 and higher values represent higher levels of reliability.

#### *Purpose*

Currently, there is no measurement tool that has been validated to measure fatigue in a population of adolescents and young adults living with CP. The purpose of this study was to establish the validity and reliability of the FISSA.

#### *Primary Objectives*

1. Determine the factor structure of the FISSA.
2. Determine the discriminative validity of FISSA by testing 5 hypotheses.

- a. Participants in GMFCS levels II and III will experience more fatigue than individuals in GMFCS level I.
  - b. Participants in GMFCS levels IV and V will experience more fatigue than individuals classified as GMFCS level II and III.
  - c. Individuals with a higher impact of pain will experience greater fatigue than those with less impact of pain on their daily lives.
  - d. Individuals with a higher severity of pain will experience greater fatigue than those who experience less severe pain.
  - e. Individuals with less physical activity will experience more fatigue than individuals with more physical activity (excluding outliers).
3. Determine the internal consistency and test-retest reliability of the FISSA.
    - a. As a result of the rigorous item selection and reduction criteria, it is hypothesized that the internal consistency of the FISSA will be high (above 0.85).
    - b. It is anticipated that fatigue is a relatively stable phenomenon, but given the self-report nature of the FISSA, it is hypothesized that the test-retest reliability of the FISSA will be moderate (above 0.70).

#### *Secondary Objective*

1. Determine the test-retest reliability of the pain questionnaire.
2. Determine the test-retest reliability of the exercise questionnaire.

#### *Methods*

##### *Design*

This study was a measurement development study that revealed the factor structure of the FISSA and assessed the discriminate validity, internal consistency and test-retest reliability of the FISSA for assessing fatigue in youth and young adults with CP. Ethical Approval was obtained from the Health Sciences Research Ethics Board at Western University (Appendix 6-A).

##### *Participants and Sampling*

A total of 367 youth and young adults with CP aged 14-31 years were contacted as potential participants for the study from participating children's rehabilitation centres

in Ontario, previous research studies about fatigue conducted by the primary investigator, existing facebook groups for individuals with CP and through the Ontario Federation for Cerebral Palsy (OFCP) website and newsletter. A recruitment notice was also sent to members of the Canadian Physiotherapy Association's Pediatric Division through an email newsletter; however, this method did not facilitate any study participation. Participants recruited from the children's treatment centres were initially identified as a potential participant by an administrative staff member through the database maintained at each rehabilitation centre. Potential participants identified from the children's treatment centres were mailed a survey package containing a letter of information (Appendix 6-B), survey booklet and a stamped addressed envelope to return the survey. Individuals who participated in a previous study about understanding the fatigue experience and who had provided consent to be contacted about future research were sent study information either by email or post depending on their preference. Advertisements were posted on the OFCP website and in their monthly newsletter as well as on existing facebook support groups for individuals with CP and in an email to members of the Canadian Physiotherapy Association Pediatrics Division (Appendix 6-C). Participants recruited through the online support groups, OFCP website or newsletter were provided with a study ID number and a link to complete the survey online. All participants were provided with a \$10 iTunes gift card as a token of appreciation for their participation in the study.

Study eligibility criteria included individuals aged 14 to 31 years of age and classified as GMFCS levels I-IV. Due to the nature of the surveys, the potential participants were limited to those who were English speaking and who could comprehend the questionnaires and respond to the questions with some degree of independence. Surveys returned that were completed entirely by parental proxy were excluded from the study. A small subset of the sample was asked to complete a smaller survey package a second time within two weeks to assess the test-retest reliability of the scale.

#### *Data Collection Tools*

In addition to the FISSA (construction and scaling can be found in Chapter 5 of this dissertation), a self-report version of the Gross Motor Function Classification System – Expanded and Revised Version (GMFCS-ER) (Palisano, Rosenbaum, Bartlett &

Livingston, 2008) was used to collect information on severity of CP (Appendix 6-D). The GMFCS-ER is an ordinal classification system consisting of 5 levels that describe the gross motor function of individuals with CP based on their self-initiated movement in the home, at school and in the community. The distinctions between levels are based on functional abilities, the need for assistive technology, including hand-held mobility devices (walkers, crutches, or canes) or wheeled mobility (Palisano et al., 2008). Evidence of the content validity of the GMFCS-ER has been established (Palisano et al., 2008); and recently, the reliability of the 12-18 year age band has been confirmed for ambulatory youth (Gorter, Slaman, Bartlett, & van den Berg-Emons, 2011). A simple demographic questionnaire was used to obtain self-reported distribution of involvement as well as information on age and sex (Appendix 6-E).

Furthermore, information was collected on prevalence, severity, impact and location of pain (Doralp & Bartlett, 2010) (Appendix 6-F). The pain questionnaire used in this study has not yet been objectively validated. The questionnaire was developed through expert opinion for the Adolescent Study of Quality of Life, Mobility and Exercise (ASQME). The questionnaire consists of 4 questions, participants were first asked to respond to the question “Over the past month, have you experienced physical pain?” (yes or no). Those responding “yes” were then asked “Please indicate how severe your pain was over the past month, on average,” response options range from 1 (very little pain) to 10 (extremely painful). Next, they were also asked to indicate “How much the pain gets in the way of your daily activities over the past month” with responses ranging from 1 (does not get in the way at all) to 10 (unable to carry out activities because of the pain). Finally, they were asked to indicate the regions in which they experienced pain on a body map (Doralp & Bartlett, 2010). Data were analyzed based on severity and impact for known groups validity of the FISSA and both of these plus the specific body regions were analyzed for the test-retest reliability of the measure.

An exercise questionnaire was also developed by the ASQME team at CanChild was used to collect information on physical activity (Brunton & Bartlett, 2010) (Appendix 6-G). The exercise questionnaire collected information on the amount (both time and frequency), type and intensity of physical activity in the previous week. Total minutes of activity (average time spent doing the activity multiplied by the number of

times the activity was performed over the week) were calculated for each of the different intensities (light, medium and hard) and used in both the known groups validity and test-retest analyses. Similar to the pain questionnaire, the exercise questionnaire has not yet been validated. The final page of the survey booklet asked for information regarding the amount of assistance required to complete the survey. Due to the fact that the pain and exercise questionnaires had not previously been validated for use (other than content validity through expert opinion) a secondary objective of this study was to assess the test-retest reliability of the pain and exercise questionnaires.

#### *Data Collection Procedures*

The study followed a modified Dillman method and participants were contacted either 2 or 3 times depending on their participation (Dillman, 2000). All potential participants initially received a full survey package (or an email with the letter of information and survey link) with the \$10 iTunes gift card. In an effort to increase the number of respondents, a reminder letter was mailed to each potential participant approximately two weeks following the initial package mailing. All individuals who had not yet returned the survey two weeks after the reminder letters were sent (four weeks following the initial mailing) received a second full questionnaire package. To assess the test-retest reliability of the fatigue scale as well as the exercise and pain questionnaires, a small subset of the sample were asked to complete the fatigue, pain and exercise portions of the survey a second time approximately two weeks after their initial response.

#### *Analysis*

An exploratory factor analysis was conducted using principal components analysis on the final data set obtained from the FISSA. Prior to conducting the factor analysis, the data were screened for suitability. The internal consistency of the individual items of the FISSA was analyzed to determine if all 32 items would remain as part of the scale undergoing factor analysis. Non-parametric Spearman's Rho correlations between the items were also performed to determine if the data was adequate for factor analysis. Specifically, Comrey and Lee (1992) suggest that the majority of the correlations between items should be between 0.3 and 0.9 to indicate adequacy for factor analysis. The Kaiser-Meyer-Olkin measure of sampling adequacy and Bartlett's test of sphericity



were explored to ensure that the use of factor analysis was appropriate for the data collected.

During the extraction phase, Kaiser's criterion, Cattell's Scree test and parallel analysis were used to determine the number of factors extracted and retained for the remainder of the analysis. Specifically, Kaiser's criterion suggests retaining all factors with eigenvalues greater than one, the Scree test involves plotting the eigenvalues to find the point at which the curve becomes horizontal to determine the number of factors (Cattell, 1966), and finally, the parallel analysis allows for retention of only the number of factors with eigenvalues that exceed corresponding values from a random data set of random correlation matrices based upon the number of variables in the measure and the number of participants in the sample. Parallel analysis represents a more accurate method of factor extraction compared to Kaiser's criterion and the Scree test (Zwick & Velicer, 1986). The remaining factors were then rotated, using the direct oblimin approach as it was hypothesized that the factors were related, to better understand the meaning of each factor. A final model was then created to explain the underlying structure of the questionnaire and to understand how the factors were acting in the scale.

The known groups validity of the FISSA was examined by investigating the difference in fatigue severity by GMFCS level, the difference in fatigue between high and low pain (represented by both pain severity and impact of pain on daily activities) and physical activity (total minutes of light, medium and hard exercise) groups. The GMFCS levels were grouped together to increase the subgroup sample size and consisted of individuals who self-classified as level I separately, levels II and III were grouped together and levels IV and V formed the third group. The Kruskal-Wallis statistic was used to compare the difference in fatigue between groups of GMFCS levels. A median split was applied to the pain impact and severity data and a Mann-Whitney U was used to assess the difference in fatigue by either high or low pain impact or severity. The exercise data was split at the 75<sup>th</sup> percentile (due to a median of zero across all intensities of exercise) and a Mann-Whitney U was used to assess the difference in fatigue by high or low physical activity.

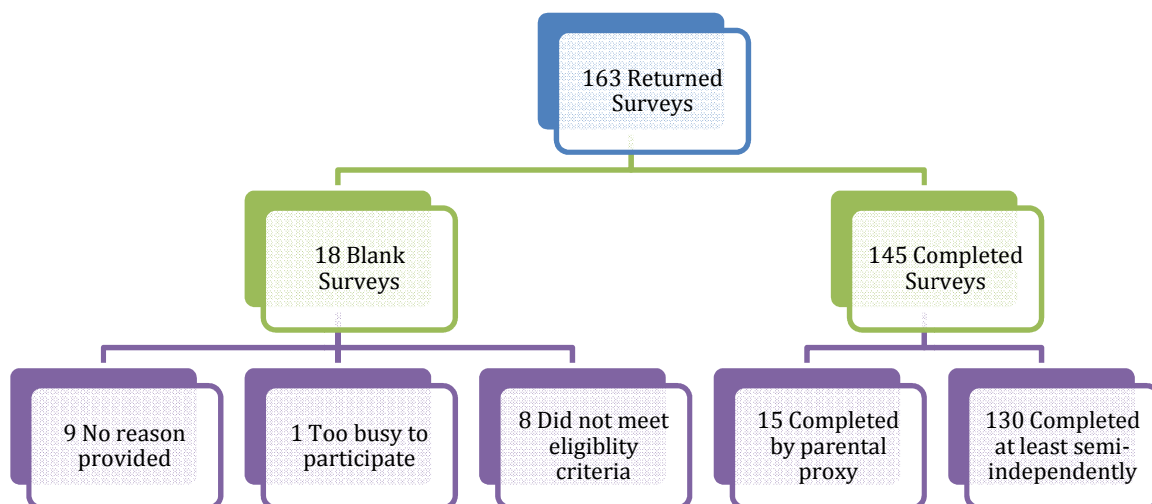
Internal consistency of the FISSA was assessed through the use of Cronbach's alpha. Test-retest reliability of the FISSA, pain and exercise questionnaire were analyzed

using an intraclass correlation, specifically the ICC (3,1) model was used (Portney & Watkins, 2000).

The sample size required for factor analysis has been reported to be anywhere from 5 to 10 participants for every one item in the analysis (Kass & Tinsley, 1979; Nunnally 1978). As a result our target sample size was 160 participants (5 participants per item, 32 items).

### *Results*

Of the 367 individuals contacted, 163 questionnaires were returned over the course of the study, for a response rate of 44.4%. A final convenience sample of 130 youth and young adults with CP participated in the study by returning a questionnaire (completed at least semi-independently) to the study team. Figure 6-1 provides a detailed breakdown of the returned questionnaires, as not all returned questionnaires were included in the study analysis. Table 6-1 contains the participant characteristics and demographic information of the 130 participants.



***Figure 6-1 – Breakdown of Inclusion and Exclusion of Returned Surveys***

**Table 6-1 Participant Characteristics and Demographic Information of the Sample**

Characteristic		Total (n=130) n (%)
Sex*	Male	61 (47%)
	Female	68 (53%)
Age	Mean, years (SD)	18.9 (4.5)
	Median	17
	Range	14-31
GMFCS Level <sup>†</sup>	I	34 (27%)
	II	39 (32%)
	III	21 (17%)
	IV	18 (14%)
	V	12 (10%)
Distribution of Involvement <sup>††</sup>	Monoplegia	6 (5%)
	Hemiplegia	31 (25%)
	Diplegia	44 (35%)
	Triplegia	11 (9%)
	Quadriplegia	34 (26%)

\*One participant did not report their sex, <sup>†</sup>Six Participants did not report their GMFCS Level, <sup>††</sup>Four participants did not report their distribution of involvement

#### *Data Screening for Factor Analysis*

The internal consistency of the 32 items together was 0.95. Each item was then scanned to determine if removal would increase the overall alpha level. One item was found to slightly increase the alpha if deleted (*I pace my physical activities to manage my fatigue*; 0.955); however, given the very slight difference in Cronbach's alpha the item was retained in the measure at that point. The non-parametric Spearman's Rho correlations did not reveal any correlations above 0.9 and 62% of the correlations were above 0.3 (see Appendix 6-H for correlation matrix) indicating adequacy for factor analysis.

The Kaiser-Meyer-Olkin measure of sampling adequacy was demonstrated to be 0.90, which is considered to be excellent in indicating the use of factor analysis (Kaiser, 1974). Bartlett's test of sphericity was found to be significant ( $p < .001$ ) again indicating that the data set was appropriate for use of factor analysis.

### *Exploratory Factor Analysis*

Following the initial item screening, all 32 items remained in the scale for factor analysis. Based on Kaiser's criterion of retaining factors with eigenvalues greater than one, seven factors would have been extracted and retained in the analysis. However, when Cattell's Scree plot (Appendix 6-I) and parallel analysis were used to determine the number of factors to be extracted, only two factors (Impact of Fatigue on Daily Living and Management and Activity Modification) were retained in the factor structure of the FISSA and together they explained 48.7% of the variance after rotation. The Impact factor explained 42.5% of the variance and the Management and Modification Factor explained an additional 6.2% of the variance in fatigue scores. The two factors were indeed related with a correlation between the factors of 0.57. Table 6-2 provides a summary of the results of the exploratory factor analysis for the FISSA. Item loadings found to be 0.4 or greater were considered significant (Ismail, 2008). In total 17 items loaded on the first factor (Impact of Fatigue on Daily Living) and 15 items loaded on the second factor (Management and Activity Modification). One item (*Fatigue interferes with my participation in social activities*) loaded moderately (and fairly equally) on both factors; however, it is considered to be part of the first factor given its slightly larger loading. Finally one item (*Fatigue interferes with my ability to control my mood*) did not load sufficiently on either factor and was removed from the scale, resulting in the final 31-item version of the FISSA (Appendix 6-J).

**Table 6-2 Summary of Exploratory Factor Analysis results for the Fatigue Severity and Impact Self Assessment (N=130)**

Item	Rotated Factor Loadings	
	Factor 1 – Impact of Fatigue on Daily Living	Factor 2 – Management and Activity Modification
Fatigue interferes with my ability to do things on my own	<b>.77</b>	.03
I use adaptive equipment to manage my fatigue	<b>.77</b>	-.27
Fatigue interferes with my ability to take care of myself	<b>.73</b>	-.07
Fatigue interferes with my ability to move around indoors	<b>.73</b>	.12
Fatigue interferes with my ability to get outside of my house	<b>.70</b>	.10
I have had to reduce my work responsibilities outside my home because of fatigue	<b>.70</b>	.06
Fatigue interferes with my ability to move around in my community	<b>.70</b>	.17
Rate your average level of fatigue for the past week	<b>.70</b>	.18
I have had to reduce my responsibilities at home because of fatigue	<b>.69</b>	.08
Rate your level of fatigue on the day within the last week that you felt the least fatigued	<b>.68</b>	-.09
On average, how much of the day do you feel fatigued	<b>.68</b>	.07
Rate your level of fatigue on the day within the last week that you felt most fatigued	<b>.64</b>	.17
Fatigue interferes with my general everyday activities	<b>.57</b>	.31
Fatigue interferes with my ability to start things	<b>.50</b>	.18
For how many days last week did you feel fatigued at least part of the day	<b>.50</b>	.24
Fatigue interferes with my ability to finish things	<b>.46</b>	.36
Fatigue interferes with my participation in social activities	<b>.43</b>	<b>.41</b>
Fatigue interferes with the length of time I can be physically active	.12	<b>.72</b>
I limit my physical activity to manage my fatigue	-.05	<b>.71</b>
My motivation to do physical activities	.05	<b>.70</b>
I stop and rest during activity to manage my fatigue	-.01	<b>.68</b>
Fatigue interferes with my balance and coordination	.03	<b>.68</b>

I pace my physical activities to manage my fatigue	-.21	<b>.66</b>
Fatigue interferes with my motivation to participate in social activities	.22	<b>.58</b>
Fatigue interferes with my leisure and recreational activities	.26	<b>.55</b>
My muscles ache when I am fatigued	.13	<b>.55</b>
I think about fatigue when I plan my day	.04	<b>.53</b>
Stress increases my fatigue	.31	<b>.45</b>
Fatigue interferes with my enjoyment of life	.34	<b>.44</b>
Long periods of inactivity increase my fatigue	.10	<b>.42</b>
Fatigue increases my stress	.31	<b>.40</b>
Fatigue interferes with my ability to control my mood	.21	.37
Eigenvalues	13.59	1.99
% of variance explained	42.5	6.2

\*Note: Factor loadings over .40 appear in bold.

#### *Known-Groups Validity*

Table 6-3 contains the descriptive information of fatigue score by grouped GMFCS level, low or high pain, and total exercise. As hypothesized, individuals who self-classified as GMFCS level I experienced significantly less fatigue than individuals classified in any other GMFCS level (II-V) ( $p < .001$ ). Contrary to the hypothesis, individuals classified as GMFCS level IV or V did not experience significantly more fatigue when compared to individuals classified as GMFCS level II or III ( $p = .063$ ). However, as hypothesized, individuals with higher pain (both impact and severity) reported higher fatigue scores ( $p < .001$ ). There were no significant differences in fatigue levels for individuals who had more physical activity at any level of intensity ( $p = 0.76$  light,  $p = 0.22$  medium and  $p = 0.74$  hard).

**Table 6-3 Summary of Fatigue Scores for Known Groups Validity Testing**

Construct	Subgroups <sup>†</sup>	FISSA Score (median, range)
Functional Level	GMFCS Level I (n=35)	69 (30, 122)
	GMFCS Level II and III (n=63)	96 (37, 147)
	GMFCS Level IV and V (n=39)	106 (38, 146)
Pain Severity*	Low Pain Severity $\leq$ 50 <sup>th</sup> percentile (n=84)	75 (30, 146)
	High Pain Severity $\geq$ 51 <sup>st</sup> percentile (n=60)	109 (39, 157)
Pain Impact*	Low Pain Impact $\leq$ 50 <sup>th</sup> percentile (n=79)	75 (30, 146)
	High Pain Impact Severity $\geq$ 51 <sup>st</sup> percentile (n=63)	108 (39, 157)
Light Exercise*	High Exercise $\geq$ 76 <sup>th</sup> percentile (n=37)	96 (36, 147)
	Low Exercise $\leq$ 75 <sup>th</sup> percentile (n=105)	92 (30, 157)
Medium Exercise*	High Exercise $\geq$ 76 <sup>th</sup> percentile (n=44)	96 (36, 144)
	Low Exercise $\leq$ 75 <sup>th</sup> percentile (n=98)	92.5 (30, 157)
Hard Exercise*	High Exercise $\geq$ 76 <sup>th</sup> percentile (n=31)	97 (43, 136)
	Low Exercise $\leq$ 75 <sup>th</sup> percentile (n=109)	93 (30, 157)

\*Boxplots of distributions of pain severity and impact and light, medium and hard exercise are contained in Appendix 6-K); †Note: The number of participants in each subgroup varies

#### *Internal Consistency*

The Cronbach's alpha for the entire questionnaire consisting of 31 items was 0.95. Independently, the Cronbach's alpha for the Impact of Fatigue on Daily Living factor (17 items) was 0.94 and the Management and Activity Modification factor (14 items) demonstrated a Cronbach's alpha of 0.90.

#### *Test-Retest Reliability*

The average test-retest interval in this study was 36 days (range 13 to 87 days). Although the test-retest packages were mailed to participants two weeks after receiving their initial response to the survey, test-retest responses returned to the investigators were

variable in length of time. A total of 31 individuals returned their test-retest package completed. The FISSA demonstrated an  $ICC(3,1) = 0.75$  (95% CI 0.54-0.87). The pain questionnaire demonstrated an  $ICC(3,1) = 0.73$  (95% CI 0.50-0.86) for the impact component, an  $ICC(3,1) = 0.78$  (95% CI 0.59-0.89) for the severity component and an  $ICC(3,1) = 0.82$  (95% CI 0.66-0.91) for the body regions that were painful. The exercise questionnaire was not shown to be reliable as evidenced by low and non-significant correlations between testing occasions (light exercise  $ICC(3,1) = -0.015$ ,  $p=0.53$ , medium exercise  $ICC(3,1) = 0.070$ ,  $p=0.36$ , hard exercise  $ICC(3,1) = 0.21$ ,  $p=0.12$ ).

### *Discussion*

The FISSA was created to examine the impact, severity and management of fatigue for youth and young adults with CP. This validation study demonstrates that the 31-item FISSA contains two related factors (impact of fatigue and management and activity modification related to fatigue) that adequately explain 49% of the variance in fatigue experienced by these individuals.

When the FISSA was originally designed, it was organized into three sections and it was hypothesized that three components would be extracted with the factor analysis. The three anticipated components of the FISSA included: impact, severity and management of fatigue. It is clear from the analysis that the factors of impact and management are present in the scale and that these factors are indeed interrelated as demonstrated by the oblimin rotation. The third anticipated factor of severity appears to be very closely linked to the impact factor and all of the items proposed to be part of that factor loaded highly (above 0.50) on the impact factor. The two-factor solution (containing 31 items) demonstrated high internal consistency of 0.95 and good test-retest reliability at 0.75.

A known groups validation approach was used to provide evidence of validity of the FISSA because there is currently no accepted measure available to provide information about fatigue in individuals with CP. The FISSA was able to discriminate between groups expected to experience more fatigue (individuals classified as having a more severe motor disability according to the GMFCS and individuals experiencing a high degree of pain).



Many individuals (regardless of GMFCS level) live with fatigue as a daily consequence of physical activity (Jahnsen, Villien, Aamodt, Stanghelle & Holm, 2003) and there are limited strategies available to these individuals to effectively manage their fatigue. Svien and colleagues (2008) suggested there is a great need to understand how fatigue is impacting a client's life and their ability to accomplish their activities of daily living. The FISSA was created, with this goal in mind, to assist with identifying individuals who are experiencing fatigue related to CP as a method of fostering a clinical discussion between clinicians and their clients about fatigue and possible management strategies. The FISSA provides a preliminary description of activities that fatigue interferes with, an overview of the severity of fatigue experienced by the individual and a report on the management strategies that an individual may or may not have tried to effectively limit fatigue. Early identification of fatigue and the activities that are affected by fatigue may assist with intervention development to interrupt the cycle of deconditioning described in individuals with CP (Tosi, Maher, Moore, Goldstein & Aisen, 2009) both on the clinical level and in future research. The progression of the activity limitations in CP as a result of fatigue are thought to be linked to functional decline that can affect independence in adult life (Tosi et al., 2009). The FISSA allows for individualized identification of the activities of daily living that may be compromised by fatigue and, once identified, strategies and adaptations to increase independence in specific areas may be more easily conceivable and available to these individuals. This self-assessment can be used, on an individual basis, to streamline a clinical conversation to the salient fatigue-related issues. The FISSA may help clinicians effectively discuss possible solutions and strategies to limit or manage the impact of fatigue while being mindful of limited time and resources in the clinical setting.

### *Limitations*

Our initial sample size calculation indicated the use of a sample size of at least 160 individuals. We did not reach this sample size in the current study; however, several authors have questioned the 5-10 participant per item ratio for indicating sample size related to factor analysis. Arrindell and van der Ende (1985) concluded that changes to the ratio of participant to item made little difference in the stability of the factor solution compared when parallel analysis was used as the primary extraction method. In addition,

Guadagnoli and Velicer (1988) argue that factors that contain four or more loadings of 0.60 or higher are reliably extracted regardless of the sample size, in the case of the FISSA, both factors readily met this criterion.

Two items (and follow-up questions) related to severity (“*Does your level of fatigue change depending on the time of day? Follow-up: What time of day is your fatigue the worst?*” and “*Does your level of fatigue change depending on the day of the week? Follow-up: On which day of the week are you most fatigued?*”) were not included in the factor analysis due to the more descriptive nature of these questions. These questions have now been moved to the additional questions section in the final version of the FISSA (Appendix 6-J).

There was a longer than anticipated and a wide range between participants in the test-retest interval (an average of almost 5 weeks). The study was initially designed to have a 2 to 3 week interval between dates of administration for the test-retest analysis; however, this was not easily controlled. Although the timeframe was larger than we had anticipated, it is unlikely that these individuals were undergoing any intervention specifically aimed at addressing their fatigue because there are no established strategies available for individuals to manage fatigue. There is the possibility of some seasonal changes that may have affected the individualized experience of fatigue during this timespan; however, it was generally assumed that the fatigue level was fairly stable and representative of the individuals’ typical life over this period of time.

Finally, information related to physical activity was collected and in the design phase of the study it was hypothesized that individuals with more physical activity would experience less fatigue. However, the questionnaire used to assess physical activity was not shown to be reliable in the test-retest phase of this study and it is therefore not surprising that no significant differences were detected in fatigue levels for this portion of the known-groups validity testing. As a result the relationship between fatigue (as measured by the FISSA) and physical activity level remains unclear, it would be helpful to address this relationship with more reliable measures in future studies.

#### *Relevance to Thesis*

The FISSA represents a validated and reliable tool that can be used to identify individuals who have a significant amount of fatigue impacting their lives. Furthermore,

once completed, the FISSA highlights the specific domains of life that fatigue is affecting the most and may help initiate conversations between clients and their clinicians to brainstorm possible solutions to decrease the impact of their fatigue. The previous chapters of this dissertation demonstrated the client-and clinician-centered approach used to develop this measure. The results of this chapter provide the necessary psychometric support for the measure to be integrated into clinical use for identification of individuals impacted by fatigue and facilitate collaborative goal setting or intervention planning by clinicians and their clients. Furthermore, this measure has the potential to enhance the self-awareness and self-management processes for youth with cerebral palsy that will be necessary for navigation of adult healthcare services following transition out of pediatric care.

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## **Chapter 7: Summary, Clinical Implications and Future Research Directions**

### *Summary and Clinical Implications*

The cumulative aim of the studies contained in this dissertation were to further the understanding of the experience of fatigue for youth and young adults with cerebral palsy (CP). The consensus definition of CP highlights the non-progressive nature of the lesion in the brain that gives rise to the motor impairment experienced by individuals with CP. However, that is not to say that the manifestations of the lesion are static. Change in the appearance of CP over time can be caused by development of secondary impairments to the musculoskeletal system. The primary focus of this work is the secondary impairment of fatigue, with lesser focus on pain, and the impact these impairments have on the daily activities of adolescents and young adults living with CP. It is crucial to further the current understanding of the impact secondary impairments for youth and young adults with CP if clinicians, and other care providers working with these individuals, want to be effective in planning for transition from the pediatric to adult healthcare settings and promoting self-management of their condition. More information regarding the experience, impact and severity of fatigue and the relationship between fatigue and pain will allow clinicians and researchers to be better equipped when planning interventions and enhancing the development of youth and young adults with CP.

The first study contained in this dissertation is a critical review focusing on the ambiguity in the literature that leads to the conclusion that individuals with CP can experience both more or less fatigue compared to their peers without a disability. The outcome (more or less fatigable) is largely dependent on how fatigue is defined and measured. When measured in isolated laboratory studies with strength normalized to the amount of force produced, individuals with CP appear to fatigue less than their non-disabled peers as a result of their lower force generating capacity (Stackhouse, Binder-Macleod & Lee, 2005; Moreau, Li, Geagan & Damiano, 2008); however, I question the usefulness of comparing fatigability in an isolated manner instead of using a functional task that depends on an absolute load (i.e. the individual's own body weight). In addition, the isolated and controlled nature of laboratory studies of fatigue do not reflect the chronicity or impact of fatigue for individuals with CP and may be confounded by structural and functional differences in the mechanisms of muscle action for individuals

with CP. Interpreting the decline in force capacity observed with fatigue by normalizing it to maximal force generating capacity removes the context of the impact of that fatigue almost entirely. These individuals who are already shown to be weaker than their peers are experiencing a large reduction in muscle force during fatiguing tasks – a 42% reduction in the study by Stackhouse, Binder-Macleod and Lee (2005) and because it was statistically significantly less than the 52% percent of decline in force exhibited by their peers, it is concluded that individuals with CP experience less fatigue. However, the experience of fatigue cannot be interpreted within this context because of the lower baseline force level, and the impact of that baseline level should not be ignored. This large reduction in force-generating capacity with fatigue has the potential to have an even greater impact on the capacity to continue performing activities due to a lower metabolic reserve. In a study of walking at a self-selected pace, individuals with CP used a larger proportion of their metabolic reserve compared to their peers without a disability (Slaman, Bussman, van der Slot, Stam, Roebroek, van den Berg-Emons et al., 2013), highlighting the possible greater impact of a fatiguing activity, regardless of the normalized reduction in force. Laboratory studies of fatigue tend to focus on the primary impairment of reduced force-generating capacity while ignoring the secondary impairment of reduced endurance that is related to the onset of fatigue. It has been well documented that youth with CP have higher energy expenditures during gait when compared to their peers at similar walking speeds (Campbell & Ball, 1978). It is important that therapists recognize how to interpret the findings in the fatigue literature within the context of functional tasks that require a certain level of absolute ability to be performed successfully.

In the process of performing the critical review, it became clear that neuromuscular fatigue was a challenge for individuals with CP, with no consensus regarding the underlying mechanisms or the impact on function for these individuals. This in part due to the limited research available on the topic and due to inconsistencies among the available studies in how fatigue is defined, measured and compared. In order to enhance the clinical understanding of fatigue, a valid and reliable assessment of fatigue is necessary; however, at the outset of this program of research, there was no published

psychometric information for any fatigue scale appropriately indicating its use in a population with CP.

As a result, a search was conducted in the CINAHL and EMBASE databases for articles that assessed psychometric properties of fatigue scales in conditions that were thought to have a similar experience and impact of fatigue related to possible common neuromuscular origins. The search and subsequent article review resulted in an examination of the psychometric properties of 15 different self-report fatigue scales that had been employed in at least one of the conditions of interest (Parkinson's disease, multiple sclerosis, chronic fatigue syndrome and/or postpoliomyelitis). The 15 measures were reviewed for content and possible applicability to the population of CP. In the review, data were extracted about the reliability and validity of the scales from each primary source article. After data extraction, each measure was classified into one of three categories: adequate psychometric properties, inadequate psychometric properties and measures needing more validation efforts. Only three of the fatigue scales were classified as having satisfactory data supporting their use in a neurological condition, the Parkinson's Fatigue Scale, the Fatigue Scale for Motor and Cognitive Functions and the Neurological Fatigue Index. Each of these scales were designed and validated for a specific neurological population and were reviewed for applicability to a population with CP, yet no one single fatigue scale appeared to be entirely appropriate. The review highlighted the advantages of a diagnosis-specific fatigue scale and served as the initial stage in the creation of a new fatigue scale specific to individuals with CP, the Fatigue Impact and Severity Self-Assessment (FISSA). Items from each of the 15 identified scales were reviewed and individual items that echoed themes discussed in the fatigue in CP literature were noted for possible inclusion in the FISSA.

A phenomenological inquiry served the dual purpose of understanding the bodily experience of living with cerebral palsy as well as a client-focused method of generating items for the FISSA. Participants were asked to describe a normal day, a day in which they experienced fatigue and/or pain, and more general questions about how they experienced their body. Overall, the participants in this study described fatigue that resulted from a variety of activities and had an impact on numerous aspects of their daily lives. Self-awareness emerged as the most important theme and each participant



described or demonstrated a different level of self-awareness. Themes of adaptation, planning and restricting activities were closely related to the level of self-awareness the participants displayed. Some of the individuals interviewed were actively aware they were managing their fatigue through restricting or making adaptations to their activities or by planning their day, while others had begun to do this but were not explicitly aware of it in their description of their typical day. Finally, other participants were not yet able to find ways to manage their fatigue. The findings of this study demonstrate the need to explore the developmental course of emerging self-awareness, the need and ability of youth and young adults with CP to plan their day to mitigate fatigue. Adolescents and young adults without a physical disability generally do not have to plan their days with fatigue in mind, but it was clear that many individuals with CP have begun to make choices about their activities while considering fatigue and pain. Jones (2009) provided personal accounts of aging with CP and has stressed the need to create strong partnerships within the medical community for these individuals and the need to further the understanding of the lived experience of CP. She wrote of her own struggles to balance the effects of fatigue and pain within the context of her activities of daily living. She describes having periods of “massive fatigue” and muscle pain but never thinking this was something to see a doctor about, believing instead that she had to force herself to keep going with no other alternative (Jones, 2009). Conversations about fatigue with health care providers can start to create a space to talk about options for managing fatigue and the impact of fatigue on participation in life activities. These conversations have the power to foster emerging self-awareness and problem solving abilities that will become necessary once these youth become adults and are required to self-direct their own care.

Interestingly, many of the individuals interviewed understand the importance of maintaining participation in their daily activities but felt they had to restrict or avoid certain activities in order to be able to complete essential tasks in their lives. The participants interviewed for this study used rich descriptions of their experiences to describe a complicated and multifaceted nature of their fatigue that could be incorporated into the FISSA. Specifically, many of the items related to the Management and Activity Modification factor were created directly as a result of the interview data. I felt it was extremely important to give voice to the concerns the participants raised in the

phenomenology and generating items for the FISSA was one method of adding content validity to the FISSA while honouring the experience of the youth and young adults with CP in this study.

After generating potential items for the FISSA, two focus groups were held with health care providers (from various professions) who normally interact with individuals with CP to ensure content validity for the measure while reducing the number of items to a reasonable and relevant subset of questions. Finally, the feasibility within the population of CP was assessed by pilot testing the FISSA in a small sample of youth and young adults with CP. Overall the FISSA was well received by the youth in the feasibility testing and no changes were made as a result of the feedback provided.

The FISSA was initially created to examine the severity, impact and management of fatigue for youth and young adults with CP. It was important that the item generation and reduction phases of the creation of the FISSA were comprehensive in nature and reflected the opinions of the end-users of the scale (both clinicians and individuals with CP) as manner of incorporating “consumer” input into the scale to enhance its usefulness. This idea of having consumer input into research about fatigue experienced by individuals with CP is another recommendation provided by Jones (2009) to enhance the potential success of a project’s outcome. In this case, the careful and purposeful detail in the creation of the FISSA provided a solid infrastructure for the scale that was reflected in the strong psychometric properties demonstrated in the validation efforts described in the final study of this dissertation. The 31-item FISSA was shown to contain two related factors (impact of fatigue and management and activity modification related to fatigue) that together were able to explain 49% of the variance in fatigue experienced by individuals with CP. A known groups validation approach provided some evidence of the construct validity of the FISSA because there is currently no gold-standard measure available to provide information about fatigue in individuals with CP. When assessed the FISSA demonstrated the ability to discriminate between groups hypothesized to experience more fatigue (including individuals classified as more severe on the Gross Motor Function Classification System (GMFCS) and individuals experiencing a high degree and impact of pain). The FISSA was also shown to be reliable with high internal consistency and good test-retest reliability.

The FISSA was created to gain a better understanding of the fatigue experienced by individuals with CP and to identify individuals who have a significant impact of fatigue on their life. It is my hope that clinicians will use the FISSA to initiate conversations about the functional restrictions individuals are experiencing as a result of fatigue and use the measure as a starting point for discussion about possible fatigue management strategies. The final recommendation made by Jones (2009) was to identify and collaborate on interventions to address functional problems; the FISSA can assist with this collaboration. Clinicians can examine the responses to individual items of the FISSA as a method of identifying the specific areas of the individual's life that fatigue is impacting most, especially given the highly individualized presentation of CP. Ideally, the FISSA will then be used to facilitate collaborative goal setting and future intervention planning between clinicians and their clients and family.

#### *Future Directions*

Although the mechanistic and physiological process of fatigue was not a focus of this dissertation, the critical review performed here highlights gaps in the current literature. One important area for future study is to understand the process of fatigue for individuals with CP with a specific focus on functional tasks such as walking or standing. Additionally, an understanding of the rate of the development of, and recovery from, fatigue is needed to better understand the chronic presentation of fatigue in individuals with CP. Finally, laboratory studies designed to assess the fatigability of individuals with CP compared to a control population, should account for differences in absolute strength and altered muscle function and consider the effect of a lower force reserve and the impact that may have for the individual with CP.

The phenomenology presented in this dissertation was the first study to explore the bodily experience of living with CP. In addition to furthering the understanding of fatigue and its impact, the descriptions provided by the participants revealed other aspects of the lived experience of individuals with CP that warrant further exploration including the development of self-awareness. Future research should explore the process of becoming self-aware and determine methods to facilitate the development of adaptive strategies appropriate to maintain participation in life activities. Self-advocacy is related

to the concept of self-awareness and is an important life skill needed to navigate the complex world of adult health care. Jones (2009) suggests that achieving “optimal health” requires a strong collaborative partnership to be formed between individuals with CP and their healthcare providers with mutual respect and sharing of information between both parties. Learning to become self-aware is a key element to being able to advocate for, and participate in, directing their own healthcare needs.

Learning to manage fatigue levels and find a balance between activity and rest could be beneficial in preventing the early deterioration of ambulatory abilities documented in the literature (Mockford & Caulton, 2010). In a recent study, Opheim and colleagues (2013) hypothesized that deterioration in walking abilities could be explained by differences in kinematic variables of gait. However, the findings did not support the hypothesis, indicating that walking cessation and declining functional mobility of individuals with CP may be much more complex and multifactorial in nature. Many factors may be at work to influence functional mobility changes that occur with aging in CP, including the expectations the person has regarding their mobility status, the social and environmental constraints the person faces and the impact of secondary conditions like fatigue and pain. Clinical conversations regarding items on the FISSA may provide the opportunity to discuss the factors contributing to loss of functional status and to foster development of self-awareness and advance self-management techniques for youth with CP necessary for lifelong management of their health condition.

In the study conducted by van der Slot and colleagues (2012), fatigue (as assessed by the Fatigue Severity Scale and the Multidimensional Fatigue Inventory) was not shown to be associated with difficulty in daily functioning. The authors hypothesized that this may be due to low or limited participation levels to begin with; however, I question whether the tools used to measure fatigue were appropriate as both measures used to assess fatigue in that study were deemed to have either inadequate psychometric properties or require further psychometric testing in the review completed for this dissertation. This is a potential area for future research using the FISSA given that the descriptions provided by individuals with CP in the phenomenology and in the two personal published accounts (Jones, 2009; Howe, 2009) clearly demonstrate the impact of fatigue on life participation.

The FISSA was developed for both discriminative and predictive purposes and at present it has only been validated for discriminative use. Future research should explore the predictive validity of the FISSA as well as focus on the development of meaningful cut points to provide an enhanced interpretation of individual scores obtained on the FISSA. Large, population-based studies, are needed to further elucidate the fatigue profiles for each GMFCS level. In the validation study, fatigue levels were different between individuals classified as GMFCS level I and those classified as GMFCS II or III. However, no differences were detected between individuals classified as level II or III and those classified as level IV or V as was initially hypothesized. Fatigue should be examined across all GMFCS levels, and potentially, each level independently given that each GMFCS level represents clinically meaningful distinctions in functional abilities (Palisano, Rosenbaum, Walter, Russell, Wood & Galuppi, 1997).

One of the goals of at the outset of this dissertation was to understand the relationship between fatigue and physical activity. As a result of an unreliable measure of physical activity, this relationship remains unclear. Information gathered in the phenomenology indicates that there is potentially both a protective and detrimental effect of physical activity for individuals with CP. Once again, further research is required to understand the level of physical activity that may be beneficial for these individuals as well as what level physical activity provides only harmful effects on function. A dose-response study of physical activity would assist in providing advice regarding recommended levels of physical activity to enhance physical functioning for individuals wishing to preserve ambulatory skills and manage the fatigue that physical activity can create.

Finally, it is important to remember that each chapter in this dissertation describes an integral component to the development of the FISSA; however, each chapter is composed of a distinct study with separate inclusion and exclusion criteria. The specific subgroup of the population of interest continued to change over the course of the four studies described here as more information was gathered and new objectives were created for subsequent studies. In order to ensure the most useful measure was developed with maximum practicality in mind, it was decided to continue to change the inclusion and exclusion criteria as new information emerged and as new subject groups were interested

in participating in the different phases of the research program. I considered the pragmatic and emergent nature of this research to be important in ensuring usefulness of the FISSA. The novelty of this research area called for a more exploratory and inclusive approach to the studies contained in this dissertation. I felt that including as many perspectives as possible would lead to the construction a comprehensive measure that was suitable to all potential end users.

In conclusion, the final product of the program of research described in this dissertation was the development of the reliable and valid Fatigue Impact and Severity Self-Assessment measure. At the present time the FISSA can be used to determine the severity level of fatigue that is experienced by an individual with CP, the impact that is currently having on their life, as well as assess some of the management strategies they have tried to combat or limit their fatigue. This also serves as a starting point for a clinical conversation about other management strategies or intervention planning opportunities to prevent or reduce the impact of fatigue for that specific individual. The FISSA can also be used in research studies to determine levels of fatigue in the population with CP and explore relationships between fatigue and other health concepts such as cardiovascular fitness. In the future, studies exploring the predictive and potentially evaluative validity of the FISSA are warranted. In addition, exploring the distribution of FISSA scores in a large heterogeneous population with CP may provide useful information to identify clinically meaningful reference points for the FISSA. Ultimately, quality of life for individuals with CP has the potential to be improved with more effective self-management of the secondary impairment of fatigue and the FISSA is another tool in the toolkit for rehabilitation practitioners to enhance the self-awareness and self-management for individuals with CP.

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***Appendix 2-A – Copyright Permission From Developmental Neurorehabilitation  
to Reproduce “Fatigue in Cerebral Palsy: A Critical Review”***

**From:** Jeff Sigafoos  
**Subject:** RE: Permission for 15(1): 54–62  
**Date:** February 14, 2012 at 1:54 PM  
**To:** Laura Brunton

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Dear Laura,

You have my permission as Editor of Developmental Neurorehabilitation to reprint a version of the paper published in Feb 2012 Volume 15 Issue 1 pages 54-62 entitled "Fatigue in Cerebral Palsy: A Critical Review" in you PhD Dissertation.

Sincerely,

Jeff Sigafoos

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**From:** Laura Brunton  
**Sent:** Wednesday, February 15, 2012 1:55 AM  
**To:** Jeff Sigafoos  
**Subject:** Permission for 15(1): 54–62

Hi Dr. Sigafoos,

I am writing to ask for permission to reprint a version of the paper published in Feb 2012 Volume 15 Issue 1 pages 54-62 entitled "Fatigue in Cerebral Palsy: A Critical Review" in my PhD Dissertation. I appreciate your consideration on this matter.

Thank you,

Laura

Laura Brunton BKin (Hon), MSc  
PhD Candidate, Health and Rehabilitation Sciences Program  
The University of Western Ontario  
London, ON



**Appendix 3-A – Psychometric Properties of Identified Scales**

Psychometric Properties of Identified Scales

Scale	Reference	Condition of Interest	Internal Consistency (Cronbach's Alpha)	Test-retest Reliability	Type of Validity	Validity Values
Fatigue Severity Scale						
	Grace et al. 2007	Parkinson's Disease	0.94	Not Reported	Convergent Validity	Highly correlated with the Parkinson Fatigue Scale (r=0.84), and a one-question fatigue rating (r=0.80).
	Horemans et al. 2004	Postpoliom yelitis	0.85 and 0.80 Estimates from two different time points	ICC=0.83 (95% CI= 0.72-0.90)	Convergent Validity	Significant Spearman correlations with The Nottingham Health Profile Energy Category (0.50) and the Postpolio List (0.60) and the Short Fatigue Questionnaire (0.47).
					Test of Unidimensionality	Mokken scale analysis revealed that the first two items of the scale misfit. The original 9-item measure is not a unidimensional scale.
	Vasconcelos et al. 2006	Postpoliom yelitis	Not Reported	Not Reported	Convergent Validity	Significant correlations with a visual analog scale for fatigue r=0.45 only. Correlation with the Fatigue Impact Scale was low and non-significant.
					Divergent Validity	Non-significant low correlations with scales measuring depression and excessive sleepiness evidence of divergent validity.

Scale	Reference	Condition of Interest	Internal Consistency (Cronbach's Alpha)	Test-retest Reliability	Type of Validity	Validity Values
Fatigue Severity Scale						
					Known Groups Validity	Able to discriminate between those with disabling fatigue and those without.
	Mills et al. 2009	Multiple Sclerosis	Not Reported	Not Reported	Rasch Analysis	The original 9-item scale did not fit the Rasch model. Items 1 and 2 were removed because of misfit with the scale and items 6 and 8 were removed because of persistent differential item functioning. The remaining 5 items achieved unidimensionality and fit the Rasch model. However, the 5-item version demonstrated a ceiling effect of 13% and this may affect the validity of the scale in individuals reporting the most severe levels of fatigue.
	Flachenecker et al. 2002	Multiple Sclerosis	Not Reported	Not Reported	Convergent Validity	Correlated moderately with the Multiple Sclerosis Specific Fatigue Severity Scale (r=0.44), the Modified Fatigue Impact Scale (r=0.56) and a visual analog scale(r=0.38).
					Known Groups Validity	Able to discriminate between Multiple Sclerosis patients with and without fatigue. A cut-off value for determining fatigue was set at 4.6.

Scale	Reference	Condition of Interest	Internal Consistency (Cronbach's Alpha)	Test-retest Reliability	Type of Validity	Validity Values
Fatigue Severity Scale						
	Burger et al. 2010	Postpoliomyelitis	Above 0.95	Not Reported	Convergent Validity	Correlated well with visual analog scales measuring impact of fatigue on daily life ( $r=0.78$ ), self-care activities ( $r=0.64$ ) and household and occupation ( $r=0.74$ ).
	Chipchase et al. 2003	Multiple Sclerosis	Not Reported	Not Reported	Known Groups Validity	The Fatigue Severity Scale was able to discriminate between individuals with Multiple Sclerosis and those without.
	Krupp et al. 1989	Multiple Sclerosis	0.91	ICC=0.84	Convergent Validity	Moderately correlated with a visual analog scale for fatigue ( $r=0.47$ ).
Fatigue Impact Scale						
	Vasconcelos et al. 2006	Postpoliomyelitis	Not Reported	Not Reported	Convergent Validity	Correlations with the Fatigue Severity Scale the visual analog scale for fatigue were low and non-significant.
					Divergent Validity	Non-significant low correlations with scales measuring depression and excessive sleepiness provide evidence of divergent validity.
					Known Groups Validity	Not able to detect differences between those with disabling fatigue and those without.

Scale	Reference	Condition of Interest	Internal Consistency (Cronbach's Alpha)	Test-retest Reliability	Type of Validity	Validity Values
Fatigue Impact Scale						
	Mathiowetz 2003	Multiple Sclerosis	Not Reported	ICC=0.76 for the total scale, and ranged from 0.68-0.76 for the subscales	Convergent Validity	A significant but low correlation with the Fatigue Severity Scale (r=0.44) but significant and moderate correlations with the vitality (r=-0.55), social functioning (r=-0.54) and mental health (r=-0.62) subscales of the Short Form-36. Similar subscales of the Short Form-36 and the Fatigue Impact Scale demonstrated stronger correlations.
	Fisk et al. 1994	Multiple Sclerosis, Chronic Fatigue Syndrome	0.98	Not Reported	Convergent Validity	Correlated with the Sickness Impact Profile for each diagnosis group (r=0.57 Chronic Fatigue Syndrome; r=0.53 Multiple Sclerosis).
					Known Groups Validity	Able to discriminate between groups based on diagnosis of Chronic Fatigue Syndrome, Multiple Sclerosis and Hypertension and levels of fatigue.
	Chipchase et al. 2003	Multiple Sclerosis	Not Reported	Not Reported	Known Groups Validity	The cognitive, physical and social dimensions of the Fatigue Impact Scale were all able to discriminate between individuals with Multiple Sclerosis and those without.

Scale	Reference	Condition of Interest	Internal Consistency (Cronbach's Alpha)	Test-retest Reliability	Type of Validity	Validity Values
Fatigue Assessment Inventory						
	Schwartz et al. 1993	Multiple Sclerosis and Chronic Fatigue Syndrome	Range from 0.70-0.92 for the subscales	Range of $r=0.29-0.69$ for subscales.	Convergent Validity	The severity and psychological consequences subscales correlated with the Rand Vitality Index ( $r= -0.72$ Severity; $r= -0.41$ Psychological Consequences).
					Known Groups Validity	All of the subscales are able to discriminate between individuals with chronic fatiguing syndromes from healthy controls.
	Chipchase et al. 2003	Multiple Sclerosis	Not Reported	Not Reported	Known Groups Validity	The fatigue severity subscale was able to discriminate between individuals with Multiple Sclerosis and those without.

Scale	Reference	Condition of Interest	Internal Consistency	Test-retest Reliability	Type of Validity	Validity Values
Multidimensional Fatigue Inventory						
	Lin et al. 2009	Chronic Fatigue Syndrome	Total scale = 0.93, range from 0.71-0.86 for individual subscales.	Not Reported	Convergent Validity	The total score and subscales correlated with the eight subscales of the Short Form-36 but most strongly correlated with the Short Form-36 vitality subscale (total score $r=-0.81$ ). Additionally, the total score and all subscales were significantly correlated with depression (total score $=0.72$ ), anxiety (total score $r=0.62$ (Trait Anxiety) $r=0.50$ (State Anxiety)).
					Known Groups Validity	Each subscale was able to discriminate between individuals in the Chronic Fatigue Syndrome group, the chronically-unwell group and the well group.
	Smets et al. 1995	Chronic Fatigue Syndrome	Range from 0.82-0.91 for the separate subscales	Not Reported	Known Groups Validity	Able to discriminate between individuals chronically fatigued and other conditions and non-fatigued individuals.

Scale	Reference	Condition of Interest	Internal Consistency	Test-retest Reliability	Type of Validity	Validity Values
Modified Fatigue Impact Scale						
	Mills et al. 2010b	Multiple Sclerosis	Not Reported	Not Reported	Rasch Analysis	The 21-item scale did not fit the Rasch model. In the physical subscale items 4, 14 and 17 were deleted because of misfit with the scale. In the cognitive subscale items 1, 2, 3, 5 and 11 were removed as a result of misfit with the scale.
Modified Fatigue Impact Scale						
	Flachenecker et al. 2002	Multiple Sclerosis	Not Reported	Not Reported	Convergent Validity	Correlated moderately with the Fatigue Severity Scale ( $r=0.56$ ) and the visual analog scale ( $r=0.47$ ).
					Known Groups Validity	Able to discriminate between Multiple Sclerosis patients with and without fatigue. A cut-off value for determining fatigue was set at 38.
Parkinson Fatigue Scale						
	Grace et al. 2007	Parkinson's Disease	0.97	Not Reported	Convergent Validity	Significantly correlated ( $r=0.84$ ) with the Fatigue Severity Scale and the one question Fatigue Rating ( $r=0.78$ ).

Scale	Reference	Condition of Interest	Internal Consistency	Test-retest Reliability	Type of Validity	Validity Values
Parkinson Fatigue Scale						
	Brown et al. 2005	Parkinson's Disease	0.98	Spearman correlations between 0.52 and 0.72	Convergent Validity	Correlated highly with the Rhoten Fatigue Scale ( $r=0.71$ ).
					Known Groups Validity	Able to discriminate between individuals who considered themselves to have fatigue and those who did not. Also able to discriminate within the fatigued group and detected differences between individuals for which fatigue was or was not considered a problem.



Scale	Reference	Condition of Interest	Internal Consistency	Test-retest Reliability	Type of Validity	Validity Values
Chalder Fatigue Scale						
	Morriss et al. 1998	Chronic Fatigue Syndrome	Not Reported	Not Reported	Convergent Validity	The factor of cognitive difficulties correlated with the cognitive failures questionnaire (r=0.69), concentration (r=0.46) and learning recall (r=0.36). The strength and endurance factor correlated with grip strength (r=0.28 left and r=0.51 right), heart rate (r=0.24), VO2 (r=0.24) and functional work capacity (r=0.22). Additionally, the factor loss of interest and motivation correlated with depression (r=0.46).
					Factor Analysis	A factor analysis demonstrated a 4-factor structure to the scale.
Empirical Fatigue Scale						
	Bailes et al. 2006	Chronic Fatigue Syndrome	Not reported	r=0.87 and 0.91	Convergent Validity	Negatively correlated with handgrip strength (r=-0.33) suggesting increased fatigue with less strength.
					Divergent Validity	Also negatively correlated with the multiple sleep latency test (r=-0.40) suggesting discrimination between fatigue and objective sleep propensity.

Scale	Reference	Condition of Interest	Internal Consistency	Test-retest Reliability	Type of Validity	Validity Values
Empirical Fatigue Scale						
					Known Groups Validity	Able to discriminate between individuals with Chronic Fatigue Syndrome from individuals with narcolepsy and individuals with no health condition.
Fatigue Scale for Motor and Cognitive Functions						
	Penner et al. 2009	Multiple Sclerosis	0.93 (Cognitive) and 0.91 (Motor) and 0.95 for the total scale.	r=0.85 (Cognitive) and 0.86 (Motor) and 0.87 for the entire scale over a period of 4 weeks.	Convergent Validity	High correlations were demonstrated with the Fatigue Severity Scale (r=0.80) and with the Modified Fatigue Impact Scale (r=0.83) as well as fatigue rated by neurologists (r=0.51).
					Divergent Validity	Low correlations with the Beck Depression Inventory (r=0.49) and depression measured by neurologists (r=0.24).
					Known Groups Validity	Able to discriminate between individuals with Multiple Sclerosis and those without.

Scale	Reference	Condition of Interest	Internal Consistency	Test-retest Reliability	Type of Validity	Validity Values
Multiple Sclerosis Specific Fatigue Severity Scale						
	Flachenecker et al. 2002	Multiple Sclerosis	Not Reported	Not Reported	Convergent Validity	The Multiple Sclerosis Specific Fatigue Severity Scale correlated moderately with the Fatigue Severity Scale ( $r=0.44$ ), and demonstrated a low correlation with the Modified Fatigue Impact Scale ( $r=0.18$ ) while not significantly correlated with the visual analog scale.
					Known Groups Validity	Able to discriminate between Multiple Sclerosis patients with and without fatigue. However the Fatigue Severity Scale and the Modified Fatigue Impact Scale were better at discriminating between patients with and without fatigue
Neurological Fatigue Index						
	Mills et al. 2010a	Multiple Sclerosis	Not Reported	Above 0.70 (range 0.79 to 0.86) at 2 and 4 weeks for all subscales	Convergent Validity	Significantly correlated with the Modified Fatigue Impact Scale Physical Scale ( $r=0.71$ ), Modified Fatigue Impact Scale Cognitive Scale ( $r=0.58$ ), the 5-item Fatigue Severity Scale ( $r=0.71$ ) and visual analog scale ( $r=0.67$ ).

Scale	Reference	Condition of Interest	Internal Consistency	Test-retest Reliability	Type of Validity	Validity Values
Piper Fatigue Scale						
	Strohschein et al. 2003	Postpoliomyelitis	0.98	ICC=0.98	Convergent Validity	Piper fatigue scale correlated highly with the Chalder Fatigue Scale (r=0.80).
					Known Groups Validity	The Piper fatigue scale scores were significantly different between individuals with postpoliomyelitis and controls.
Short Fatigue Questionnaire						
	Horemans et al. 2004	Postpoliomyelitis	0.79 and 0.77	ICC = 0.84 (0.73-0.90)	Convergent Validity	Significant Spearman correlations with the Nottingham Health Profile energy category (0.67) and the Postpolio Problems List (0.68) and the Fatigue Severity Scale (0.47).
Swedish Occupational Fatigue Inventory						
	Johansson et al. 2008	Multiple Sclerosis	Range from 0.68 to 0.92 depending on the subscale	Not Reported	Convergent Validity	Other than moderate correlations between the Lack of Energy Subscale and the Fatigue Severity Scale (r=0.53-0.61) at the three points of data collection, the correlations were low between the subscales and the Fatigue Severity Scale.

Scale	Reference	Condition of Interest	Internal Consistency	Test-retest Reliability	Type of Validity	Validity Values
Unidimensional Fatigue Impact Scale						
	Meads et al. 2009	Multiple Sclerosis	0.96	r=0.86	Convergent Validity	Correlated highly with the Nottingham Health Profile Energy Level (r=0.64), and moderately with the Nottingham Health Profile Pain Scale (r=0.51), the Nottingham Health Profile Emotional Reactions Scale (r=0.52), the Nottingham Health Profile Physical Mobility Scale (r=0.47) and the Nottingham Health Profile Social Isolation Scale (r=0.54). Additionally, the U-FIS correlated moderately with the Hospital Anxiety and Depression Scale Anxiety Scale and moderately with the Hospital Anxiety and Depression Scale depression scale (r=0.60).
					Known Groups Validity	Able to discriminate between participants with different levels of self-perceived severity of Multiple Sclerosis and general health and between people experiencing a flare-up of symptoms and those who were not.

r=Pearson's Correlation Coefficient (other correlations are specified in text); ICC= Intraclass Correlation Coefficient; 95% CI= 95% Confidence Interval

## Appendix 4-A Ethics Approval from Western University for Phenomenology



### Office of Research Ethics

The University of Western Ontario  
 Room 4180 Support Services Building, London, ON, Canada N6A 5C1  
 Telephone: (519) 661-3036 Fax: (519) 850-2466 Email: ethics@uwo.ca  
 Website: www.uwo.ca/research/ethics

### Use of Human Subjects - Ethics Approval Notice

**Principal Investigator:** Dr. D.J. Bartlett

**Review Number:** 16899E

**Review Level:** Expedited

**Review Date:** February 24, 2010

**Approved Local # of Participants:** 10

**Protocol Title:** Understanding the bodily experience of cerebral palsy: a phenomenology

**Department and Institution:** Physical Therapy, University of Western Ontario

**Sponsor:**

**Ethics Approval Date:** March 11, 2010

**Expiry Date:** February 28, 2011

**Documents Reviewed and Approved:** UWO Protocol, Letter of Information and Consent, Assent Form.

**Documents Received for Information:**

This is to notify you that The University of Western Ontario Research Ethics Board for Health Sciences Research Involving Human Subjects (HSREB) which is organized and operates according to the Tri-Council Policy Statement: Ethical Conduct of Research Involving Humans and the Health Canada/ICH Good Clinical Practice Practices: Consolidated Guidelines; and the applicable laws and regulations of Ontario has reviewed and granted approval to the above referenced study on the approval date noted above. The membership of this REB also complies with the membership requirements for REB's as defined in Division 5 of the Food and Drug Regulations.

The ethics approval for this study shall remain valid until the expiry date noted above assuming timely and acceptable responses to the HSREB's periodic requests for surveillance and monitoring information. If you require an updated approval notice prior to that time you must request it using the UWO Updated Approval Request Form.

During the course of the research, no deviations from, or changes to, the protocol or consent form may be initiated without prior written approval from the HSREB except when necessary to eliminate immediate hazards to the subject or when the change(s) involve only logistical or administrative aspects of the study (e.g. change of monitor, telephone number). Expedited review of minor change(s) in ongoing studies will be considered. Subjects must receive a copy of the signed information/consent documentation.

Investigators must promptly also report to the HSREB:

- a) changes increasing the risk to the participant(s) and/or affecting significantly the conduct of the study;
- b) all adverse and unexpected experiences or events that are both serious and unexpected;
- c) new information that may adversely affect the safety of the subjects or the conduct of the study.

If these changes/adverse events require a change to the information/consent documentation, and/or recruitment advertisement, the newly revised information/consent documentation, and/or advertisement, must be submitted to this office for approval.

Members of the HSREB who are named as investigators in research studies, or declare a conflict of interest, do not participate in discussion related to, nor vote on, such studies when they are presented to the HSREB.

Chair of HSREB: Dr. Joseph Gilbert  
 FDA Ref. #: IRB 0000940

Ethics Officer to Contact for Further Information			
<input type="checkbox"/> Janice Sutherland	<input type="checkbox"/> Elizabeth Wambolt	<input checked="" type="checkbox"/> Grace Kelly	<input type="checkbox"/> Denise Grafton

File



## *Amendment to Change Assent Form*



### Use of Human Participants - Ethics Approval Notice

**Principal Investigator:** Dr. Doreen Bartlett  
**Review Number:** 16899E  
**Review Level:** Delegated  
**Approved Local Adult Participants:** 0  
**Approved Local Minor Participants:** 10  
**Protocol Title:** Understanding the bodily experience of cerebral palsy: a phenomenology  
**Department & Institution:** Physical Therapy, University of Western Ontario  
**Sponsor:**  
**Ethics Approval Date:** June 09, 2011      **Expiry Date:** April 30, 2012  
**Documents Reviewed & Approved & Documents Received for Information:**

Document Name	Comments	Version Date
Assent	The letter of information (assent letter) for children under the age of 18 has been revised to add information about privacy and confidentiality.	

This is to notify you that The University of Western Ontario Research Ethics Board for Health Sciences Research Involving Human Subjects (HSREB) which is organized and operates according to the Tri-Council Policy Statement: Ethical Conduct of Research Involving Humans and the Health Canada/ICH Good Clinical Practice Practices: Consolidated Guidelines; and the applicable laws and regulations of Ontario has reviewed and granted approval to the above referenced revision(s) or amendment(s) on the approval date noted above. The membership of this REB also complies with the membership requirements for REB's as defined in Division 5 of the Food and Drug Regulations.

The ethics approval for this study shall remain valid until the expiry date noted above assuming timely and acceptable responses to the HSREB's periodic requests for surveillance and monitoring information. If you require an updated approval notice prior to that time you must request it using the UWO Updated Approval Request Form.

Members of the HSREB who are named as investigators in research studies, or declare a conflict of interest, do not participate in discussion related to, nor vote on, such studies when they are presented to the HSREB.

The Chair of the HSREB is Dr. Joseph Gilbert. The UWO HSREB is registered with the U.S. Department of Health & Human Services under the IRB

#### Ethics Officer to Contact for Further Information

<input type="checkbox"/> Janice Sutherland	<input checked="" type="checkbox"/> Grace Kelly	<input type="checkbox"/> Shantel Walcott
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*This is an official document. Please retain the original in your files.*

### The University of Western Ontario

Office of Research Ethics

Support Services Building Room 5150 • London, Ontario • CANADA - N6A 3K7  
 PH: 519-661-3036 • F: 519-850-2466 • [ethics@uwo.ca](mailto:ethics@uwo.ca) • [www.uwo.ca/research/ethics](http://www.uwo.ca/research/ethics)



**Appendix 4-B Ethics Approval from Thames Valley Children's Centre for Phenomenology**



20 April 2010

Ms. Laura Brunton

**Re: Understanding the Bodily Experiences of Cerebral Palsy**

Dear Ms. Brunton,

I have reviewed the additional information that the Thames Valley Children's Centre's (TVCC) Research Advisory Committee requested that you provide for the above-named study and then relay in a letter to me. You have satisfactorily answered the Committee's queries.

You have identified Mary Ann Tucker as the person who will be initially contacted should participants experience emotional distress during the interview. You have explained that the TVCC Research Contact Person, Wendy McDonald, will now provide a clinical judgement about the client's ability to participate in the study (i.e., interact and communicate with an unfamiliar conversation partner), as opposed to administering the Communication Function Classification System. Finally, you have explained that your sample size of 8 to 10 participants should be adequate to reach saturation, and that follow-up interviews may be used if additional information is required from participants.

Thank you for providing the TVCC Research Program with a copy of the letter of approval for your study from the University of Western Ontario's Health Sciences Research Ethics Board.

You may now proceed with your study. Please arrange to present your findings to TVCC staff members at the completion of your study. In addition, it would be appreciated if you provide the TVCC Research Program with a copy of any journal articles or conference papers/abstracts that arise from your study.

Best wishes for a successful project!

Janeke McDonough, Ph.D.  
 Researcher, Research Program  
 Chair, Research Advisory Committee  
 Thames Valley Children's Centre

Cc: Karen Lowry, Director, CCIR, Privacy Officer  
 Mary Ann Tucker, Director, ECSAPP  
 Wendy McDonald, ECSAPP

Beyond disability

*Appendix 4-C Letter of Information and Consent Forms for Phenomenological Inquiry*

Letter of Information  
(Adolescents aged 14 years up to the 18<sup>th</sup> birthday)

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**Title of Study:** Understanding the Bodily Experience of Cerebral Palsy: A Phenomenology

**Investigators:** Laura Brunton, MSc, PhD (Student) Health and Rehabilitation Sciences, Graduate Program, The University of Western Ontario

Doreen Bartlett, PT, PhD, Associate Professor, School of Physical Therapy, Faculty of Health Sciences, The University of Western Ontario

The pronouns “you” and “your” in this letter should be read as referring to the participant and not the parent and/or guardian who is signing the consent form for the participant.

You are being invited to participate in a research study exploring the bodily experience of cerebral palsy. Currently there is no information about how adolescents and young adults experience their bodies during their daily activities. This study will use interviews with 8-10 adolescents and young adults aged 14 to 25 years to gain knowledge of how the body is perceived by persons with cerebral palsy.

If you agree to participate in this study, we will ask you to participate in one interview that lasts approximately one (1) hour in which you will be asked questions about your daily activities and how your body feels during those activities. The interview will be coordinated at a time that is convenient for you and will be conducted at your home, at The University of Western Ontario, or another location of your preference. If you wish, your parent may be present at the interview. The interview will be audio-taped and transcribed at a later date so the researcher can be focused on the interview and to ensure the transcript will be accurate.

If more information is needed there may be the possibility of returning for a second interview to elaborate on concepts and themes that arise from the initial interview. Again this interview will last no longer than one (1) hour and will be coordinated with your preferences in mind. This interview will also be audio-taped. Both interviews will be transcribed and given to you for your review and approval.

You will not experience any direct benefits from this study, however, this knowledge may help physiotherapists and other care providers when planning for interventions and transitions out of pediatric care models. There are no known risks associated with your participation. Information from this study may also be used to develop a tool to measure muscle fatigue and pain in adolescents and young adults with cerebral palsy. There is an option to participate in a future study about muscle fatigue in adolescents and young adults with cerebral palsy, you will be asked to provide your name and phone number if you wish to be contacted for future studies.

Should the interview take place at The University of Western Ontario, parking costs will be covered by the study.

Participation in this study is voluntary. You may refuse to participate, refuse to answer any questions, ask to have some components of the interview excluded or withdraw from the study at any time.

Your confidentiality will be respected. Your research records will be stored in a locked cabinet in a secure office in Elborn College at The University of Western Ontario in London and will be destroyed after 7 years. Audio-tapes will be erased after you have reviewed and approved the transcripts. Only those individuals listed as investigators will be able to access your information. When the results of this study are published, neither your name nor any identifying information will be used.

Representatives from The University of Western Ontario Health Sciences Research Ethics Board may contact you or require access to your study-related records to monitor the conduct of the research.

If you have any questions about this study, please contact Dr. Doreen Bartlett at \_\_\_\_\_, extension \_\_\_\_\_ or by email at \_\_\_\_\_.

If you have any questions about your rights as a research participant or the conduct of the study, you may contact The Office of Research Ethics at \_\_\_\_\_ or by email at \_\_\_\_\_.

You do not waive any legal rights by signing this consent form.

This letter is for you to keep.

## Consent Form

**Title of Study:** Understanding the Bodily Experience of Cerebral Palsy: A Phenomenology

**Investigators:** Laura Brunton, MSc, PhD (Student) Health and Rehabilitation Sciences, Graduate Program, The University of Western Ontario

Doreen Bartlett, PT, PhD, Associate Professor, School of Physical Therapy, Faculty of Health Sciences, The University of Western Ontario

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I have read the Letter of Information, have had the nature of the study explained to me and I agree to participate. All questions have been answered to my satisfaction.

---

Name of Participant (Please Print)

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Name of Parent/Guardian (Please Print)

---

Signature of Parent/Guardian

---

Signature of Investigator

---

Date

Letter of Information  
(Participants aged 18-25 years)

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**Title of Study:** Understanding the Bodily Experience of Cerebral Palsy: A Phenomenology

**Investigators:** Laura Brunton, MSc, PhD (Student) Health and Rehabilitation Sciences, Graduate Program, The University of Western Ontario

Doreen Bartlett, PT, PhD, Associate Professor, School of Physical Therapy, Faculty of Health Sciences, The University of Western Ontario

You are being invited to participate in a research study exploring the bodily experience of cerebral palsy. Currently there is no information about how adolescents and young adults experience their bodies during their daily activities. This study will use interviews with 8-10 adolescents and young adults aged 14 to 25 years to gain knowledge of how the body is perceived by persons with cerebral palsy.

If you agree to participate in this study, we will ask you to participate in one interview that lasts approximately one (1) hour in which you will be asked questions about your daily activities and how your body feels during those activities. The interview will be coordinated at a time that is convenient for you and will be conducted at your home, at The University of Western Ontario, or another location of your preference. This interview will be audio-taped.

There may be the possibility of returning for a second interview to elaborate on concepts and themes that arise from the initial interview. Again this interview will last no longer than one (1) hour and will be coordinated with your preferences in mind. This interview will also be audio-taped. Both interviews will be transcribed and given to you for your review and approval.

You will not experience any direct benefits from this study, however, this knowledge will help physiotherapists and other care providers when planning for interventions and transitions out of pediatric care models. Information from this study will also be used to develop a tool to measure muscle fatigue and pain in adolescents and young adults with cerebral palsy.

Should the interview take place at The University of Western Ontario, parking costs will be covered by the study.

Participation in this study is voluntary. You may refuse to participate, refuse to answer any questions, ask to have components of the interview deleted or withdraw from the study at any time with no effect on your future care.

Your confidentiality will be respected. Your research records will be stored in a locked cabinet in a secure office in Elborn College at The University of Western Ontario in London and will be destroyed after 7 years. Audio-tapes will be erased after they are transcribed and reviewed by you. Only those individuals listed as investigators will be able to access your information. When the results of this study are published, neither your name nor any identifying information will be used.

Representatives from The University of Western Ontario Health Sciences Research Ethics Board may contact you or require access to your study-related records to monitor the conduct of the research.

If you have any questions about this study, please contact Dr. Doreen Bartlett at \_\_\_\_\_, extension \_\_\_\_\_ or by email at \_\_\_\_\_.

If you have any questions about your rights as a research participant or the conduct of the study, you may contact The Office of Research Ethics at \_\_\_\_\_ or by email at \_\_\_\_\_.

You do not waive any legal rights by signing this consent form.

This letter is for you to keep.

## Consent Form

**Title of Study:** Understanding the Bodily Experience of Cerebral Palsy: A Phenomenology

**Investigators:** Laura Brunton, MSc, PhD (Student) Health and Rehabilitation Sciences, Graduate Program, The University of Western Ontario

Doreen Bartlett, PT, PhD, Associate Professor, School of Physical Therapy, Faculty of Health Sciences, The University of Western Ontario

---

I have read the Letter of Information, have had the nature of the study explained to me and I agree to participate. All questions have been answered to my satisfaction.

---

Name of Participant (Please Print)

---

Signature of Participant

---

Date

---

Signature of Investigator

Letter of Information  
(Participants aged 14 years up to the 18<sup>th</sup> birthday)

---

**Title of Study:** Understanding the Bodily Experience of Cerebral Palsy: A Phenomenology

**Investigators:** Laura Brunton, MSc, PhD (Student) Health and Rehabilitation Sciences, Graduate Program, The University of Western Ontario

Doreen Bartlett, PT, PhD, Associate Professor, School of Physical Therapy, Faculty of Health Sciences, The University of Western Ontario

**Why you are here:**

We would like to tell you about a study about youth with cerebral palsy. We want to ask if you would like to be in the study.

**Why are we doing this study?**

We want to know how your body feels during your daily activities.

**What will happen to you?**

If you agree to be in the study: we will ask you questions about how you feel about your body as you move, walk and play. You will be interviewed for less than one hour, and if we need help understanding some of the things you talked about in your first interview we may ask you for second interview. We will tape these interviews and will ask you to read them once they are typed up.

**Will the study hurt?**

The study will not hurt, it is only asking you questions, and you can choose not to answer a question if it makes you uncomfortable.

**Will you get better if you are in the study?**

This study will not help you feel better or get well, but it might help us understand and could help other people with cerebral palsy in the future.

**What if you have any questions?**

You can ask any questions you have at any time. You can talk to your family, or your doctor or someone else.



**What happens to the information you give in the study?**

You will get to pick a “nickname” to be used for the study instead of your real name. None of the answers you give will be shared with anyone else and no one will know they were your answers. You will get to read a printed version of your answers to the study questions after the interview and you can choose to remove anything you want.

**Do you have to be in the study?**

You do not have to be in this study. No one will be mad at you if you choose not to do this. If you do not want to be in the study just say so. If you say yes, you can change your mind and say no later. It is up to you.

## Assent Form

**Title of Study:** Understanding the Bodily Experience of Cerebral Palsy: A Phenomenology

**Investigators:** Laura Brunton, MSc, PhD (Student) Health and Rehabilitation Sciences, Graduate Program, The University of Western Ontario

Doreen Bartlett, PT, PhD, Associate Professor, School of Physical Therapy, Faculty of Health Sciences, The University of Western Ontario

I want to participate in this study.

---

Print name of Youth

---

Signature of Youth

---

Age

---

Date

---

Signature of Investigator

---

Date

*Appendix 4-D Semi-Structured Interview Guide*

**Understanding the Bodily Experience of Cerebral Palsy: A Phenomenology  
Semi-Structured Interview Guide**

**Thank you for agreeing to participate in this study. Today we'll be discussing your experience of your body. I will be recording this interview so I can focus on the conversation and type it out later. I just want to remind you that you can choose not to respond to any question without any problem. After this interview you will be given the written transcript of what we discuss today and you will have the option to include it in the study or remove any parts you wish. Are you ready to begin?**

1. Can you describe your typical day?
2. How does your body feel as the day goes on? [Probe "positive" or "negative" responses to gain a deep understanding of the typical day]
3. Does your experience of your body differ based on your daily activities [weekend vs. weekday or as the week progresses] and if so how?
4. Can you describe a day in which you experienced some fatigue or tiredness in your muscles?
5. Can you tell me about a day in which you had physical pain?
6. How does your body feel when you are being physically active? [Probe intensity of activities]
7. As you've gotten older, how has your body changed? [Probe differing experiences]

Note: Further questions may be added or questions may be removed and adapted as necessary to the individual being interviewed. Probing will occur to gain a deeper understanding of the topics arising from these questions.

## *Appendix 5-A Ethics Approval from Western University for Focus Groups and Feasibility Testing*



### Use of Human Participants - Ethics Approval Notice

**Principal Investigator:** Dr. Doreen Bartlett  
**Review Number:** 17998E  
**Review Level:** Delegated  
**Approved Local Adult Participants:** 0  
**Approved Local Minor Participants:** 0  
**Protocol Title:** Constructing a New Clinical Measure of Fatigue for Adolescents and Young Adults with Cerebral Palsy  
**Department & Institution:** Physical Therapy, University of Western Ontario  
**Sponsor:**  
**Ethics Approval Date:** June 14, 2011      **Expiry Date:** March 31, 2012  
**Documents Reviewed & Approved & Documents Received for Information:**

Document Name	Comments	Version Date
UWO Protocol		
Letter of Information & Consent	Health Care Professionals	
Letter of Information & Consent	Parents / Guardians	
Letter of Information & Consent	14-18 years old	
Other	Email	

This is to notify you that The University of Western Ontario Research Ethics Board for Health Sciences Research Involving Human Subjects (HSREB) which is organized and operates according to the Tri-Council Policy Statement: Ethical Conduct of Research Involving Humans and the Health Canada/ICH Good Clinical Practice Practices: Consolidated Guidelines; and the applicable laws and regulations of Ontario has reviewed and granted approval to the above referenced revision(s) or amendment(s) on the approval date noted above. The membership of this REB also complies with the membership requirements for REB's as defined in Division 5 of the Food and Drug Regulations.

The ethics approval for this study shall remain valid until the expiry date noted above assuming timely and acceptable responses to the HSREB's periodic requests for surveillance and monitoring information. If you require an updated approval notice prior to that time you must request it using the UWO Updated Approval Request Form.

Members of the HSREB who are named as investigators in research studies, or declare a conflict of interest, do not participate in discussion related to, nor vote on, such studies when they are presented to the HSREB.

The Chair of the HSREB is Dr. Joseph Gilbert. The UWO HSREB is registered with the U.S. Department of Health & Human Services under the IRB registration number IRB 00000940.

---

#### Ethics Officer to Contact for Further Information

<input type="checkbox"/> Janice Sutherland	<input checked="" type="checkbox"/> Grace Kelly	<input type="checkbox"/> Shantel Walcott
--	---	--

*This is an official document. Please retain the original in your files.*

**The University of Western Ontario**  
Office of Research Ethics

Support Services Building Room 5150 • London, Ontario • CANADA - N6A 3K7  
PH: 519-661-3036 • F: 519-850-2466 • [ethics@uwo.ca](mailto:ethics@uwo.ca) • [www.uwo.ca/research/ethics](http://www.uwo.ca/research/ethics)



## Appendix 5-B Focus Groups and Feasibility Testing Approval from McMaster University



Hamilton  
Health Sciences

### RESEARCH ETHICS BOARD



REB Office, 293 Wellington St. N., Suite 102, Hamilton, ON L8L 8E7  
Telephone: 905-521-2100, Ext. 42013  
Fax: 905-577-8378

#### Research Ethics Board Membership

Suzette Salama PhD  
Interim-Chair/Ethics Representative  
Donald Arnold MD, MSc FRCP(C)  
Hematology & Thromboembolism  
Uma Athale MBBS, MD, M.Sc. FRCP  
Pediatric Hematology/Oncology  
Mary Bedek CCHRA (C)  
Privacy Officer  
Joseph Beyene PhD  
Clinical Epidemiology & Biostatistics  
David Clark MD PhD FRCP(C)  
Medicine  
Jean Crowe MHSc  
Rehabilitation Science  
Lynn Donohue BA(Hons)  
Community Representative  
Melanie Griffiths FRCP (UK)  
Diagnostic Imaging  
Ali Hersi MD, PhD, FRCP  
Emergency Medicine  
Cindy James BScN  
Gastroenterology  
David Jewell M. Sc., MHSC  
Geriatrics  
Graham Jones BSc, MSc, PhD, MD,  
FRCP, FCCP  
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Peter Kavsak PhD, FCACB, FACB  
Laboratory Medicine  
Rosanne Kent RN BA MHSc(M)  
Cardiology  
Oliver Klimck MDir, MA  
Research Ethics Officer  
Grigorios Leontiadis MD PhD,  
Gastroenterology  
Steve Lloyd MD  
Family Medicine  
Cheryl Main MD FRCP, Medical  
Microbiology and Infectious Diseases  
Shelly McLean MBA  
Community Representative  
Leslie Murray RT (R), BAppSc (MI)  
Medical Radiation  
Madhu Natarajan MD, FRCP, FACC  
Cardiology  
Kesava Reddy MBBS FRCS FACS  
Neurosurgery  
Susan Rivers RN MSC (T),  
Geriatrics  
Gita Sobhi BSc Phm  
Pharmacy  
Alex Spyropoulos MD, FACP, FCCP,  
FRCP  
Medicine/Thrombosis Service  
Brian Timmons PhD  
Pediatrics  
Marie Townsend BA(Hons), MBA  
Administration  
Stephen Walter PhD  
Clinical Epidemiology & Biostatistics  
Kathryn Weibert MD  
Transfusion Medicine  
Andrew Worster MD  
Emergency Medicine  
Ed Younglai PhD  
Obstetrics/Gynecology

May 20, 2011

**PROJECT NUMBER:** 11-166

**PROJECT TITLE:** Constructing a New Clinical Measure of Fatigue for Adolescents and Young Adults with Cerebral Palsy

**PRINCIPAL INVESTIGATOR:** Marilyn Wright

This will acknowledge receipt of your letter dated May 10, 2011 which enclosed revised copies of the Information/Consent Form, Application Form, Protocol and the Recruitment Email for the above-named study. These issues were raised by the Research Ethics Board at their meeting held on April 19, 2011. Based on this additional information, we wish to advise your study has been given *final* approval from the full REB. The submission, Study Protocol version dated May 4, 2011 including the Information/Consent Form-Focus Group Health Care Professionals, Information/Consent Form- Legal Representative or Guardian and Information/Assent Form – Participant with Cerebral Palsy Under 18 Years, all version 2 dated May 4, 2011 together with Sample Email for Recruitment of Participant with CP version dated May 4, 2011; Sample Email for Recruitment of Healthcare Professionals; Appendix A: New Fatigue Scale (Work in Progress); Appendix B: Feasibility Questionnaire and Appendix F were found to be acceptable on both ethical and scientific grounds. **Please note** attached you will find the Information/Consent Form with the REB approval affixed; all consent forms used in this study must be copies of the attached materials.

We are pleased to issue final approval for the above-named study for a period of 12 months from the date of the REB meeting on April 19, 2011. Continuation beyond that date will require further review and renewal of REB approval. Any changes or revisions to the original submission must be submitted on an REB amendment form for review and approval by the Research Ethics Board.

The Hamilton Health Sciences/McMaster Health Sciences Research Ethics Board operates in compliance with and is constituted in accordance with the requirements of: The Tri-Council Policy Statement on Ethical Conduct of Research Involving Humans; The International Conference on Harmonization of Good Clinical Practices; Part C Division 5 of the Food and Drug Regulations of Health Canada; and the provisions of the Ontario Personal Health Information Protection Act 2004 and its applicable Regulations.

PLEASE QUOTE THE ABOVE-REFERENCE PROJECT NUMBER ON  
ALL-FUTURE CORRESPONDENCE

Sincerely,

Suzette Salama PhD,  
Interim Chair, Research Ethics Board

**Appendix 5-C Ethics Approval from Thames Valley Children's Centre for Focus Groups and Feasibility Testing**



16 June 2011

Ms. Laura Brunton

**Re: Constructing a new clinical measure of fatigue for adolescents and young adults with cerebral palsy**

Dear Ms. Brunton,

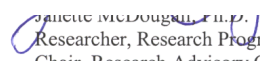
I have reviewed the additional information that the Thames Valley Children's Centre's (TVCC) Research Advisory Committee requested that you provide for the above-named study and then relay in a letter to me. You have satisfactorily answered the Committee's queries.

You have identified the correct number of participants to be recruited for the study. You will be submitting an amendment to the University of Western Ontario's Health Sciences Research Ethics Board to be able to include recreational therapists in the study. A definition of fatigue will be provided on the final version of the questionnaire you are constructing. Your plans for validity and reliability testing of the measure have been described. In addition, the process for analyzing client feedback on the questionnaire has been explained. You acknowledge an alternative recruitment strategy may be necessary at TVCC. Finally, you have simplified the Letter of Information for youth.

Once the TVCC Research Program has received a copy of the letter of approval for your study from the University of Western Ontario's Health Sciences Research Ethics Board, you may proceed with your study.

Please arrange to present your findings to TVCC staff members at the completion of your study. In addition, it would be appreciated if you provide the TVCC Research Program with a copy of any journal articles or conference papers/abstracts that arise from your study.

Best wishes for a successful project!

  
 Jennette McDougall, M.P.  
 Researcher, Research Program, Quality Management  
 Chair, Research Advisory Committee  
 Thames Valley Children's Centre

Cc: Karen Lowry, Director, QM, Privacy Officer  
 Linda Bolack, Director, CaTSS  
 Jenny Kermer, Manager, CaTSS/Clinic

*Beyond*   
 disability

***Appendix 5-D Feasibility Testing and Focus Groups Letter of Information and Consent Forms***

Letter of Information  
Focus Groups for Health Care Professionals

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**Title of Study:** Constructing a New Clinical Measure of Fatigue for Adolescents and Young Adults with Cerebral Palsy

**Investigators:** L. Brunton, MSc, PhD (Candidate), Health and Rehabilitation Sciences graduate program; D. Bartlett, PT, PhD, Associate Professor, Faculty of Health Sciences

You are being invited to participate in a research study aimed at creating a clinical measure of fatigue for adolescents and young adults with cerebral palsy. Currently there are no self-report measures of fatigue validated for use by individuals with cerebral palsy. A systematic review of fatigue measures validated in other neurological conditions has been conducted and a new fatigue scale consisting of items relevant to individuals with cerebral palsy as been created. This study will consist of focus groups with healthcare professionals who regularly treat individuals with cerebral palsy to gain feedback on how to shorten this scale and keep it relevant to individuals with cerebral palsy.

If you agree to participate in this study, we will ask you to participate in one focus group that lasts approximately ninety (90) minutes in which you will be asked to review the newly created fatigue scale and vote to delete or add items to the scale. The focus group will be coordinated at a time that is convenient for you and other participants from your centre. The focus group will consist of between 3-6 individuals from your center and will be audio-taped for accuracy.

There are no known risks, harms or discomforts associated with this study; however, if you feel uncomfortable at any time you may choose to withdraw from the study.

You will not experience any direct benefits from this study, however, the validation of a fatigue scale for individuals with cerebral palsy has the potential help physiotherapists and other healthcare providers when planning for interventions and transitions between care models.

Participation in this study is voluntary. You may refuse to participate, refuse to answer any questions posed by the focus group leader or withdraw from the study at any time.

Your confidentiality will be respected. The transcripts of the focus group will be stored in a locked cabinet in a secure office in Elborn College at The University of Western Ontario in London and will be destroyed after 7 years. Audio-tapes will be erased after they are transcribed and reviewed by the investigator. Only those individuals listed as investigators will be able to access your information. When the results of this study are published, neither your name nor any identifying information will be used. If you would



like a summary of the results of this study, please complete the attached form with your contact information.

Focus group members are asked to keep everything that they hear confidential and not to discuss it outside of the meeting. However, we cannot guarantee that confidentiality will be maintained by group members.

Representatives from The University of Western Ontario Health Sciences Research Ethics Board may contact you or require access to your study-related records to monitor the conduct of the research.

If you have any questions about this study, please contact Dr. Doreen Bartlett at \_\_\_\_\_, extension \_\_\_\_\_ or by email at \_\_\_\_\_.

If you have any questions about your rights as a research participant or the conduct of the study, you may contact The Office of Research Ethics at \_\_\_\_\_ or by email at \_\_\_\_\_.

You do not waive any legal rights by signing this consent form.

This letter is for you to keep.

## Consent Form

**Title of Study:** Constructing a New Clinical Measure of Fatigue for Adolescents and Young Adults with Cerebral Palsy

**Investigators:** L. Brunton, MSc, PhD (Candidate), Health and Rehabilitation Sciences graduate program; D. Bartlett, PT, PhD, Associate Professor, Faculty of Health Sciences

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I have read the Letter of Information, have had the nature of the study explained to me and I agree to participate. All questions have been answered to my satisfaction.

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Name of Participant (Please Print)

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Signature of Participant

---

Date

---

Signature of Investigator

---

Date

Letter of Information  
Parent or Legal Guardian of Individual with Cerebral Palsy

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**Title of Study:** Constructing a New Clinical Measure of Fatigue for Adolescents and Young Adults with Cerebral Palsy

**Investigators:** L. Brunton, MSc, PhD (Candidate), Health and Rehabilitation Sciences graduate program; D. Bartlett, PT, PhD, Associate Professor, Faculty of Health Sciences

The pronouns “you” and “your” in this letter should be read as referring to the participant and not the parent and/or guardian who is signing the consent form for the participant.

You are being asked to participate in a research study to create a measure of fatigue for youth and young adults with cerebral palsy. Currently there are no measures of fatigue for use by people with cerebral palsy. A review of fatigue measures used in similar conditions has been completed and a new fatigue scale with of items specific to people with cerebral palsy as been created. Healthcare professionals with experience treating people with cerebral palsy have given feedback on the new scale. We are asking youth with cerebral palsy to review the new scale and comment on the clarity of the questions and layout of the questionnaire.

If you agree to take part in this study, we will ask you to read the new fatigue scale and answer questions about the scale by email, this should take no longer than 45 minutes.

There are no known risks, harms or discomforts associated with this study; however, if you feel uncomfortable at any time you may choose to withdraw from the study.

You will not directly benefit from this study; however, the responses you give will assist in the creation of a fatigue scale for individuals with cerebral palsy that may help physiotherapists and other healthcare providers when planning for interventions.

Participation in this study is voluntary. You may refuse to participate, refuse to answer any questions or withdraw from the study at any time. If you wish to receive a summary of the results of this study please complete the attached summary of results form with your contact information.

Your confidentiality will be respected. The questionnaire will be stored in a locked cabinet in a secure office in Elborn College at The University of Western Ontario in London and will be destroyed after 7 years. Only individuals listed as investigators will be able to access your information. When the results of this study are published, neither your name nor any identifying information will be used.

Representatives from The University of Western Ontario Health Sciences Research Ethics Board may contact you or require access to your study-related records to monitor the conduct of the research.

If you have any questions about this study, please contact Dr. Doreen Bartlett at \_\_\_\_\_, extension \_\_\_\_\_ or by email at \_\_\_\_\_.

If you have any questions about your rights as a research participant or the conduct of the study, you may contact The Office of Research Ethics at \_\_\_\_\_ or by email at \_\_\_\_\_.

You do not waive any legal rights by signing this consent form.

This letter is for you to keep.

## Consent Form

**Title of Study:** Constructing a New Clinical Measure of Fatigue for Adolescents and Young Adults with Cerebral Palsy

**Investigators:** L. Brunton, MSc, PhD (Candidate), Health and Rehabilitation Sciences graduate program; D. Bartlett, PT, PhD, Associate Professor, Faculty of Health Sciences

---

I have read the Letter of Information, have had the nature of the study explained to me and I agree to participate. All questions have been answered to my satisfaction.

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Name of Participant (Please Print)

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Name of Guardian (Please Print)

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Signature of Guardian

---

Date

---

Signature of Investigator

---

Date

Letter of Information  
Individual with Cerebral Palsy Under Age 18

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**Title of Study:** Constructing a New Clinical Measure of Fatigue for Adolescents and Young Adults with Cerebral Palsy

**Investigators:** L. Brunton, MSc, PhD (Candidate), Health and Rehabilitation Sciences graduate program; D. Bartlett, PT, PhD, Associate Professor, Faculty of Health Sciences

You are being asked to participate in a research study to create a measure of fatigue for youth and young adults with cerebral palsy. Currently there are no measures of fatigue for use by people with cerebral palsy. A review of fatigue measures used in similar conditions has been completed and a new fatigue scale with of items specific to people with cerebral palsy as been created. Healthcare professionals with experience treating people with cerebral palsy have given feedback on the new scale. We are asking youth with cerebral palsy to review the new scale and comment on the clarity of the questions and layout of the questionnaire.

If you agree to take part in this study, we will ask you to read the new fatigue scale and answer questions about the scale by email, this should take no longer than 45 minutes.

There are no known risks, harms or discomforts associated with this study; however, if you feel uncomfortable at any time you may choose to withdraw from the study.

You will not directly benefit from this study; however, the responses you give will assist in the creation of a fatigue scale for individuals with cerebral palsy that may help physiotherapists and other healthcare providers when planning for interventions.

Participation in this study is voluntary. You may refuse to participate, refuse to answer any questions or withdraw from the study at any time. If you would like to receive a summary of the results of this study please complete the attached summary of results form with your contact information.

Your confidentiality will be respected. The questionnaire will be stored in a locked cabinet in a secure office in Elborn College at The University of Western Ontario in London and will be destroyed after 7 years. Only individuals listed as investigators will be able to access your information. When the results of this study are published, neither your name nor any identifying information will be used.

Representatives from The University of Western Ontario Health Sciences Research Ethics Board may contact you or require access to your study-related records to monitor the conduct of the research.

If you have any questions about this study, please contact Dr. Doreen Bartlett at \_\_\_\_\_, extension \_\_\_\_\_ or by email at \_\_\_\_\_.

If you have any questions about your rights as a research participant or the conduct of the study, you may contact The Office of Research Ethics at \_\_\_\_\_ or by email at \_\_\_\_\_.

You do not waive any legal rights by signing this consent form.

This letter is for you to keep.

## Assent Form

**Title of Study:** Constructing a New Clinical Measure of Fatigue for Adolescents and Young Adults with Cerebral Palsy

**Investigators:** L. Brunton, MSc, PhD (Candidate), Health and Rehabilitation Sciences graduate program; D. Bartlett, PT, PhD, Associate Professor, Faculty of Health Sciences

---

I want to participate in this study.

---

Print name of Youth

---

Signature of Youth

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Age

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Date

---

Signature of Investigator

---

Date



*Appendix 5-E Feasibility Questionnaire*

1. How easy was it to answer these questions?

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2. Are there any questions on the questionnaire that you do not understand? If yes, which question(s) (provide the number) and tell us what is confusing about the question?

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3. Were the answer choices/style of answer appropriate for the questions asked? If not, what would you prefer to see?

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4. Is there anything else you would like to tell us about this questionnaire?

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***Appendix 5-F Items for Consideration for the Fatigue Impact and Severity Self-Assessment***

Potential Items for the Fatigue Impact and Severity Self-Assessment

Brief Fatigue Inventory:

- Circle the number that describes how, during the past 24 hours, fatigue has interfered with your: (0-10 scale)
  - A. General Activity
  - C. Walking Ability
  - F. Enjoyment of Life

FACIT Fatigue Scale:

- Circle the number as it applies to the past 7 days (0-4 scale):
  - I have trouble starting things because I am tired
  - I have trouble finishing things because I am tired
  - I am frustrated by being too tired to do the things I want to do
  - I have to limit my social activity because I am tired

Fatigue Impact Scale:

- Circle the number that indicates best how much of a problem fatigue has been for you these past 4 weeks, including today (0-4 scale): “Because of my fatigue”
  - I feel that I am more isolated from social contact
  - I have to reduce my workload or responsibilities
  - I have difficulty paying attention for a long period of time
  - I have to rely more on others to help me or do things for me
  - I have difficulty planning activities ahead of time because my fatigue may interfere with them
  - I am more clumsy and uncoordinated
  - I am more irritable and more easily angered
  - I have to be careful about pacing my physical activities
  - I am less motivated to do anything that requires physical effort
  - I am less motivated to engage in social activities
  - My ability to travel outside my home is limited
  - I have trouble maintaining physical effort for long periods
  - I have few social contacts outside my home
  - Normal day-to-day events are stressful for me
  - My muscles feel much weaker than they should
  - My physical discomfort is increased
  - I have difficulty dealing with anything new
  - I feel unable to meet the demand that people place on me
  - I am less able to complete tasks that require physical effort
  - I worry about how I look to other people
  - I have to limit my physical activities
  - I require more frequent or longer periods of rest
  - Minor difficulties seem like major difficulties

Modified Fatigue Impact Scale:

- Because of my fatigue during the past four weeks...(0-4 scale):
  - I have had to pace myself in my physical activities
  - I have been less motivated to do anything that requires physical effort
  - I have been less motivated to participate in social activities
  - I have been limited in my ability to do things away from home
  - I have had trouble maintaining physical effort for long periods
  - My muscles have felt weak
  - I have been physically uncomfortable
  - I have been less able to complete tasks that require physical effort
  - I have limited my physical activities
  - I have needed to rest more often or for longer periods

Fatigue Symptom Inventory:

- Circle the number that best indicates how that item applies to you (0-10 scale):
  - Rate your level of fatigue on the day you felt most fatigued during the past week
  - Rate your level of fatigue on the day you felt least fatigued during the past week
  - Rate your level of fatigue on average during the past week
  - Rate how much, in the past week, fatigue interfered with your general level of activity
  - Indicated how many days, in the past week, you felt fatigued for any part of the day (0-7)
  - Rate how much of the day, on average, you felt fatigued in the past week
  - Indicate which of the following best describes the daily pattern of your fatigue in the past week (0-4, not at all, worse in morning, in afternoon, in evening, no pattern)

Multidimensional Fatigue Symptom Inventory:

- Circle the number which best describes how true that statement has been for you in the past 7 days (0-4 scale):
  - My muscles ache
  - I feel weak all over
  - I need help doing my usual activities
  - I have trouble starting things
  - I ache all over
  - I have no energy

Fatigue Severity Scale:

- During the past week, I have found that (1-7 Scale):
  - My motivation is lower when I am fatigued
  - Exercise brings on my fatigue
  - I am easily fatigued

- Fatigue interferes with my physical functioning
- Fatigue causes frequent problems for me
- My fatigue prevents sustained physical functioning
- Fatigue interferes with carrying out certain duties and responsibilities
- Fatigue is among my three most disabling symptoms
- Fatigue interferes with my work, family or social life

Multidimensional Assessment of Fatigue Scale:

- Circle the number that most closely indicates how you have been feeling during the past week (0-10 scale):
  - How severe is the fatigue which you have been experiencing
  - To what degree has fatigue caused you distress
  - In the past week, to what degree has fatigue interfered with your ability to:
    - Engage in leisure and recreational activities
  - To what degree has your fatigue changed during the past week (4-1 scale, increased, fatigue has gone up and down, stayed the same, decreased)

Piper Fatigue Scale:

- Qualitative questions:
  - Overall, what do you believe is most directly contributing to or causing your fatigue?
  - Overall, the best thing you have found to relieve your fatigue is?

***Appendix 5-G Initial Draft of the Fatigue Impact and Severity Self-Assessment for Focus Group 1***

Initial Draft of the Fatigue Impact and Severity Self-Assessment

**Severity**

- Rate your level of fatigue on the day last week that you felt **most** fatigued:  
 No Fatigue                                  Moderate Fatigue                                  Severe Fatigue
- Rate your level of fatigue on the day last week that you felt **least** fatigued:  
 No Fatigue                                  Moderate Fatigue                                  Severe Fatigue
- Rate your **average** level of fatigue for the past week:  
 No Fatigue                                  Moderate Fatigue                                  Severe Fatigue
- How many days last week did you feel fatigued at least part of the day?  
 1                  2                  3                  4                  5                  6                  7
- How much of the day, on average, do you feel fatigued?  
 None                                  Half The Day                                  All Day

**Impact**

**Using the scale below, to what extent do you agree with the following statements?**

- Completely Disagree                                  Neither Agree nor Disagree                                  Completely Agree

6. Fatigue interferes with my general every day activities
7. Fatigue interferes with my ability to walk
8. Fatigue interferes with my enjoyment of my life
9. I have trouble starting things because of fatigue
10. I have trouble finishing things because of fatigue
11. I get frustrated because fatigue stops me from doing the things I would like to do
12. I have to limit my social activities because of fatigue
13. I feel more isolated from social contacts because of fatigue
14. I have had to reduce my workload or responsibilities because of fatigue
15. Fatigue makes me more reliant on others to help me or do things for me
16. I find it hard to plan activities in advance because my fatigue might interfere with them
17. Fatigue makes me more clumsy and uncoordinated
18. I am easily angered when I am tired
19. I have to pace my physical activities because of fatigue

20. I have a hard time finding the motivation to do things that require physical effort because of fatigue
21. I have a hard time finding the motivation to participate in social activities because of fatigue
22. Fatigue limits my ability to travel outside my house
23. I have trouble being physically active for long periods at a time because of fatigue
24. Fatigue makes normal day-to-day events stressful
25. My muscles feel weak due to fatigue
26. Fatigue makes me feel physically uncomfortable
27. Fatigue makes unexpected or new things more difficult
28. Fatigue makes it harder for me to meet the demands other people place on me
29. Fatigue makes me less capable of completing tasks that require physical effort
30. I have to limit my physical activity because of fatigue
31. Fatigue makes me stop and rest more or for longer periods
32. Fatigue makes minor difficulties seem like major difficulties
33. Fatigue makes my muscles ache
34. Fatigue makes me feel weak all over
35. I need help doing my usual activities because of fatigue
36. Exercise or physical activity brings on fatigue
37. I am easily fatigued
38. My motivation to do other tasks is lower when I am fatigued
39. Fatigue is among three of my most disabling symptoms
40. Long periods of rest or inactivity can make fatigue worse
41. Stress increases my fatigue
42. Fatigue interferes with my leisure and recreational activities
43. My fatigue is worse in the morning
44. My fatigue is worse in the afternoon
45. My fatigue is worse in the evening
46. My fatigue is the same all day
47. My fatigue changes depending on the time of day
48. Fatigue makes me feel upset

Qualitative Questions:

49. Overall, what factor is responsible or contributes the most to your fatigue?

50. What have you found is the best way to reduce your fatigue?

***Appendix 5-H Second Draft of the Fatigue Impact and Severity Self-Assessment for  
Focus Group 2***

Second Draft of the Fatigue Impact and Severity Self-Assessment

**Severity**

1. Rate your level of fatigue on the day last week that you felt **most** fatigued:

No Fatigue                                      Moderate Fatigue                                      Severe Fatigue

2. Rate your level of fatigue on the day last week that you felt **least** fatigued:

No Fatigue                                      Moderate Fatigue                                      Severe Fatigue

3. Rate your **average** level of fatigue for the past week:

No Fatigue                                      Moderate Fatigue                                      Severe Fatigue

4. How many days last week did you feel fatigued at least part of the day?

1                      2                      3                      4                      5                      6                      7

5. How much of the day, on average, do you feel fatigued?

None                                      Half The Day                                      All Day

6. Does your level of fatigue change depending on the time of day?

Yes (if yes please answer question 6b)                                      No

6b) What time of day is your fatigue worse?

Early Morning	Mid morning	Noon	Late afternoon	Evening
1	2	3	4	5

**Impact**

**Using the scale below, to what extent do you agree with the following statements?**

Completely Disagree	Neither Agree nor Disagree	Completely Agree
------------------------	-------------------------------	---------------------

6. Fatigue interferes with my general every day activities
7. Fatigue interferes with my ability to move around indoors
8. Fatigue interferes with my enjoyment of my life
9. Fatigue interferes with my ability to start things
10. Fatigue interferes with my ability to finish things
11. Fatigue interferes with my ability to do things I would like to do
12. Fatigue interferes with my participation in social activities

13. I have had to reduce my workload or responsibilities because of fatigue
14. Fatigue interferes with my ability to do my usual activities on my own
15. I think about fatigue when I plan my day
16. Fatigue interferes with my balance and coordination
17. Fatigue interferes with my ability to control my temper
18. I pace my physical activities to manage my fatigue
19. My motivation to participate in social activities is lower when I am fatigued.
20. My motivation to do physical activities is lower when I am fatigued.
21. Fatigue interferes with my ability to get outside of my house.
22. Fatigue interferes with my ability to move around in my community.
23. Fatigue interferes with how long I can be physically active.
24. Fatigue interferes with my ability to deal with unexpected things.
25. I limit my physical activity to manage my fatigue
26. I stop and rest during activity to manage my fatigue
27. My muscles ache when I am fatigued.
28. I feel weak when I am fatigued.
29. Long periods of inactivity increase my fatigue.
30. Stress increases my fatigue
31. Fatigue increases my stress.
32. Fatigue interferes with my leisure and recreational activities
33. I get frustrated when I am fatigued.

Qualitative Questions:

34. Overall, what factor is responsible or contributes the most to your fatigue?
35. What have you found is the best way to reduce your fatigue?
36. What could other people do to help reduce your fatigue?



**Appendix 5-I Draft of the Fatigue Impact and Severity Self-Assessment for Feasibility Testing**

## Fatigue Impact and Severity Self-Assessment (FISSA)

Please answer the following questions about your experience with fatigue. For the purposes of this questionnaire we would like you to think about fatigue in terms:

- physical tiredness,
- muscle soreness
- exhaustion of your muscles and body
- or any related feeling

When answering the questions, please try to focus on fatigue as it is defined above and not pain you may experience that is different from muscle soreness.

### Impact Scale

Completely Disagree	Somewhat Disagree	Neither Agree nor Disagree	Somewhat Agree	Completely Agree
1	2	3	4	5

Using the scale above and thinking about a **typical week** (7 days), to what extent do you agree with the following statements?

### Fatigue interferes with ...

1. my general everyday activities	1	2	3	4	5
2. my ability to move around indoors	1	2	3	4	5
3. my enjoyment of life	1	2	3	4	5
4. my ability to start things	1	2	3	4	5
5. my ability to finish things	1	2	3	4	5
6. my participation in social activities	1	2	3	4	5
7. my leisure and recreational activities	1	2	3	4	5
8. the length of time I can be physically active	1	2	3	4	5
9. my ability to get outside of my house	1	2	3	4	5
10. my ability to move around in my community	1	2	3	4	5
11. my ability to do things on my own	1	2	3	4	5
12. my balance and coordination	1	2	3	4	5
13. my ability to control my mood (examples: I get upset, cranky, irritable, sad, frustrated etc.)	1	2	3	4	5



24. On average, how much of the day do you feel fatigued?

None	A Quarter of the Day	Half the Day	Three Quarters of the Day	All Day
1	2	3	4	5

25. Does your level of fatigue change depending on the time of day?

Yes (If yes, please answer question 25b) No

25b. What time of day is your fatigue the worst?

Early Morning	Mid-morning	Noon	Late afternoon	Evening
1	2	3	4	5

26. Does your level of fatigue change depending on the day of the week?

Yes (If yes, please answer question 26b) No

26b. On which day of the week are you most fatigued?

Monday	Tuesday	Wednesday	Thursday	Friday	Saturday	Sunday
1	2	3	4	5	6	7

27. For how many days last week did you feel fatigued at least part of the day?

1	2	3	4	5	6	7
---	---	---	---	---	---	---

### Management Scale

Using the scale below and thinking about a **typical week** (7 days), to what extent do you agree with the following statements?

Completely Disagree 1	Somewhat Disagree 2	Neither Agree nor Disagree 3	Somewhat Agree 4	Completely Agree 5
-----------------------------	---------------------------	------------------------------------	------------------------	--------------------------

28. I pace my physical activities to manage my fatigue	1	2	3	4	5
29. I think about fatigue when I plan my day	1	2	3	4	5
30. I limit my physical activity to manage my fatigue	1	2	3	4	5
31. I stop and rest during activity to manage my fatigue	1	2	3	4	5
32. I use adaptive equipment to manage my fatigue (examples: a walker, manual wheelchair, power wheelchair etc.)	1	2	3	4	5
33. I have had to reduce my work responsibilities outside of my home because of fatigue (examples: school work, job-related work, volunteering etc.)	1	2	3	4	5
34. I have had to reduce my responsibilities at home because of fatigue	1	2	3	4	5

### Additional Questions:

35. What factors are responsible for or contribute to your fatigue?

36. What do you do to reduce or manage your fatigue?

37. What else could you do to reduce or manage your fatigue?

38. What could other people do to help reduce your fatigue?

## *Appendix 6-A Western University Ethical Approval*



### Use of Human Participants - Ethics Approval Notice

**Principal Investigator:** Dr. Doreen Bartlett  
**Review Number:** 18701E  
**Review Level:** Delegated  
**Approved Local Adult Participants:** 0  
**Approved Local Minor Participants:** 0  
**Protocol Title:** Validating a New Measure of Fatigue for Youth and Young Adults with Cerebral Palsy: The Fatigue Impact and Severity Self-Assessment  
**Department & Institution:** Physical Therapy, University of Western Ontario  
**Sponsor:** Canadian Institutes of Health Research

**Ethics Approval Date:** February 17, 2012                      **Expiry Date:** January 31, 2013  
**Documents Reviewed & Approved & Documents Received for Information:**

Document Name	Comments	Version Date
UWO Protocol		
Letter of Information	Adolescents aged 14 years up to the 16th birthday	2012/01/24
Letter of Information	Young Adult aged 16 years and older	2012/01/24
Letter of Information	Parents / Guardians of Children 14-16	2012/01/24

This is to notify you that The University of Western Ontario Research Ethics Board for Health Sciences Research Involving Human Subjects (HSREB) which is organized and operates according to the Tri-Council Policy Statement: Ethical Conduct of Research Involving Humans and the Health Canada/ICH Good Clinical Practice Practices: Consolidated Guidelines; and the applicable laws and regulations of Ontario has reviewed and granted approval to the above referenced revision(s) or amendment(s) on the approval date noted above. The membership of this REB also complies with the membership requirements for REB's as defined in Division 5 of the Food and Drug Regulations.

The ethics approval for this study shall remain valid until the expiry date noted above assuming timely and acceptable responses to the HSREB's periodic requests for surveillance and monitoring information. If you require an updated approval notice prior to that time you must request it using the UWO Updated Approval Request Form.

Members of the HSREB who are named as investigators in research studies, or declare a conflict of interest, do not participate in discussion related to, nor vote on, such studies when they are presented to the HSREB.

The Chair of the HSREB is Dr. Joseph Gilbert. The UWO HSREB is registered with the U.S. Department of Health & Human Services under the IRB registration number IRB 00000940.

#### Ethics Officer to Contact for Further Information

Janice Sutcliffe		Shantal Watson
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*This is an official document. Please retain the original in your files.*



### Use of Human Participants - Ethics Approval Notice

**Principal Investigator:** Dr. Doreen Bartlett  
**Review Number:** 18701E  
**Review Level:** Delegated  
**Approved Local Adult Participants:** 0  
**Approved Local Minor Participants:** 0  
**Protocol Title:** Validating a New Measure of Fatigue for Youth and Young Adults with Cerebral Palsy: The Fatigue Impact and Severity Self-Assessment  
**Department & Institution:** Health Sciences\Physical Therapy, University of Western Ontario  
**Sponsor:** Canadian Institutes of Health Research

**Ethics Approval Date:** March 29, 2012                      **Expiry Date:** January 31, 2013  
**Documents Reviewed & Approved & Documents Received for Information:**

Document Name	Comments	Version Date
Revised Western University Protocol	The children's treatment centre will now pass along contact information of people who have consented to be contacted for research by other researchers to the researcher. Recruitment and advertisement of the study have also been revised.	
Advertisement	Advertisement for the Ontario Federation of Cerebral Palsy and Facebook Groups related to Cerebral Palsy.	
Advertisement	Recruitment Advertisement for the Canadian Physiotherapy Association, Pediatric Division	
Advertisement	Snowball sampling script	
Other	Revised questionnaire question	

This is to notify you that The University of Western Ontario Research Ethics Board for Health Sciences Research Involving Human Subjects (HSREB) which is organized and operates according to the Tri-Council Policy Statement: Ethical Conduct of Research Involving Humans and the Health Canada/ICH Good Clinical Practice Practices: Consolidated Guidelines; and the applicable laws and regulations of Ontario has reviewed and granted approval to the above referenced revision(s) or amendment(s) on the approval date noted above. The membership of this REB also complies with the membership requirements for REB's as defined in Division 5 of the Food and Drug Regulations.

The ethics approval for this study shall remain valid until the expiry date noted above assuming timely and acceptable responses to the HSREB's periodic requests for surveillance and monitoring information. If you require an updated approval notice prior to that time you must request it using the University of Western Ontario Updated Approval Request Form.

Members of the HSREB who are named as investigators in research studies, or declare a conflict of interest, do not participate in discussion related to, nor vote on, such studies when they are presented to the HSREB.

The Chair of the HSREB is Dr. Joseph Gilbert. The HSREB is registered with the U.S. Department of Health & Human Services under the IRB registration number IRB 00000940.

#### Ethics Officer to Contact for Further Information

<input type="checkbox"/> Janice Sutherland	<input checked="" type="checkbox"/> Grace Kelly	<input type="checkbox"/> Shantel Walcott
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### The University of Western Ontario

Office of Research Ethics

Support Services Building Room 5150 • London, Ontario • CANADA - N6G 1G9  
 PH: 519-661-3036 • F: 519-850-2466 • ethics@uwo.ca • www.uwo.ca/research/ethics



## Use of Human Participants - Ethics Approval Notice

**Principal Investigator:** Dr. Doreen Bartlett  
**File Number:** 102145  
**Review Level:** Delegated  
**Approved Local Adult Participants:** 0  
**Approved Local Minor Participants:** 0  
**Protocol Title:** Validating a New Measure of Fatigue for Youth and Young Adults with Cerebral Palsy: The Fatigue Impact and Severity Self-Assessment - 18701E  
**Department & Institution:** Health Sciences\Physical Therapy, Western University  
**Sponsor:** Canadian Institutes of Health Research

**Ethics Approval Date:** June 20, 2012 **Expiry Date:** January 31, 2013

**Documents Reviewed & Approved & Documents Received for Information:**

Document Name	Comments	Version Date
Revised Western University Protocol	Revised participant recruitment and number of study participants.	
Increase in number of local Participants	Increase from 50 to 150.	

This is to notify you that The University of Western Ontario Research Ethics Board for Health Sciences Research Involving Human Subjects (HSREB) which is organized and operates according to the Tri-Council Policy Statement: Ethical Conduct of Research Involving Humans and the Health Canada/ICH Good Clinical Practice Practices: Consolidated Guidelines; and the applicable laws and regulations of Ontario has reviewed and granted approval to the above referenced revision(s) or amendment(s) on the approval date noted above. The membership of this REB also complies with the membership requirements for REB's as defined in Division 5 of the Food and Drug Regulations.

The ethics approval for this study shall remain valid until the expiry date noted above assuming timely and acceptable responses to the HSREB's periodic requests for surveillance and monitoring information. If you require an updated approval notice prior to that time you must request it using the University of Western Ontario Updated Approval Request Form.

Members of the HSREB who are named as investigators in research studies, or declare a conflict of interest, do not participate in discussion related to, nor vote on, such studies when they are presented to the HSREB.

The Chair of the HSREB is Dr. Joseph Gilbert. The HSREB is registered with the U.S. Department of Health & Human Services under the IRB registration number IRB 00000940.

**Ethics Officer to Contact for Further Information**

<input type="checkbox"/> Janice Sutherland	<input checked="" type="checkbox"/> Grace Kelly	<input type="checkbox"/> Shantel Walcott
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*This is an official document. Please retain the original in your files.*





## Use of Human Participants - Ethics Approval Notice

**Principal Investigator:** Dr. Doreen Bartlett  
**File Number:** 102145  
**Review Level:** Delegated  
**Approved Local Adult Participants:** 0  
**Approved Local Minor Participants:** 0  
**Protocol Title:** Validating a New Measure of Fatigue for Youth and Young Adults with Cerebral Palsy: The Fatigue Impact and Severity Self-Assessment - 18701E  
**Department & Institution:** Health Sciences\Physical Therapy, Western University  
**Sponsor:** Canadian Institutes of Health Research

**Ethics Approval Date:** September 14, 2012 **Expiry Date:** January 31, 2013  
**Documents Reviewed & Approved & Documents Received for Information:**

Document Name	Comments	Version Date
Revised Western University Protocol	The number of study participants has been revised to 400. In addition the study team will now advertise also at upcoming conferences.	
Advertisement		

This is to notify you that The University of Western Ontario Research Ethics Board for Health Sciences Research Involving Human Subjects (HSREB) which is organized and operates according to the Tri-Council Policy Statement: Ethical Conduct of Research Involving Humans and the Health Canada/ICH Good Clinical Practice Practices: Consolidated Guidelines; and the applicable laws and regulations of Ontario has reviewed and granted approval to the above referenced revision(s) or amendment(s) on the approval date noted above. The membership of this REB also complies with the membership requirements for REB's as defined in Division 5 of the Food and Drug Regulations.

The ethics approval for this study shall remain valid until the expiry date noted above assuming timely and acceptable responses to the HSREB's periodic requests for surveillance and monitoring information. If you require an updated approval notice prior to that time you must request it using the University of Western Ontario Updated Approval Request Form.

Members of the HSREB who are named as investigators in research studies, or declare a conflict of interest, do not participate in discussion related to, nor vote on, such studies when they are presented to the HSREB.

The Chair of the HSREB is Dr. Joseph Gilbert. The HSREB is registered with the U.S. Department of Health & Human Services under the IRB registration number IRB 00000940.

## Ethics Officer to Contact for Further Information

<input type="checkbox"/> Janice Sutherland	<input checked="" type="checkbox"/> Grace Kelly	<input type="checkbox"/> Shantel Walcott
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*This is an official document. Please retain the original in your files.*



*Appendix 6-B Letter of Information and Consent Forms for Validation Study*

Letter of Information  
(Young Adult aged 16 years and older)

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**Title of Study:** Validating a New Measure of Fatigue for Youth and Young Adults with Cerebral Palsy: The Fatigue Impact and Severity Self-Assessment

**Investigators:** Laura Brunton, MSc, PhD (Student) Health and Rehabilitation Sciences, Graduate Program, Faculty of Health Sciences, The University of Western Ontario

Doreen Bartlett, PT, PhD, Professor, School of Physical Therapy, Faculty of Health Sciences, The University of Western Ontario

You are being invited to take part in a research study to test a new questionnaire about fatigue. Currently there is no way of understanding the impact or severity of fatigue for youth and young adults with cerebral palsy. This study will use a survey to measure levels of fatigue, pain, physical activity, and personal aspects of youth and young adults aged 14 to 30 years to understand how fatigue affects these individuals. The information from this study will be used to understand any relationships that may exist between pain, fatigue and physical activity level. This information will also be used to assess the stability and usefulness of the new questionnaire about fatigue.

If you agree to take part in this study, we will ask you to fill out a survey that should take no longer than one (1) hour. The survey booklet is with this letter and will ask questions about fatigue, pain, physical activity and your outlook on life. The survey package will also ask about how you usually move around in different places, how many of your limbs are affected and to what degree they are affected by cerebral palsy. You can complete this survey online if you prefer. You will find a link to the online survey on the front page of the survey package.

You may be asked to complete a second survey package two weeks after the first one. This survey package will be shorter and should take no more than thirty (30) minutes to complete. The second survey will only ask about your fatigue, pain and physical activity levels. This survey can also be done online if you prefer. You will find a link to the online survey on the front of the survey package.

All finished survey materials should be returned to the study team in the self-addressed, stamped envelope included with this letter, or completed online if you choose that method. If you decide not to take part in the study, please mail the blank survey back to the study team and you will not be contacted again.

Mailing a completed survey package to the study team implies that you have read this letter and agree to your information being used in the study.

You will not experience any direct benefits from this study; however, this information may help physiotherapists and other care providers when planning for interventions and transitions from pediatric to adult care. Included with the survey is a \$10 iTunes gift card as a thank you for participating in the study.

There are no known risks to participating in this study. Participation in this study is voluntary. You may refuse to participate or refuse to answer any questions at any time with no effect on your future care.

Your confidentiality will be respected. You will not be asked to provide any information that will identify you in any way. Your research records will be stored in a locked cabinet in a secure office in Elborn College at The University of Western Ontario in London, Ontario and will be destroyed after 7 years. Digital copies of the survey data will be erased at this time and paper copies will be shredded. Only those people listed as investigators will be able to access your information. When the results of this study are published, neither your name nor any identifying information will be used.

Representatives from The University of Western Ontario Health Sciences Research Ethics Board may contact you or require access to your study-related records to monitor the conduct of the research.

If you have any questions about this study, please contact Dr. Doreen Bartlett at \_\_\_\_\_, extension \_\_\_\_\_ or by email at \_\_\_\_\_.

If you have any questions about your rights as a research participant or the conduct of the study, you may contact The Office of Research Ethics at \_\_\_\_\_ or by email at \_\_\_\_\_.

This letter is for you to keep.

Letter of Information  
(Parent/Guardian of Adolescents aged 14 years up to the 16<sup>th</sup> birthday)

---

**Title of Study:** Validating a New Measure of Fatigue for Youth and Young Adults with Cerebral Palsy: The Fatigue Impact and Severity Self-Assessment

**Investigators:** Laura Brunton, MSc, PhD (Student) Health and Rehabilitation Sciences, Graduate Program, Faculty of Health Sciences, The University of Western Ontario

Doreen Bartlett, PT, PhD, Professor, School of Physical Therapy, Faculty of Health Sciences, The University of Western Ontario

The pronouns “you” and “your” in this letter should be read as referring to the participant and not the parent and/or guardian who is responsible for agreeing for the participant. We ask you to read this letter and talk about this study with your child before agreeing to take part in the study.

You are being invited to take part in a research study to test a new questionnaire about fatigue. Currently there is no way of understanding the impact or severity of fatigue for youth and young adults with cerebral palsy. This study will use a survey to measure levels of fatigue, pain, physical activity, and personal aspects of youth and young adults aged 14 to 30 years to understand how fatigue affects these individuals. The information from this study will be used to understand any relationships that may exist between pain, fatigue and physical activity level. This information will also be used to assess the stability and usefulness of the new questionnaire about fatigue.

If you agree to take part in this study, we will ask you to fill out a survey that should take no longer than one (1) hour. The survey booklet is with this letter and will ask questions about fatigue, pain, physical activity and your outlook on life. The survey package will also ask about how you usually move around in different places, how many of your limbs are affected and to what degree they are affected by cerebral palsy. You can complete this survey online if you prefer. You will find a link to the online survey on the front page of the survey package.

You may be asked to complete a second survey package two weeks after the first one. This survey package will be shorter and should take no more than thirty (30) minutes to complete. The second survey will only ask about your fatigue, pain and physical activity levels. This survey can also be done online if you prefer. You will find a link to the online survey on the front of the survey package.

All finished survey materials should be returned to the study team in the self-addressed, stamped envelope included with this letter, or completed online if you choose that method. If you decide not to take part in the study, please mail the blank survey back to the study team and you will not be contacted again.

Mailing a completed survey package to the study team implies that you have read this letter and agree to your information being used in the study.

You will not experience any direct benefits from this study; however, this information may help physiotherapists and other care providers when planning for interventions and transitions from pediatric to adult care. Included with the survey is a \$10 iTunes gift card as a thank you for participating in the study.

There are no known risks to participating in this study. Participation in this study is voluntary. You may refuse to participate or refuse to answer any questions at any time with no effect on your future care.

Your confidentiality will be respected. You will not be asked to provide any information that will identify you in any way. Your research records will be stored in a locked cabinet in a secure office in Elborn College at The University of Western Ontario in London, Ontario and will be destroyed after 7 years. Digital copies of the survey data will be erased at this time and paper copies will be shredded. Only those people listed as investigators will be able to access your information. When the results of this study are published, neither your name nor any identifying information will be used.

Representatives from The University of Western Ontario Health Sciences Research Ethics Board may contact you or require access to your study-related records to monitor the conduct of the research.

If you have any questions about this study, please contact Dr. Doreen Bartlett at \_\_\_\_\_, extension \_\_\_\_\_ or by email at \_\_\_\_\_.

If you have any questions about your rights as a research participant or the conduct of the study, you may contact The Office of Research Ethics at \_\_\_\_\_ or by email at \_\_\_\_\_.

This letter is for you to keep.

Letter of Information  
(Adolescents aged 14 years up to the 16<sup>th</sup> birthday)

---

**Title of Study:** Validating a New Measure of Fatigue for Youth and Young Adults with Cerebral Palsy: The Fatigue Impact and Severity Self-Assessment

**Investigators:** Laura Brunton, MSc, PhD (Student) Health and Rehabilitation Sciences, Graduate Program, Faculty of Health Sciences, The University of Western Ontario

Doreen Bartlett, PT, PhD, Professor, School of Physical Therapy, Faculty of Health Sciences, The University of Western Ontario

You are being asked to take part in a research study to test a new questionnaire about fatigue. Currently there is no way to understand the impact or severity of fatigue for youth and young adults with cerebral palsy. This study will ask questions about your levels of fatigue, pain, and physical activity. The information from this study will be used to understand the relationships between fatigue, pain, and physical activity. This information will also be used to assess the strength and usefulness of the new questionnaire about fatigue.

If you agree to take part in this study, we will ask you to fill out a survey that should take no longer than one (1) hour. The survey booklet is with this letter and will ask questions about fatigue, pain, physical activity and your outlook on life. The survey package will also ask about how you usually move around in different places, how many of your limbs are affected and to what amount they are affected by cerebral palsy. You can complete this survey online if you prefer. You will find a link to the online survey on the front page of the survey package.

You may be asked to complete a second survey package two weeks after the first one. This survey package will be shorter and should take no more than thirty (30) minutes to complete. The second survey will only ask about your fatigue, pain and physical activity levels. This survey can also be done online if you prefer. You will find a link to the online survey on the front of the survey package.

All finished survey materials should be returned to the study team in the self-addressed, stamped envelope included with this letter, or completed online if you choose that way. If you decide not to take part in the study, please mail the blank survey back to the study team and you will not be contacted again.

Mailing a completed survey package to the study team means that you have read this letter and agree to your information being used in the study.

You will not experience any direct benefits from this study; however, this information may help physiotherapists and other care providers when planning for therapy and transitions from pediatric to adult care. Included with the survey is a \$10 iTunes gift card as a thank you for participating in the study.

There are no known risks to participating in this study. Participation in this study is voluntary. You may refuse to participate or refuse to answer any questions at any time with no effect on your future care.

Your privacy will be respected. You will not be asked to provide any information that will identify you in any way. Your research records will be stored in a locked cabinet in a secure office in Elborn College at The University of Western Ontario in London, Ontario and will be destroyed after 7 years. Digital copies of the survey data will be erased at this time and paper copies will be shredded. Only those people listed as investigators will be able to access your information. When the results of this study are published, neither your name nor any identifying information will be used.

Representatives from The University of Western Ontario Health Sciences Research Ethics Board may contact you or require access to your study-related records to monitor the conduct of the research.

If you have any questions about this study, please contact Dr. Doreen Bartlett at \_\_\_\_\_, extension \_\_\_\_\_ or by email at \_\_\_\_\_.

If you have any questions about your rights as a research participant or the conduct of the study, you may contact The Office of Research Ethics at \_\_\_\_\_ or by email at \_\_\_\_\_.

This letter is for you to keep.

*Appendix 6-C Recruitment Advertisement Flyer*

Recruitment Advertisement for the Ontario Federation of Cerebral Palsy and Various Facebook Groups Related to Cerebral Palsy

Researchers at The University of Western Ontario are conducting a study involving individuals with cerebral palsy aged 14 to 30 years and are looking for participants. A survey package would be mailed out to participants with the questionnaire covering topics such as fatigue, physical activity, pain, level of optimism and mobility. The questionnaire can also be completed online. If you have questions, would like to participate, or know of someone who might like to take part in their study, please contact Laura Brunton at \_\_\_\_\_ for further information.

Recruitment Advertisement for the Canadian Physiotherapy Association, Pediatric Division

Researchers at The University of Western Ontario are conducting a study involving individuals with cerebral palsy aged 14 to 30 years and are looking for participants. A survey package would be mailed out to participants with the questionnaire covering topics such as fatigue, physical activity, pain, level of optimism and mobility. The questionnaire can also be completed online. If you have questions, or know of someone who might like to take part in their study, please contact Laura Brunton at \_\_\_\_\_ for further information.

*Appendix 6-D Self-Report Measure of the Gross Motor Function Classification System  
Expanded and Revised Version*

Please read this and mark **only one box** beside the description that best fits how you move.

**I have difficulty sitting on my own and controlling my head and body posture in most positions**

**and** have difficulty achieving any voluntary control of movement  
**and** need a specially adapted chair to sit comfortably and be transported anywhere  
**and** have to be lifted or hoisted by another person or special equipment to move

---

**I can sit with some pelvic and trunk support but do not stand or walk without significant support**

**and** therefore always rely on a wheelchair when outdoors  
**and** can achieve movement by myself using a powered wheelchair  
**and** can crawl or roll to a limited extent to move around indoors

---

**I can stand on my own and walk if using a hand-held walking aid** (such as a walker, rollator, crutches, canes, etc.)

**and** find it difficult to climb stairs, or walk on uneven surfaces without support  
**and** use a variety of means to move around depending on the circumstances  
**and** prefer to use a wheelchair to travel quickly or over longer distances

---

**I can walk on my own without any walking aids, but need to hold the handrail when going up or down stairs**

**and** therefore walk in most settings  
**and** often find it difficult to walk on uneven surfaces, slopes or in crowds  
**and** may occasionally prefer to use a walking aid (such as a cane or crutch) or a wheelchair to travel quickly or over longer distances

---

**I can walk on my own without using walking aids, and can go up or down stairs without needing to hold the handrail**

**and** walk wherever I want to go (including uneven surfaces, slopes or in crowds)  
**and** can run and jump although my speed, balance, and coordination may be limited



### Appendix 6-E Demographic Questionnaire

How old are you? \_\_\_\_\_ (years)

What is your gender?  Male  
 Female

Have you ever been told you have cerebral palsy?

Yes  No

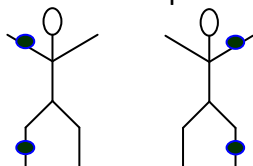
Please check **THE ONE** description of distribution that best describes your body (please refer to descriptions below):

monoplegia  hemiplegia  diplegia  triplegia  quadriplegia

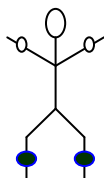
#### DESCRIPTIONS

**'Monoplegia'** means that only one limb is involved (either upper or lower extremity).

**'Hemiplegia'** (or 'hemisynndrome') means that there is both a lot of asymmetry of CP, such that one side of the body is obviously affected and the other has little or no obvious functional impairment

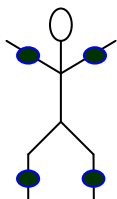


**'Diplegia'** is a term usually meant to describe CP where the legs are more functionally affected than the arms.



**'Triplegia'** means that three limbs are involved (usually both lower extremities and one upper extremity).

**'Quadriplegia'** (Tetraplegia) is also sometimes called **'total body involvement'**, and recognizes that trunk and whole body posture are importantly functionally involved with the CP.



*Appendix 6-F Pain Questionnaire*

**Date Completed:** \_\_\_\_\_

**Completed by:** Study participant independently [  ] or with assistance/interview [  ]

**Over the past month, have you experienced physical pain?**

NO [  ] - if you responded "no", you are finished this questionnaire

YES [  ]

If you have experienced physical pain **over the past month**, please circle **how much the pain gets in the way of your daily activities** over the past month (1 = doesn't get in the way at all to 10 = unable to carry out activities because of the pain).

1	2	3	4	5	6	7	8	9	10
doesn't get in the way at all									unable to carry out activities because of the pain

Please indicate **how severe your pain was over the past month** on average (1 very little pain to 10 extremely painful).

1	2	3	4	5	6	7	8	9	10
very little pain									extremely painful

**Please turn to the next page to indicate the areas of your body that were painful over the past month.**

Please check those areas of your body that were painful over the past

Neck

Shoulder Area

Elbow/Forearm

Wrist/Hand

Hip

Thigh

Knee Area

Calf Area

Ankle/Foot Area

Upper Back

Lower Back

month.

Thank you for completing this questionnaire.

*Appendix 6-G Exercise Questionnaire*

The chart on the next page asks about the exercises you did last week. By "exercise", we mean activities that involve stretching, strengthening, or physical effort. Activities that involve effort or exertion cause the following things: 1) the heart works harder and faster, 2) breathing is deeper, and 3) the body perspires or sweats.

The 1<sup>st</sup> column in the chart asks you to think about and circle the number for the exercises you did over the past week. There is some space for you to write in the other "sports", or "exercises" that you did.

The 2<sup>nd</sup> column in the chart asks you to write-in the number of different times in the past week you did each of the exercises listed. If you didn't do an exercise at all, just leave the space blank.

The 3<sup>rd</sup> column in the chart asks you to write-in the average amount of time (in minutes) you spent doing each of the exercises listed, each time. You won't need to put anything in this column for the exercises you did not do at all last week.

The 4<sup>th</sup> column in the chart asks you to write-in how hard you worked on average when you did each exercise last week. Again, you won't need to put anything in this column for the exercises you did not do at all last week.

When you are thinking about how hard you worked, please choose either **light**, **medium**, or **hard** according to the descriptions below:

<b>Light</b>	normal heart rate and breathing, no sweating
<b>Medium</b>	some increase in heart rate and breathing
<b>Hard</b>	heart working hard, breathing very deep, sweating

### Exercise Questionnaire

Study ID: \_\_\_\_ \_\_\_\_ \_\_\_\_

Date Completed: \_\_\_\_\_

	Activity	How many times did you do the activity last week? (write "zero" if you didn't do the activity)	Each time you did the activity, how much time did you spend doing it? (in minutes)	How hard did you work at the activity? (light, medium, hard)
1	Walk / run			
2	Wheel in a manual wheelchair			
3	Cycle			
4	Dance			
5	Swim / aquatics			
6	Basketball			
7	Hockey / sledge hockey			
8	Baseball / T ball			
9	Soccer			
10	Volleyball			
11	Football			
12	Bowling / Boccia ball			
13	Horseback riding			
14	Canoeing / kayaking			
15	Sailing			
16	Golf			
17	Skating / Skiing			
18	Rollerblading			
19	Martial Arts / wrestling / yoga / gymnastics			
20	Strength training/weight lifting			
21	Stretching exercises			
Other activities (please list)				
22	a.			
	b.			
	c.			
	d.			
	e.			

**Light**

normal heart rate and breathing, not sweating

**Medium**

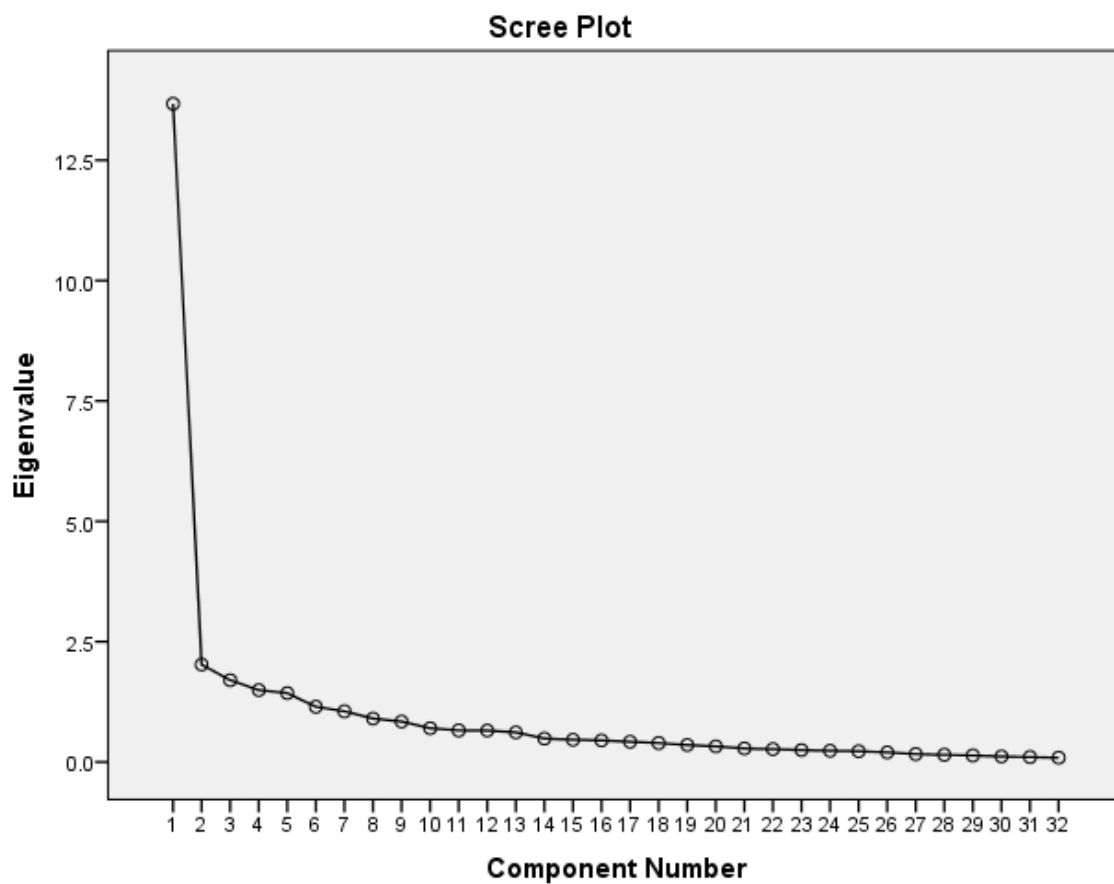
some increase in heart rate and breathing

**Hard**

heart working hard, breathing very deep, sweating



*Appendix 6-I Scree Plot of Fatigue Impact and Severity Self-Assessment Factor  
Extraction Analysis*



**Appendix 6-J Final Fatigue Impact and Severity Self-Assessment Following Factor Analysis**

**Fatigue Impact and Severity Self-Assessment (FISSA)**

Please answer the following questions about your experience with fatigue. For the purposes of this questionnaire we would like you to think about fatigue in terms:

- physical tiredness
- muscle soreness
- exhaustion of your muscles and body
- or any related feeling

When answering the questions, please try to focus on fatigue as it is defined above and not pain you may experience that is different from muscle soreness.

**Impact Scale**

Completely Disagree 1	Somewhat Disagree 2	Neither Agree nor Disagree 3	Somewhat Agree 4	Completely Agree 5
-----------------------------	---------------------------	------------------------------------	------------------------	--------------------------

Using the scale above and thinking about **a typical week** (7 days), to what extent do you agree with the following statements?

**Fatigue interferes with !**

1. my general everyday activities	1	2	3	4	5
2. my ability to move around indoors	1	2	3	4	5
3. my ability to do things on my own	1	2	3	4	5
4. my ability to move around in my community	1	2	3	4	5
5. my ability to get outside of my house	1	2	3	4	5
6. my ability to finish things	1	2	3	4	5
7. my participation in social activities	1	2	3	4	5
8. my ability to start things	1	2	3	4	5
9. my ability to take care of myself (examples: Dressing, eating, bathing, brushing my teeth/hair, toileting etc.)	1	2	3	4	5

**In addition,**

10. I use adaptive equipment to manage my fatigue (examples: a walker, manual wheelchair, power wheelchair etc.)	1	2	3	4	5
11. I have had to reduce my work responsibilities outside of my home because of fatigue (examples: school work, job-related work, volunteering etc.)	1	2	3	4	5
12. I have had to reduce my responsibilities at home because of fatigue	1	2	3	4	5



Using the scale given with each question, please think about the **last seven (7) days** and answer the following statements or questions.

13. Rate your level of fatigue on the day within the last week that you felt the **most** fatigued:

No Fatigue		Moderate Fatigue		Severe Fatigue
1	2	3	4	5

14. Rate your level of fatigue on the day within the last week that you felt the **least** fatigued:

No Fatigue		Moderate Fatigue		Severe Fatigue
1	2	3	4	5

15. Rate your **average** level of fatigue for the past week:

No Fatigue		Moderate Fatigue		Severe Fatigue
1	2	3	4	5

16. On average, how much of the day do you feel fatigued?

None	A Quarter of the Day	Half the Day	Three Quarters of the Day	All Day
1	2	3	4	5

17. For how many days last week did you feel fatigued at least part of the day?

1	2	3	4	5	6	7
---	---	---	---	---	---	---

**Management and Activity Modification Scale**

Using the scale below and thinking about a **typical week** (7 days), to what extent do you agree with the following statements?

Completely Disagree 1	Somewhat Disagree 2	Neither Agree nor Disagree 3	Somewhat Agree 4	Completely Agree 5
-----------------------------	---------------------------	------------------------------------	------------------------	--------------------------

**Fatigue interferes with !**

18. my enjoyment of life	1	2	3	4	5
19. my leisure and recreational activities	1	2	3	4	5
20. the length of time I can be physically active	1	2	3	4	5
21. my balance and coordination	1	2	3	4	5
22. my motivation to do physical activities	1	2	3	4	5
23. my motivation to participate in social activities	1	2	3	4	5

**In addition,**

24. my muscles ache when I am fatigued	1	2	3	4	5
25. long periods of inactivity increase my fatigue	1	2	3	4	5
26. stress increases my fatigue	1	2	3	4	5
27. fatigue increases my stress	1	2	3	4	5
28. I pace my physical activities to manage my fatigue	1	2	3	4	5
29. I think about fatigue when I plan my day	1	2	3	4	5
30. I limit my physical activity to manage my fatigue	1	2	3	4	5
31. I stop and rest during activity to manage my fatigue	1	2	3	4	5

**Additional Questions:**

32. Does your level of fatigue change depending on the time of day?

Yes (If yes, please answer question 25b) No

32b. What time of day is your fatigue the worst?

Early Morning	Mid-morning	Noon	Late afternoon	Evening
1	2	3	4	5

33. Does your level of fatigue change depending on the day of the week?

Yes (If yes, please answer question 26b) No

33b. On which day of the week are you most fatigued?

Monday	Tuesday	Wednesday	Thursday	Friday	Saturday	Sunday
1	2	3	4	5	6	7

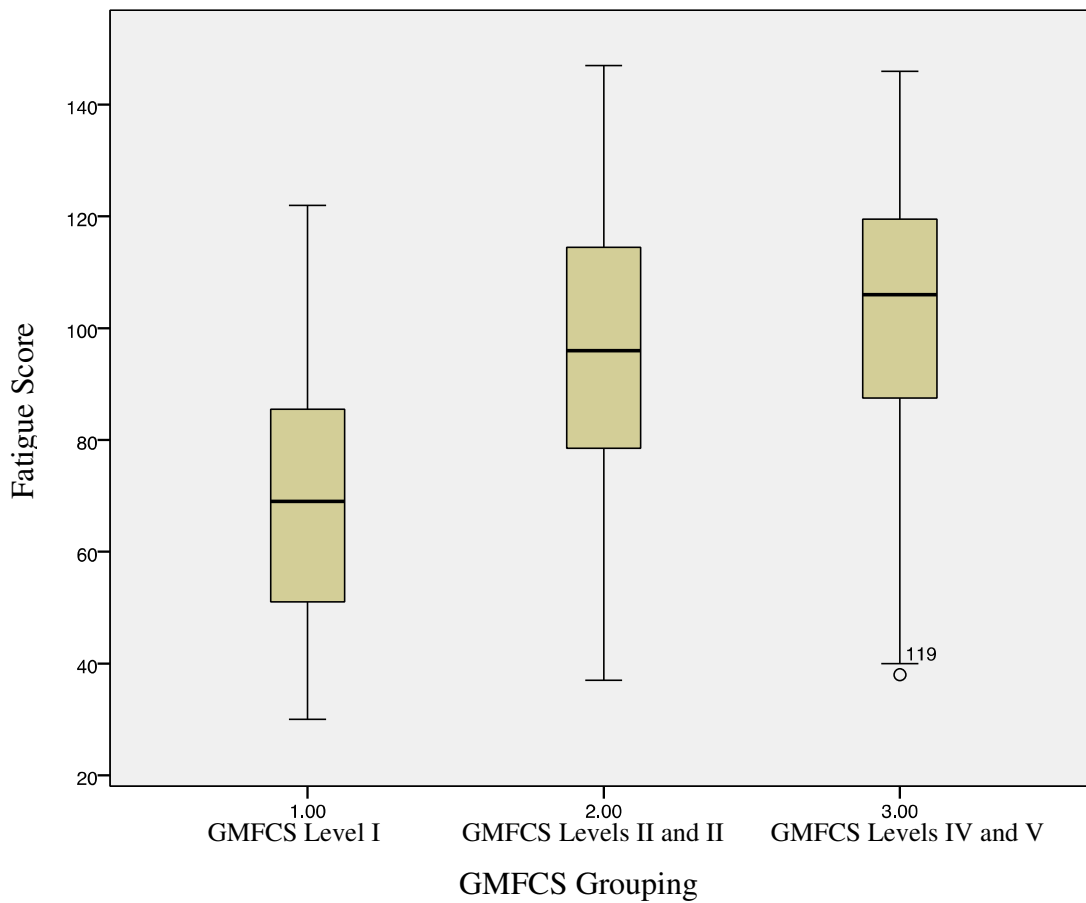
34. What factors are responsible for or contribute to your fatigue?

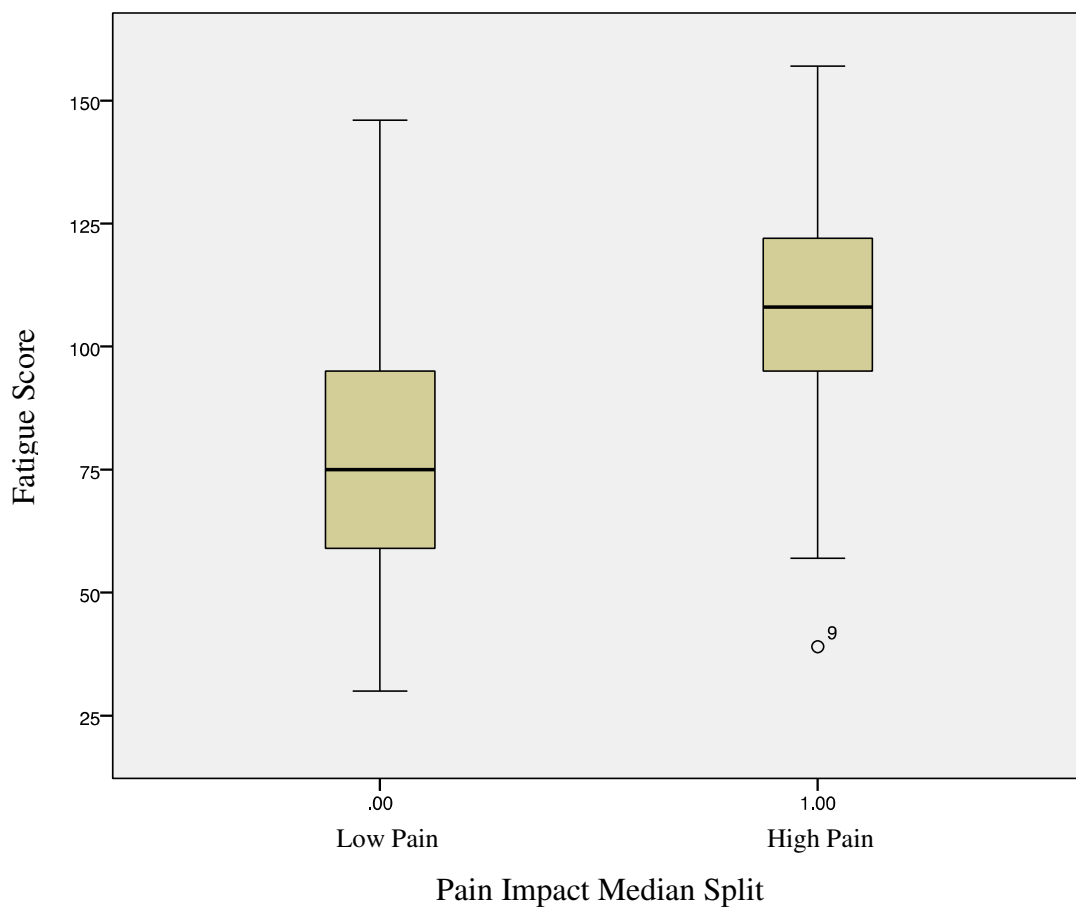
35. What do you do to reduce or manage your fatigue?

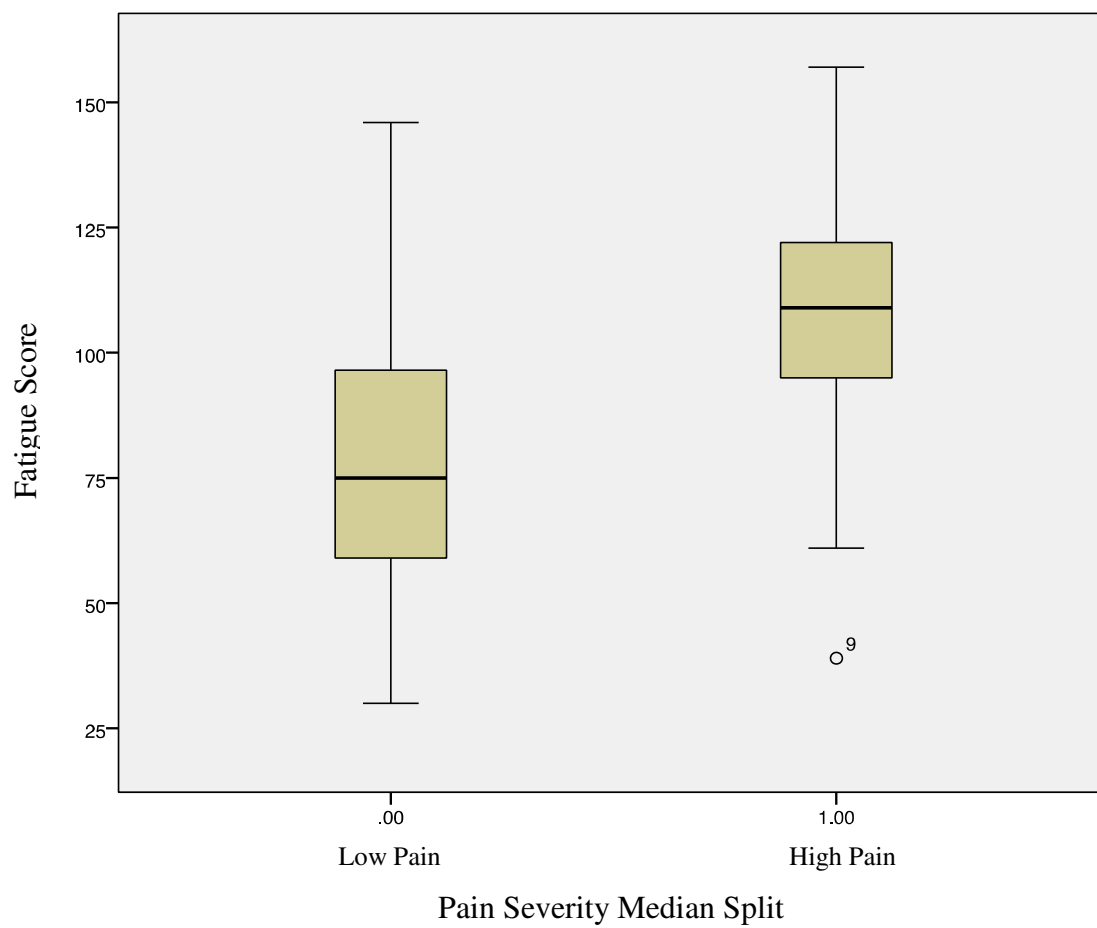
36. What else could you do to reduce or manage your fatigue?

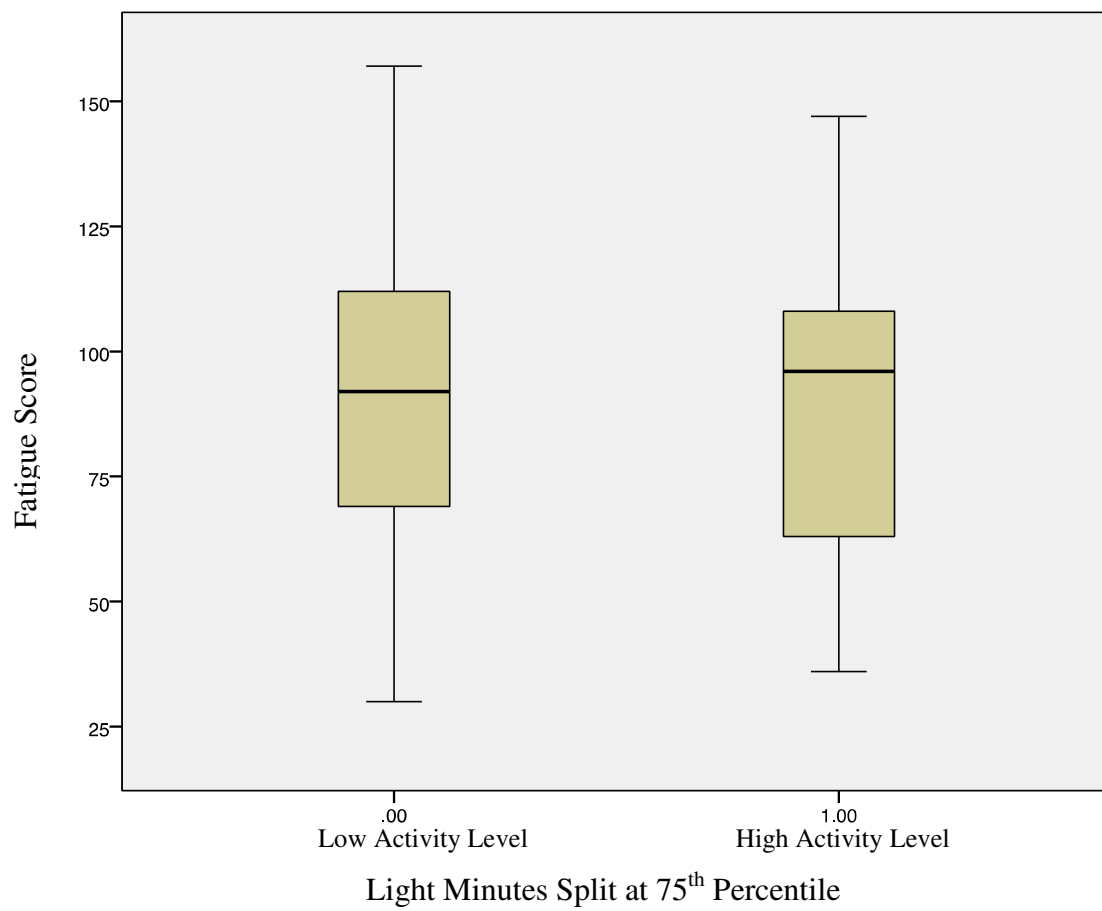
37. What could other people do to help reduce your fatigue?

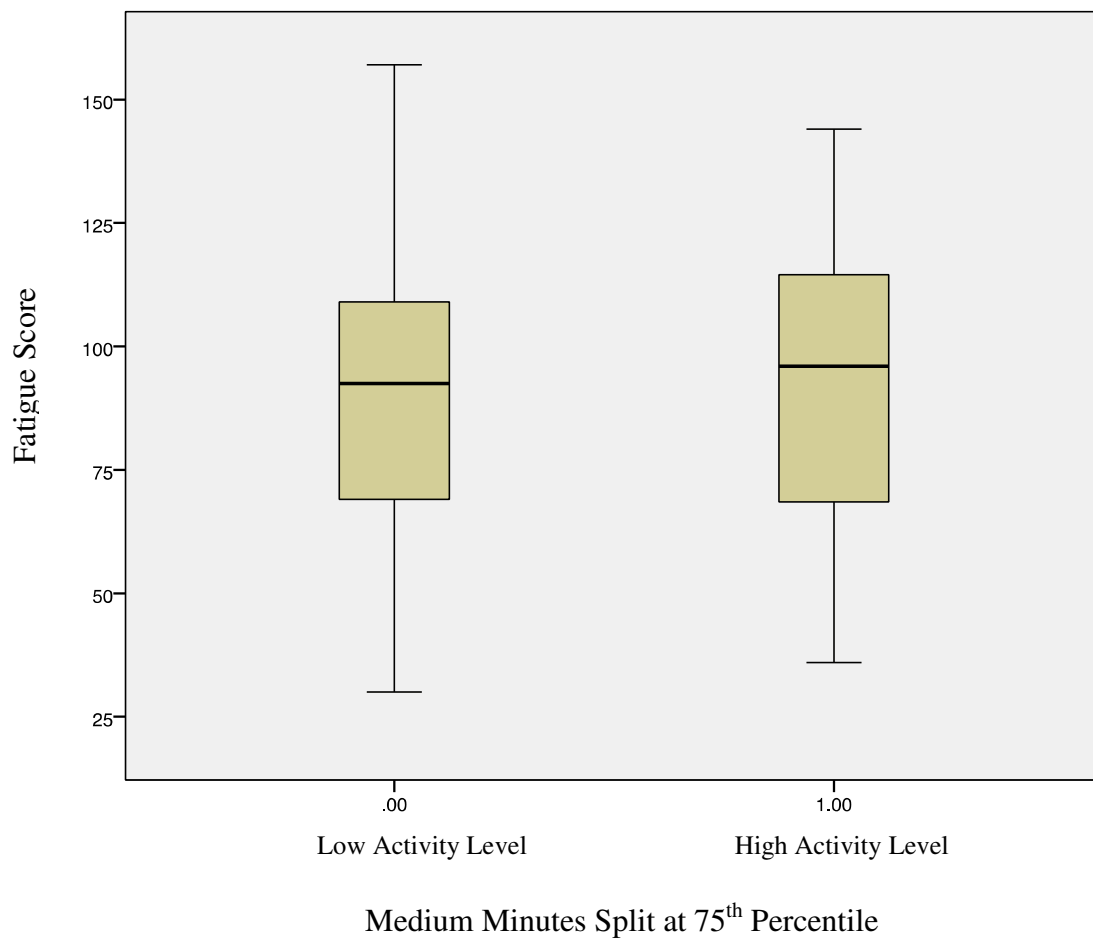
***Appendix 6-K Boxplots of Descriptive Results for Fatigue Score by Gross Motor Function Classification System Level, Pain and Physical Activity***



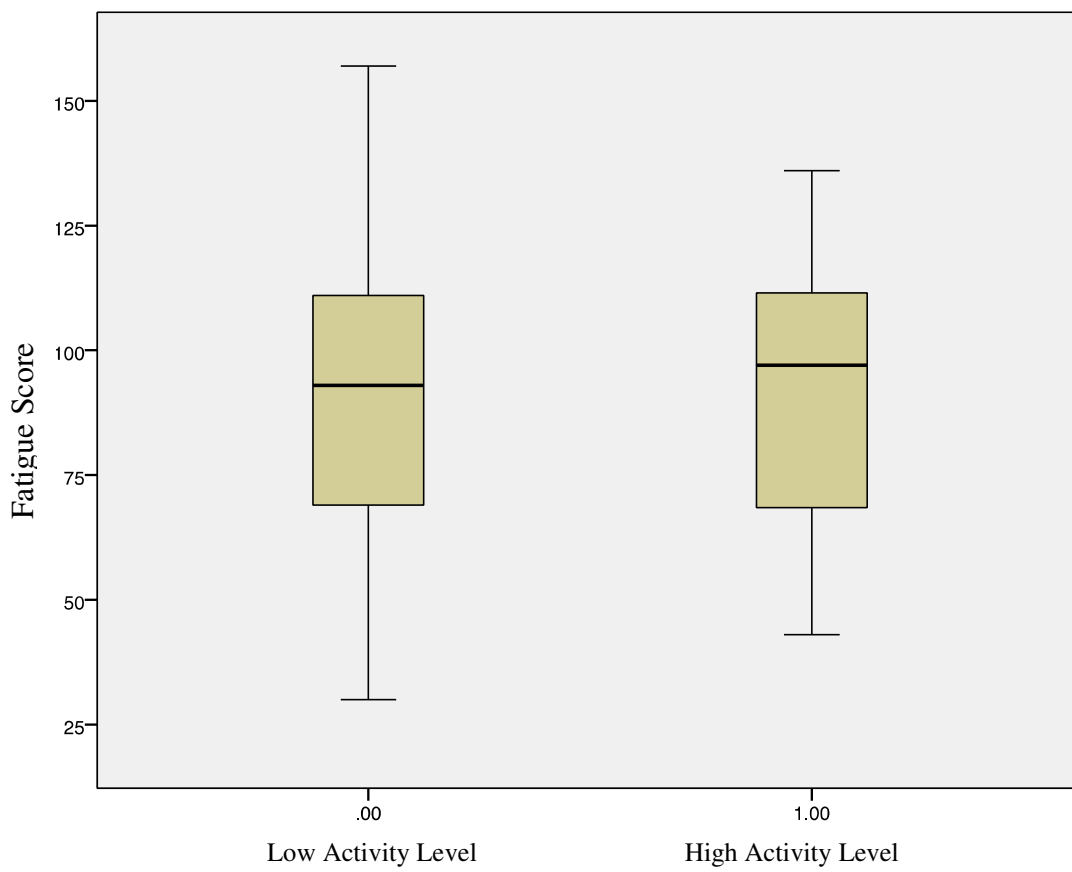












Hard Minutes Split at 75<sup>th</sup> Percentile

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**Publications:**

Falk B., Brunton L., Dotan R., Usselman C., Klentrou P., Gabriel D. Muscle Strength and Contractile Kinetics of Isometric Elbow Flexion in Girls and Women. *Ped. Exerc. Sci.*, 21(3):354-364, 2009.

Falk B., Usselman C., Dotan R., Brunton L., Klentrou P., Shaw J., Gabriel D. Child-adult Differences in Muscle Strength and Activation Pattern During Isometric Elbow Flexion and Extension. *Appl. Physiol. Nutr. Metab.*, 34:609-615, 2009.

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Brunton LK., Bartlett DJ. The Bodily Experience of Cerebral Palsy: A Journey to Self-Awareness. *Disabil Rehabil.* 35(23): 1981-1990, 2013.

**Additional Scholarly Products:**

Brunton L., Bartlett D. Psychometric Properties of Two Abbreviated Versions of the Gross Motor Function Measure for Children with Cerebral Palsy. *Developments, Pediatric Division Bulletin*, May 2010, Issue 2.

Regan, S., Orchard, C., Khalili, H., Brunton, L., Leslie K. Policy Analysis of Interprofessional Collaborative Requirements under Bill 171 and Bill 179. Technical Report submitted to Ontario Health Human Resources Research Network, March 31<sup>st</sup> 2012.

Brunton, L., & Bartlett, D. (2012). Understanding the Impact of Fatigue on Adolescents and Young Adults with Cerebral Palsy (Facts To Go, Volume 8 Issue 1). London, ON: Thames Valley Children's Centre.

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