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## Development of holistic classification systems for children with cerebral palsy

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A thesis submitted in partial fulfillment of the requirements for the Doctor of Philosophy degree in Health and Rehabilitation Sciences

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## Abstract

Cerebral palsy (CP) is a complex disorder. There is a gap in the literature in classifying children with CP broadly. The purpose of this thesis was to develop holistic classification systems for children with CP. As a first step, a search was conducted to explore the strategies used to classify children with developmental co-ordination disorder and autism-spectrum disorder. Two versions of holistic classification systems named the body function index in cerebral palsy (BFI-CP) versions I and II were developed using two methods. Then, the relationships and differences among the developed classification systems and the Gross Motor Function Classification System (GMFCS) were explored. Next, differences among subsets of the classifications that did not correspond to the ordinal levels of the GMFCS were explored. Next, the relationships between the developed classification systems (BFI-CP I and II) and the GMFCS and the change in outcome of motor function were explored. Exploration of the existing classification systems of childhood disorders (Chapter 2) demonstrated that none of the classification systems in CP addressed the majority of the key features in the international consensus definition of CP. The BFI-CP I was developed using a summing technique and the BFI-CP II was developed using cluster analysis. The findings demonstrated a strong correlation between the BFI-CP I and the GMFCS ( $r=0.92$ ), the BFI-CP II and the GMFCS ( $r=0.93$ ), and the BFI-CP I and II ( $r=0.92$ ), all ( $p<0.001$ ). There was a significant difference between the BFI-CP I and the GMFCS ( $\chi^2 = 670.49$ ,  $df=16$ ,  $p<0.001$ ) and the BFI-CP II and the GMFCS ( $\chi^2 = 685.57$ ,  $df=16$ ,  $p<0.001$ ). There was a statistically significant but weak correlations between the BFI-CP I, BFI-CP II and the GMFCS and the change in outcome of motor function based on the 50% probability that

children developed 'better than expected', 'as expected', or 'more poorly than expected' over the period of one year. The heterogeneity of the health condition of CP increases the challenges in predicting the change in gross motor function using a holistic classification system. Every child's unique features should be monitored individually to understand the strengths and weaknesses and make decisions in treatment planning.

Keywords: holistic classification, cerebral palsy, comprehensive subgrouping.

## Co-authorship Statement

Chapters 1 and 2: Introduction and Understanding issues in identifying subgroups of children with heterogeneous conditions by investigating two childhood conditions chapters were combined together as “Perspectives on classification of selected childhood neurodisabilities based on a review of literature” and has been published in *Developmental Neurorehabilitation*, 2016; 8:1-13. Dr. Bartlett contributed to this chapter through review of interpretations and critical review of the chapter and manuscript.

Chapter 3: Development of a holistic classification in children with cerebral palsy was co-authored by Dr. Bartlett. It has been submitted as “Subgrouping children with cerebral palsy from a broader perspective using two methods” in *Physiotherapy Theory and Practice*. Dr. Bartlett contributed to the design, provided access to data collected in a previous study, assisted with interpretation, and critically reviewed the chapter and manuscript.

Chapter 4 and 5: Prognostic implications of the holistic classifications in children with cerebral palsy and Summary, implications, and conclusion chapters will be combined together and will be submitted to *Pediatric Physical Therapy*. Dr. Bartlett contributed to the design, provided access to data collected in a previous study (Move & PLAY study), assisted with interpretation, and critically reviewed the chapters.

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## List of Abbreviations

ADI – R - Autism Diagnostic Interview Revised  
APTA - American Physical Therapy Association  
ASD - Autism Spectrum Disorder  
BFI-CP - Body Function Index in Cerebral Palsy  
BFMF - Bimanual Fine Motor Function  
BOTMP - Bruininks Oseretsky Test of Motor Proficiency  
CELF - Clinical Evaluation of Language Fundamentals  
CFCS - Communication Function Classification System  
CIHR - Canadian Institute of Health Research  
COMPS - Clinical Observations of Motor & Postural Skills  
CP - Cerebral Palsy  
CSI - Comprehensive Severity Index  
DCD - Developmental Co-ordination Disorder  
DSM - Diagnostic & Statistical Manual of Mental Disorders  
DT-VMI - Developmental test of Visual Motor Integration  
DTVP - Developmental Test of Visual Perception  
EASE - Early Activity Scale for Endurance  
ECAB – Early Clinical Assessment of Balance  
EEG - Electro Encephalogram  
FSA - Functional Strength Assessment  
G – Generalizability co-efficient  
GED – General equivalency diploma  
GMFCS - Gross Motor Function Classification System  
GMFM – Gross Motor Function Measure  
GMFM – 66 - B & C - Gross Motor Function Measure – 66 - Basal and Ceiling  
ICC - Intraclass Correlation Co-efficient  
ICD - International Classification of Diseases  
ICF - International Classification of Functioning, Disability and Health  
ICF-CY - International Classification of Functioning, Disability and Health – children and youth  
IQ – Intelligence Quotient  
K - Kappa  
KAT - Kinaesthetic Acuity Test  
MABC - Movement Assessment Battery for Children  
MACS - Manual Ability Classification System  
MAI – AR - Movement Assessment of Infants – Automatic Reactions section  
MAS - Modified Ashworth Scale  
MRI - Magnetic Resonance Imaging  
MVPT - Motor Free Visual Perception Test  
NA - Not Applicable  
PBE – Practice Based Evidence  
PBS - Pediatric Balance Scale  
PDD - Pervasive Developmental Disorders

PPVT - Peabody Picture Vocabulary Test  
r – Pearson's correlation co-efficient  
SAROMM - Spinal Alignment & Range of Motion Measure  
SC - Swedish Classification  
SCPE - Surveillance of Cerebral Palsy in Europe  
TLT - Tower of London Test  
VABS - Vineland Adaptive Behaviour Scale  
VMI – Visual Motor Integration  
WAIS – R - Wechsler Adult Intelligence Scale Revised  
WHO - World Health Organization  
WISC- Wechsler Intelligence Scale for Children  
Y - Years

## Chapter 1: Introduction

### Statement of the Research Problem

#### Rationale and justification for the study

Cerebral palsy (CP) is a non-progressive disorder of movement and posture which occurs in the early childhood period accompanied by secondary conditions and comorbidities. Children with CP present with heterogeneous features which increases the complexity in understanding the presentation of this condition. Classification systems in CP provide clinicians and researchers a way to sort or subgroup children so that their similarities and differences can be better understood, which in turn influences clinical decision making. Traditional classification systems are not helpful in making decisions about treatment planning and the prevailing functional classification systems do not classify children with CP from a holistic perspective. The proposed work aims to fill these gaps and increase knowledge in understanding subgroups of children with CP. The main objective of this dissertation was to develop and explore the prognostic implications of two holistic classifications for children with CP.

#### Significance of the study

The product of this work is intended to increase understanding of subgroups of children with CP and facilitate communication between the health care professionals and

A version of this chapter has been published. (Jeevanantham D, Bartlett D. Perspectives on classification of children with childhood neurodisabilities based on a review of literature. *Dev Neurorehabil.* 2016; 8:1-13. [Epub ahead of print])

among the health care professionals and parents and policy makers. This work may also help in planning effective rehabilitation strategies based on expected outcomes for groups of children with different characteristics. Knowledge derived from this work may also contribute to parents' expectations of providing intervention tailored to the unique characteristics of their children with CP, and may have a role in enhancing effective, efficient, and family-centred care. The results of this work may also contribute to service providers', parents' and policy makers' decision making on selection of services. Finally, the products of this work are expected to have applications for clinical practice, administration, teaching, and research.

## Background information on Cerebral Palsy

This chapter is focused on the International Classification of Functioning, Disability and Health (ICF), background information on cerebral palsy, the extent of the problem, and a brief discussion on the prevailing traditional and functional classification systems in this health condition.

The World Health Organization (WHO)'s ICF is a comprehensive framework of disability that provides a standard language for describing the health state of an individual.<sup>1</sup> The ICF covers all aspects of health (health domains) and some aspects of health-related well-being (health-related domains). It organizes information in two parts: *functioning and disability* and *contextual factors*. The components of functioning and disability include: (1) body structure and body functions, (2) activity, and (3) participation. The components of contextual factors include (1) environmental factors and

(2) personal factors. The ICF constructs of body structure and body function are described by variations in *body structure (anatomical)* and *body function (physiological)*. The ICF constructs of activity and participation are described in terms of *capacity* and *performance*. *Capacity* refers to an individual's ability to execute a task in a standard environment, whereas *performance* refers to the ability of the individual to execute a task in real life situations.<sup>1</sup> The environmental factors in this context include all aspects of the physical, social, and attitudinal world.<sup>1</sup> *Capacity* reflects what a child can do when an environment is standardized. The difference between the *capacity* and *performance* reflects the impact of the environment and provides guidance on potential modifications that could be done to the environment to facilitate *performance*.<sup>1</sup> Therefore in this thesis the terms *capacity* and *performance* are used rather than activity and participation. Even though *personal factors* are one of the components of the ICF, they are not classified in its entirety due to social and cultural variability. For example, beliefs, practices, and personal characteristics are personal features that are taken up differently in different cultures which prevent a shared understanding and approach.<sup>1</sup> Throughout this chapter, the identified classification systems are linked to the ICF wherever possible.

CP is the most common cause of childhood physical disability.<sup>2</sup> It occurs in 2 to 2.5 per 1000 live births.<sup>3</sup> According to the international consensus definition of CP, "Cerebral palsy (CP) describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy, and by secondary musculoskeletal



problems”.<sup>4 pg9</sup> Classification is a systematic way of assigning data, persons, or objects into categories on the basis of common characteristics.<sup>5</sup> Categorization of individuals with CP assists with decreasing the complexity in understanding and describing clinical manifestations. The purpose of classification primarily includes description, prediction, and comparison.<sup>4</sup> Morris has stated “after more than 150 years of debate we do not have an agreed method for classifying the impairment that has been shown to be robust in terms of validity and reliability”.<sup>6 pg6</sup> The debate on classification of CP continued for many years and numerous classification systems have been proposed and refined.

The prevailing classification systems of CP used in rehabilitation can be broadly divided into three categories: (1) topographical classification (i.e distribution of involvement), (2) classification based on type of motor disorder, and (3) functional classification. Table 1.1 contains a description of these classifications.

**Table 1.1: Classification systems of cerebral palsy**

<b>Authors</b>	<b>Constructs</b>	<b>Classification</b>	<b>ICF Construct</b>
Reid et al. <sup>7</sup>	Distribution of involvement	Monoplegia Hemiplegia Diplegia Triplegia quadriplegia	Body function
Surveillance of Cerebral Palsy in Europe <sup>8</sup>	Motor disorder	Spastic - Unilateral Spastic Cerebral Palsy - Bilateral Spastic Cerebral Palsy Ataxia Dyskinetic - Dystonic - Choreo athetotic	Body function
Westbom et al. <sup>9</sup>	Motor disorder (i.e Swedish Classification)	Spastic - Hemiplegia - Diplegia - Tetraplegia Ataxic - Diplegia - Simple Ataxia Dyskinetic - Dystonia - Choreo athetosis - Athetosis and dystonia Mixed	Body function
Palisano et al. <sup>10</sup>	Gross motor function	Level I – Walks without limitation Level II – Walks with limitation Level III - Walks using a hand-held mobility device Level IV - Self-mobility with limitations; may use powered mobility Level V - Transported in a manual wheelchair	Performance
Beckung et al. <sup>11</sup>	Manual function	Level I – (a) One hand: manipulates without restrictions. The other hand: manipulates with restrictions or limitations in more advanced fine motor skills Level II – (a) One hand: manipulates without restrictions. The other hand: only ability to grasp or hold (b) Both hands: limitations in more advanced fine motor skills Level III – (a) One hand: manipulates without restrictions. The other hand: no functional ability b) One hand: limitations in more advanced fine motor skills. The other hand: only ability to grasp or worse Level IV - (a) Both hands: only ability to grasp b) One hand: only ability to hold. The other hand: only ability to hold or worse Level V - Both hands: only the ability to hold or worse	Capacity
Eliasson et al. <sup>12</sup>	Manual function	Level I – Handles objects easily and successfully Level II – Handles most objects but with somewhat reduced quality and/or speed of achievement Level III - Handles objects with difficulty; needs help to prepare and/or modify activities Level IV - Handles a limited selection of easily managed objects in adapted situations Level V - Does not handle objects and has severely limited ability to perform even simple actions	Performance
Hidecker et al. <sup>13</sup>	Communication function	Level I - Sends and receives with familiar and unfamiliar partners effectively and efficiently Level II - Sends and receives with familiar and unfamiliar partners but may need extra time Level III - Sends and receives with familiar partners effectively, but not with unfamiliar partners Level IV - Inconsistently sends and/or receives even with familiar Partners Level V – Seldom effectively sends and receives, even with familiar partners	Performance

Topographical classification is widely used and it classifies children with CP into the following types based on the distribution of involvement: monoplegia, hemiplegia, diplegia, triplegia, and quadriplegia.<sup>7</sup> This classification identifies subgroups based on the number of limbs involved and falls under the 'body structure and function' ICF construct. Imprecisions and inconsistencies have been reported in using the topographical classification descriptors.<sup>7,14</sup> The topographical classification has poor reliability (K= - 0.01 to 0.59).<sup>15</sup>

The Surveillance of CP in Europe (SCPE)<sup>8</sup> and the Swedish Classification (SC)<sup>9</sup> are two classifications that use motor type to group children with CP. Although these classifications are also widely employed, the motor disorder classifications (K = 0.1 to 0.35) have poor reliability.<sup>15</sup> The SCPE classified children with CP into four categories (See Table 1.1) primarily based on predominant motor disorder.<sup>8</sup> More recently, SCPE has recommended using functional classification systems to describe functional performance.<sup>16</sup> The SCPE classification does not provide information on coexisting neurological and musculoskeletal findings, and has a moderate level of agreement (K=- 0.59) for including a child as a CP case in the SCPE database.<sup>17</sup> Work on improving the reliability of the SCPE system is described as in progress.<sup>17</sup> Recently, Sellier and colleagues studied the inter-rater reliability of the SCPE system using video observations (K = 0.85) and written vignettes (K =0.78).<sup>18</sup> These findings are supported by Randall and colleagues (K = 0.84).<sup>19</sup> The SC is a combination of the type of motor disorder and the topographical pattern (See Table 1.1), and is in practice since the start of a clinical follow-up programme in combination with a health care quality database program.<sup>9</sup> The traditional classification systems, which classify children with CP primarily based on

muscle tone, and/or type of motor disorder, lack evidence, and have poor reliability and poor prognostic value.<sup>7</sup> The underlying framework of both of the motor disorder classifications is the 'body structure and function' dimension of ICF.

More recently, efforts have been made to classify children with CP based on their functional profiles. The Gross Motor Function Classification System (GMFCS),<sup>10, 20</sup> Bimanual Fine Motor Function (BFMF),<sup>11</sup> Manual Ability Classification System (MACS),<sup>12</sup> and Communication Function Classification System (CFCS)<sup>13</sup> (see Table 1.1) are four functional classification systems that classify children with CP based on their functional abilities in everyday life.

The GMFCS,<sup>20</sup> along with its recently revised and expanded version,<sup>10</sup> serves as a standard tool that classifies children primarily based on self-initiated movement. It is a five-point ordinal- level classification system which has specific descriptions for five different age bands. Children in level I are completely independent in walking, running, and other gross motor functions; however, the speed at which they perform gross motor functions may be reduced. Children in level V are completely dependent. The psychometric properties of the GMFCS have been extensively investigated. Content validity of the second version of the GMFCS was most recently explored using the following two consensus methods: the nominal group process through group discussions via teleconferences and a Delphi survey in which iterations to questions were done online.<sup>10</sup> This expanded and revised version of the GMFCS has an excellent agreement between parents and physiotherapists with an intraclass correlation coefficient (ICC) of 0.96 for children between 4 to 18 years of age.<sup>21</sup> The use of the GMFCS to classify children under 2 years of age has to be done with caution due to a lower inter-rater

agreement ( $K=0.55$ ) compared to children older than 2 years of age ( $K=0.75$ ).<sup>20</sup> The GMFCS is stable over a period of one year<sup>22</sup> as well as following single-event multilevel surgery.<sup>23</sup>

The BFMF is a bimanual grading system of fine motor function.<sup>11</sup> Since its publication in 2002, only one study has explored the reliability of the BFMF and reported an excellent correlation coefficient as determined by a Kappa value of 0.98.<sup>19</sup> Elvrum et al. recently explored the construct and content validity of the BFMF.<sup>24</sup> They found excellent correlation between the BFMF and the MACS (Spearman's  $\rho=0.89$ ). The content validity of the BFMF was explored through literature review and using the International Classification of Functioning, Disability and Health (ICF-CY)<sup>25</sup> framework to compare the BFMF with the MACS.

The MACS is a recently developed tool for classifying children's ability to perform bimanual activities of daily living.<sup>12</sup> The MACS is analogous to the GMFCS in that it is a five-level classification system that classifies children based on self-initiated performance; however, the MACS does not contain specific age bands and the levels are ascertained with respect to children's appropriate developmental activities.<sup>12</sup> Content validity of the MACS was analyzed using a consensus process and qualitative methodology.<sup>12, 26</sup> The agreement between therapists as analyzed using the intraclass correlation coefficient was high ( $ICC = 0.97$ ) for ages between 4 and 18 years.<sup>12</sup> Concurrent validity was explored by correlations with the Functional Independence Measure for Children ( $r = -0.78$ )<sup>27</sup> and the Pediatric Evaluation of Disability Inventory ( $r = -0.72$ )<sup>28</sup> and were statistically significant for both. The MACS was stable over a one-

year interval in children with CP aged between 4 and 17 years with an ICC value of 0.97.<sup>29</sup>

The CFCS is a recently developed tool for categorizing the communication ability of children with CP with familiar and unfamiliar partners.<sup>13</sup> The CFCS is also a five-level classification system and the levels are determined based on the child's ability to communicate by using any method of communication in a real life situation. Preliminary evidence on psychometric properties of the CFCS has been reported.<sup>13</sup> Content validity was explored by consultation with expert groups, using both the nominal group process and the Delphi technique. Intra-rater reliability was 0.82 and inter-rater reliability was 0.66, as measured using the Kappa co-efficient.<sup>13</sup>

The GMFCS, the MACS, and the CFCS map to *performance* as they focus on real life situations whereas the BFMF maps to the *capacity* construct of the ICF as it focus on what the child can do rather than what the child usually does.

Children with CP exhibit heterogenous features and the prevailing classification systems categorize children with CP primarily based on any one feature. Before deciding on ways of classifying children with CP holistically, it is useful to study prevailing classification systems in other selected childhood conditions to understand the strategies associated with identifying subgroups that might be useful for clinical decision making. The term holistic classification used in this thesis refers to “a classification that addresses the majority of the key features of CP described in the international consensus definition<sup>4</sup>”.

The biological plausibility of considering CP and Developmental Co-ordination Disorder (DCD) as a continuum of movement disorder is still under debate.<sup>30</sup> Also, a recent study on prevalence of the co-occurrence of CP and Autism Spectrum Disorder (ASD) estimated that almost 7% of children with CP had a co-occurrence of ASD, and specifically the frequency of co-occurrence of ASD was higher (18.4%) in children with non-spastic CP.<sup>31</sup> Although CP, DCD, and ASD are three different conditions, the features of heterogeneity and co-occurrence provided inspiration to study the subtypes of these two neurodisabilities (ie. DCD and ASD) in detail which may assist in selecting appropriate methods for developing a holistic classification system for children with CP. The next chapter is focused on describing classification systems in children with two other neurodisabilities (i.e DCD and ASD).

## Chapter 2: Understanding issues in identifying subgroups of children with heterogeneous conditions by investigating two childhood conditions

This chapter is focused on understanding prevailing classification systems in children with Developmental Co-ordination Disorder (DCD) and Autism Spectrum Disorders (ASD), interpreting the identified classification systems in terms of the utility of the classification systems, and identifying gaps in the literature.

### Introduction

Children with the two selected childhood conditions (i.e DCD and ASD) are diverse in clinical presentation and comorbidities. DCD is an idiopathic neurodevelopmental disorder that occurs in isolation or as a co-morbidity with other neurodevelopmental and neurobehavioural disorders, which complicates the diagnosis of this disorder.<sup>32</sup> According to the Diagnostic and Statistical Manual of Mental disorders (DSM IV), children with DCD demonstrate marked disturbance in development of motor co-ordination to the extent that it interferes with academic performance as well as activities of daily living, in the absence of other medical conditions and pervasive developmental disorder.<sup>33</sup> According to the European Academy for Childhood Disability, DCD is better defined by the DSM IV criteria than ICD-10 criteria, leading to the recommendation to use developmental coordination disorder as the official terminology

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for use in the English language.<sup>32</sup> The estimated prevalence of DCD ranges from 5 to 8% of all school aged children.<sup>33</sup>

According to the DSM - IV,<sup>33</sup> the ASD are referred to as pervasive developmental disorders (PDD) which include five disorders: autistic disorder, Rett's syndrome, childhood disintegrative disorder, Asperger's disorder, and PDD not otherwise specified. The uncertainty associated with the diagnostic standard that categorizes the subtypes made researchers advocate for an umbrella term. The subsequent version, DSM V,<sup>34</sup> uses the term ASD and eliminates the use of other diagnoses including Asperger's disorder, autism, PDD, and childhood disintegrative disorder. As stated by DSM IV, ASD consist of 3 domains: (1) abnormalities in social interaction, (2) communication deficit, and (3) repetitive behaviours and restricted interests. The DSM V, however, consists of only two domains in which the social interaction and communication domains are combined together as one domain (social communication) and the other domain is repetitive behaviours and fixated interests. In order to be diagnosed with ASD, the child will have exhibited these symptoms from early childhood. The estimated prevalence of children with ASD ranges from 4.8–21.2 per 1,000 children under 8 years of age.<sup>35</sup>

The aims of this chapter are: (1) to identify various classification systems of the two neurodisability conditions and align them to the International Classification of Functioning, Disability and Health (ICF), (2) to analyze the utility of the identified classification systems including those for CP, and (3) to propose a method for developing a holistic classification system for children with CP.

## Methods

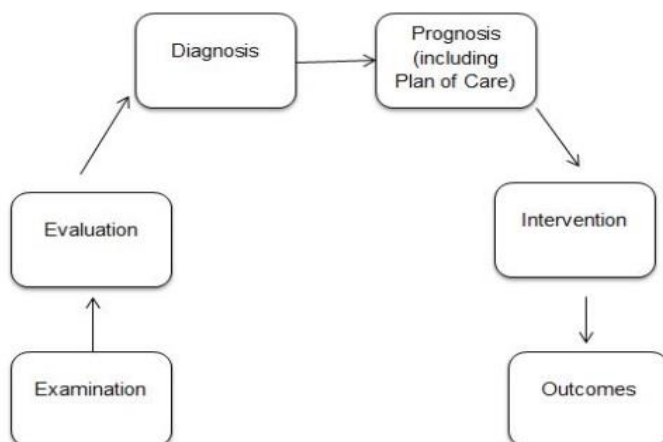
A literature search aiming to identify different subgrouping systems in DCD and ASD was done in 6 different databases and Google using a combination of terms. Detailed description of the database searches and the procedure is provided in Appendix 2.1. The results of the search of the prevailing classification systems of DCD and ASD in terms of constructs, measures used, as well as how they align with the ICF are provided in tables under corresponding sections. The term clinical utility used in this thesis refers to the usefulness of a measure/classification in clinical practice determined using criteria selected based on experience. The clinical utility of classification systems for use with children with neurodisabilities was determined through a combination of the psychometric properties of the underlying measures and/or classification systems, the specific purposes for which they are used in practice, their focus on both functioning and development, and the time and resources required to obtain a classification. Guidelines for determining the adequacy of psychometric properties, description of the range of purposes that classification systems can have in rehabilitation practice, review of the role of both functioning and development, and description of the duration and availability of resources required to establish a classification system are outlined in the following paragraphs.

Strong measurement properties are an essential criterion that determine the clinical utility of an instrument. Reliability refers to the degree of consistency of a measure (within a rater, between raters, or within a participant over time) and validity refers to the degree to which an instrument measures what it is designed to measure. The

ICC is an indicator of reliability with values ranging from 0.00 to 1.00. ICC values greater than 0.75 indicate good reliability and the ICC values less than 0.75 indicate moderate to poor reliability.<sup>36</sup> Validity is typically determined by the magnitude of relationship between the measures of other constructs. Correlation co-efficients greater than 0.75 indicate a strong correlation, those between 0.50 and 0.74 indicate a moderate correlation, and those below 0.50 indicate a weak correlation.<sup>37</sup> Appendices 2.2 and 2.3 contain details of the psychometric properties of the tests used in the identified studies. In many cases, the identified studies used older versions of the tests. In summarizing the psychometric properties, I err on the side of being conservative for measures with values reported in ranges.

Whereas physicians use the ICD to label and identify a disorder,<sup>5</sup> rehabilitation practitioners explore the functional ability of a person with a health condition using the ICF<sup>1</sup> for many purposes. The American Physical Therapy Association (APTA)<sup>38</sup> provides a useful framework to establish a high quality, standardized patient care approach facilitating functional independence. Figure 2.1 illustrates the framework proposed by the APTA. Classification of a health condition occupies a primary part in the examination element in which the rehabilitation practitioner administers various tests and measures to obtain data to assist in identifying a subgroup in which individuals' best fit. A stable classification predicts the prognosis of a health condition which, along with the examination element, helps in planning appropriate effective and efficient interventions for subgroups of people with similar characteristics, ultimately leading to optimal outcomes.

Figure 2.1: Reprinted from Guide to Physical Therapist Practice. 2<sup>nd</sup> ed. Phys Ther. 2001; 81:9-744, with permission of the American Physical Therapy Association. Copyright © 2001 American Physical Therapy Association.<sup>38</sup>



Whereas the components of functioning of the ICF were summarized in the previous section, the next focus is on the extent to which classification in the three selected neurodisabilities attend to the criterion of development, as suggested by the International Classification of Functioning, Disability and Health – Children and Youth (ICF-CY).<sup>25</sup> Specifically I contrast the extent to which various classifications attend to age-related changes.

The training, procedures, and time required to establish a subgroup and the availability of resources are final criteria that determine the utility of a classification system. This section identifies a range of subgrouping systems: from classification systems that are freely available online that take only minutes to complete, to commercial products that are expensive to purchase and require extensive training to learn to administer and requires significant time (hours and probably days) to both administer and

score. The time required to reconcile each child's pattern of scores to obtain a classification requires even more additional time.

## Results

### Subtypes of Developmental Co-ordination Disorder

The search yielded the following three classification systems used in children with DCD. Table 2.1 provides details of the results of the three subtyping systems of DCD. Detailed descriptions of the psychometric properties of the measures used in these identified studies are provided in Appendix 2.2.

Macnab and colleagues identified five clusters of children with DCD using constructs such as kinaesthetic acuity, visual-perception, visual motor integration, manual dexterity, balance, and complex gross motor tasks analyzed using different measures.<sup>39</sup> Green and colleagues established five clusters by studying constructs including manual dexterity, visual-spatial skills, motor skills, postural skills, and kinaesthesia.<sup>40</sup> Green et al. also attempted to analyze the predictive validity of the classification and found limited predictive value. Vaivre-Douret and colleagues identified three subtypes by studying neuropsychological, neuro-psychomotor, and neuro-visual examination constructs using a variety of tests and measures.<sup>41</sup> They identified the subgroups using inferential clinical analysis and validated the results using factor analysis and cluster analysis.

**Table 2.1: Subtypes of Developmental Co-ordination Disorder**

Authors	Constructs	Measures	ICF constructs (Overall Best fit)	Classification system
Macnab et al. <sup>39</sup> (2001)	Kinaesthetic Acuity	Kinaesthetic Acuity Test	Body Function	<p><b>Good Balance</b> – Normal standing balance and visual perception</p> <p><b>Good visual-motor</b> – Good performance on measures of upper-limb speed and dexterity, visual motor integration, and visual perception and poor performance on measures of kinaesthetic acuity and balance</p> <p><b>General perceptual-motor</b> – Severe difficulty in all areas</p> <p><b>Poor fine motor/visual motor</b> – poor performance in fine motor skills, visual motor integration, and visual perception</p> <p><b>Poor gross motor</b> - Poor performance on the complex gross motor subtest (measured using the running speed and agility subtest of BOTMP)</p>
	Visual-Perception	The Motor Free Visual-Perception Test	Body Function	
	Visual-motor integration	Developmental Test of Visual Motor Integration	Body function in the context of capacity	
	Manual Dexterity (Upper Limb Speed and Dexterity subtest)	Bruininks Oseretsky Test of Motor Proficiency	Body function and capacity	
	Complex gross motor task (Running Speed and Agility subtest)			
	Balance	Test of Motor Impairment	Body Function and Capacity	
Green et al. <sup>40</sup> (2008)	Manual dexterity and balance*	Movement Assessment Battery for Children (M-ABC)	Body Function and Capacity	<p><b>Relative strength across perceptual-motor items</b> – Lower scores for Kinesthetic acuity, than the visual motor integration and visual subtests, manual dexterity, and static and dynamic balance.</p> <p><b>Relative strength in perceptual functions and fine motor skills</b> – Better scores on kinaesthetic acuity, visual motor integration and visual subtest, manual dexterity, and dynamic balance.</p> <p><b>Poor static and dynamic balance</b> – Relative weakness in visual perceptual skills, and static and dynamic balance. Better scores on Visual Motor Integration and Visual subtest, Manual dexterity, and kinaesthetic acuity.</p> <p><b>Poor perceptual and fine motor tasks</b> – Poor scores on visual spatial, kinaesthesia, manual dexterity items. Relative strength in balance items.</p> <p><b>Poor across all items</b></p>
	Visual-spatial*	Developmental Test of Visual-Motor Integration	Body function in the context of capacity	
	Motor, postural skills and Kinaesthesia*	The Clinical Observations of Motor and Postural Skills	Body Function	

Vaivre-Douret et al. <sup>41</sup> (2011)	Neuropsychological	Wechsler measure of Intelligence	Capacity	<b>Ideomotor dyspraxia</b> – abnormalities for crawling, digital praxis, slowness, imitation of gestures, digital gnosis, dynamic balance, body spatial integration, handwriting, hypotonia, abnormalities in standing tone and homogeneous tonic laterality, and visual pursuits. No impairment of the pyramidal tract motor pathway or manual dexterity, or visual perceptual motor or VEP disorder. <b>Visual spatial and visual constructional dyspraxia</b> – abnormalities in puzzles, visual motor integration, visual spatial structuring, lego blocks, arithmetic, visual spatial constructional tasks, handwriting, vertical pursuit, and visual refraction. <b>Mix dyspraxia</b> – abnormalities in all measures
		Block Design	Capacity	
		Manual Copy and Visual Spatial Memory of A Complex Geometric Figure	Body function and capacity	
		Developmental Test of Visual-Motor Integration	Body function in the context of capacity	
		Bell Crossing test	Body function	
		Porteus Labyrinth test	Capacity	
		Tower of London	Body function	
		Developmental test of visual perception	Body function tasks	
		Hand writing scale	N/A	
		Language Screening battery	N/A	
	kinaesthetic perception	Body function		
	Neuro-psychomotor	Neuro-psychomotor Functions in Children	Body function and capacity	
	Neuro-visual examination	Electroretinogram	Body structure	
Visually Evoked Potentials		Body function		
Motor Electro-Oculogram		Body structure		

\* Constructs determined by us

ICF – International Classification of Functioning, Disability and Health, N/A = Not applicable as the tool was not located

Different authors used a variety of instruments to measure the specific constructs of interest (Table 2.1). Certain tests and measures are very straight forward in determining their underlying ICF constructs (eg. Wechsler Intelligence Scale for Children is a *capacity* measure). However, some measures examined the *body function* construct of the ICF in the context of *capacity* (eg. Visual Motor Integration (VMI) Test). The VMI test is used to assess the ability of an individual to integrate the visual and motor abilities which involve copying simple designs. Copying is a *capacity* construct (according to ICF) whereas visual motor integration is a *body function* construct (according to ICF). From my perspective, such measures map to the *body function* in the context of *capacity* (ICF constructs).

In both Macnab's<sup>39</sup> and Green's studies,<sup>40</sup> the subgroups appear relatively similar as they both used similar constructs and some similar measures. The most similar subgroups include: the *general perceptual-motor* cluster in Macnab's study and the *poor across all items* cluster in Green's study, both of which were characterized by poor scores across all items (Table 2.1). Green et al. in addition to identifying the subgroups also attempted to analyze the predictive validity of the subgrouping system and found limited predictive value. Both Macnab and Green teams studied dynamic balance, gross and fine motor skills, and perceptual motor skills, whereas, Vaivre-Douret and colleagues<sup>41</sup> studied neuropsychological, neuro-psychomotor and neuro-visual examination using batteries of tests. Green et al. also reported that they did not find any conclusive evidence supporting the stability of the classifications derived.



DCD is typically diagnosed using the DSM criteria and/or a combination of tests and batteries. All three studies attempted to subgroup children with DCD using cluster analysis, highlighting the complexity and heterogeneity in classifying children with DCD. Overall, the majority of the measures map onto *body function* and a few map onto *capacity* and a combination of *body function* and *capacity*. None of the measures map to *performance*.

## Subtypes of Autism Spectrum Disorders

The detailed literature search provided eight subgrouping systems in children with ASD. Table 2.2 summarizes the results of eight separate investigative teams that have explored subtypes of autism using different methods. Elaboration on the results of these studies is discussed below. Details of the psychometric properties of the measures used are contained in Appendix 2.3.

**Table 2.2: Subtypes of Autism Spectrum Disorder**

<b>Authors</b>	<b>Constructs</b>	<b>Measures</b>	<b>ICF construct (Overall best fit)</b>	<b>Classification</b>
Stevens et al. <sup>42</sup> (2000)	Interaction, communication, and restricted repetitive behavior*	Wing Autistic Disorder Interview Checklist	Performance (except one item - impairment)	1. High functioning 2. Low functioning
	Cognition*	Stanford Binnet Intelligence Scale	Capacity	
		Bayley Scales of Infant Developmental	Capacity	
	Communication*	Peabody Picture Vocabulary Test	Body function in the context of Capacity	
Social Behavior*	Vineland Adaptive Behaviour Scales	Performance		
Cuccaro et al. (2003) <sup>43</sup>	Restricted and Repetitive Behaviors	Autism Diagnostic Interview-Revised	N/A	1. Repetitive sensory-motor behaviours 2. Resistance to change
Miles et al. <sup>44</sup> (2005)	Microcephaly	Head circumference	Body structure	1. Essential Autism 2. Complex Autism
	Imaging*	Brain MRI	Body structure	
	Electrodiagnosis*	Brain EEG,	Body function	
	Language*	Vineland Adaptive Behavior Scales	Performance	
		Clinical Evaluation of language fundamentals – III	Capacity	
	Cognition*	Leiter International performance scale	Capacity	
		Wechsler Intelligence scale for Children	Capacity	
Stanford Binnet Intelligence Scale		Capacity		

The development of Autism Classification of Functioning: Social Communication (ACSF: SC) was in progress when this thesis was written; therefore not included.

		Vineland Adaptive Behavior Scales	Performance	
Liss et al. <sup>45</sup> (2006)	Sensory*	Sensory Questionnaire	Performance	1. Overfocused 2. High functioning 3. Low functioning 4. Mildly overfocused
	Attention*	Kinsbourne Overfocusing Scale	Performance	
	Socialization, communication and perseveration*	Vineland Adaptive Behavior Scales	Performance	
DSM - IV checklist		N/A		
Lam et al. <sup>46</sup> (2008)	Restricted and Repetitive Behaviors	Autism Diagnostic Interview-Revised	N/A	1. Repetitive motor behaviours 2. Insistence on sameness 3. Circumscribed interests
Rapin et al. <sup>47</sup> (2009)	Expressive phonology	Photoarticulation Test	Body Function	1. Persistent and severe impairment in expressive phonologic skills 2. Average expressive phonology
Lane et al. <sup>48</sup> (2010)	Sensory	The Short Sensory Profile	Performance	1. Sensory based inattentive seeking 2. Sensory modulation with movement sensitivity 3. Sensory modulation with taste/smell insensitivity
Anagnostou et al. <sup>49</sup> (2011)	Repetitive Behavior, Obsessions and Compulsions	The Yale Brown Compulsive Scale	Performance	1. Obsessions, 2. Higher-order repetitive behaviors 3. Lower-order repetitive behaviors 4. Hoarding

\* constructs derived by us

ICF – International Classification of Functioning, Disability and Health, N/A = Not applicable as the tool was not located

DSM - Diagnostic and Statistical Manual of Mental disorders

The development of Autism Classification of Functioning: Social Communication (ACSF: SC) was in progress when this thesis was written; therefore not included.

Stevens and colleagues identified two subgroups of autism using cluster analysis: (1) *high functioning* and (2) *low functioning* group based on cognition, communication, and social behaviour.<sup>42</sup> They also found that preschool cognitive functioning (non-verbal intelligence quotient (IQ)) is the potential predictor of school age functioning.

Cuccaro and colleagues identified two subgroups of restricted and repetitive behaviors in children with autism based on factor extraction.<sup>43</sup> Children in *factor 1* exhibited un-purposeful repetitive sensory-motor behaviors and children in *factor 2* exhibited resistance to change. The two factor subgrouping was replicated and supported by several groups of researchers including groups led by Shao,<sup>50</sup> Szatmari,<sup>51</sup> and Bishop.<sup>52</sup>

Miles and colleagues identified two subgroups of autism based purely on abnormality of morphogenesis: essential autism and complex autism.<sup>44</sup> Children with complex autism had significant dysmorphology or microcephaly and a lower IQ, more abnormal electro-encephalogram (EEG), abnormalities in magnetic resonance imaging (MRI), and identifiable autism-related syndrome. Children with essential autism were non-dysmorphic and non-microcephalic and had higher sibling recurrence, more relatives with autism, and a higher IQ, as well as fewer seizures. They also analyzed the features that best predicted poor outcomes and found that microcephaly strongly predicted poor outcome, followed by dysmorphology.

Liss and colleagues<sup>45</sup> studied the sensory and attention abnormalities in children with autism and identified four subgroups based on cluster analysis: over-focused, high

functioning, low functioning, and mildly over-focused. Children in the over-focused subgroup were over reactive to sensory stimuli, highly over-focused, and had exceptional memory for selective material and exhibited perseverative behavior. Children in the high functioning group had fewer problems. Children in the low functioning group had prominent under-sensitivity and sensory seeking. Children in the mildly over-focused subgroup had fewer autistic features, were relatively high functioning and were similar in all features to the over-focused group but were mildly over-focused.

Lam and colleagues<sup>46</sup> identified three subtypes of restricted and repetitive behaviors based on exploratory factor analysis. Children in type 1 exhibited repetitive motor behaviours and had associated social/communication deficits, children in type 2 exhibited insistence of sameness and had associated social and communication deficit, and children in type 3 had circumscribed interest and exhibited behaviours such as strong preoccupations and attachment to certain objects.

Rapin and colleagues<sup>47</sup> identified two types of language disorders in school-aged children with autism through cluster analysis based on expressive phonology and validated the cluster solution with other cognitive, social, and language measures. Children in type 1 had persistent and severe impairment in expressive phonologic skills and children in type 2 had low to better than average expressive phonology.

Lane and colleagues<sup>48</sup> studied sensory processing in children with autism and identified three subgroups using cluster analysis: sensory-based inattentive seeking, sensory modulation with movement sensitivity, and sensory modulation with taste/smell

sensitivity. Children in the sensory-based inattentive seeking category had typical sensory processing function and had attentional difficulties. Children in the sensory modulation with movement sensitivity category exhibited under and over responsiveness and had difficulty with movement function such as weak muscles, poor grasp, and low endurance. Children in the sensory modulation with taste/smell sensitivity category exhibited only sensory modulation difficulties. They also found that the sensory processing subgroups predicted communication skill and maladaptive behaviour.

Anagnostou and colleagues<sup>49</sup> derived a four-group classification of repetitive behaviours in children with autism using factor analysis: obsessions, higher-order repetitive behaviors, lower-order repetitive behaviors, and hoarding. Children in the obsessions group had fear of contamination. Children in the higher-order repetitive behaviors group exhibited behaviors such as ordering, washing, repeating, and checking. Children in the lower order repetitive behaviors group exhibited self-damaging behaviors and games/superstitious behaviors. Children in the hoarding group exhibited obsessions and compulsions related to hoarding.

Different authors proposed different ways of classifying children with autism based on specific areas of deficit (Table 2.2) using different methodologies. The two group classification system derived by Miles and colleagues<sup>44</sup> is distinct from others as it focuses mainly on morphological abnormalities. The classification derived by Rapin and colleagues<sup>47</sup> is also different from others as they exclusively focused on expressive phonology. The classification systems derived by Cuccaro and colleagues<sup>43</sup> and Lam and

The development of Autism Classification of Functioning: Social Communication (ACSF: SC) was in progress when this thesis was written; therefore not included.

colleagues<sup>46</sup> are relatively similar as they focused on same domain (i.e restricted repetitive behavior) and used same measure (i.e Autism Diagnostic Interview Revised). Anagnostou and colleagues<sup>49</sup> studied repetitive behaviors along with obsessions and compulsions using a different scale and the classification derived is a mixture of the results derived in earlier studies on repetitive behavior with added components of compulsions and obsessions. The sub typing derived by Stevens<sup>42</sup> focused on cognition, communication, and socialization components. Lane and colleagues<sup>48</sup> derived the classification primarily based on sensory domain, whereas the classification system derived by Liss and colleagues<sup>45</sup> is a combination of categorizations proposed by Lane and colleagues<sup>48</sup> and Stevens and colleagues<sup>42</sup> as they focused on sensory, attention, and adaptive behavior components.

Among all classification systems, the one proposed by Stevens and colleagues<sup>42</sup> captures the breadth of dimensions of ASD and the measures used are a combination of *performance* and *capacity*. This classification was published before the publication of the ICF. Perhaps a revision of Steven's classification could be considered by future researchers.

Most of the measures used by different authors to derive different classification systems in ASD map onto either *capacity* or *performance*, except a few. The head circumference measurement and brain MRI map to the *body structure* construct of the ICF. The photoarticulation test and brain EEG map to the *body function* construct of the ICF. Although the Peabody Picture Vocabulary Test (PPVT) measures the *body function*

The development of Autism Classification of Functioning: Social Communication (ACSF: SC) was in progress when this thesis was written; therefore not included.

aspect it is administered in the context of capacity. Interestingly, the term impairment is used in the Wing Autistic Disorder Interview Checklist, although the items specifically address the *performance* of children with autism in their everyday lives. With the publication of the ICF, and its clear description of the distinction among “*impairment*”, “*capacity*” and “*performance*”, specific attention should be given when using these terms.

## Summary of results of the classifications

As discussed in chapters 1 and 2, there are different ways of classifying children with CP, DCD, and ASD. The prevailing classification systems of CP map to *body function/body structure*, *capacity*, and *performance* constructs of the ICF. Children with DCD and ASD are classified using different combinations of multiple measures. The existing subtypes of DCD are derived through cluster analysis. The measures used in different classification systems map to *body function* or *capacity* and none of the measures map to *performance*. Children with autism are classified based on various constructs using various measures and using cluster or factor analysis or simple description based on morphology. The majority of the measures used for classifying children with ASD map to *capacity* or *performance*. Given the variety of ways in which children with these selected neurodisabilities are classified, it is useful to investigate factors associated with the utility of various approaches.



## Utility of classification systems of Cerebral Palsy, Developmental Co-ordination Disorder, and Autism Spectrum Disorders

### Cerebral Palsy

Detailed description of the psychometric properties of classification systems in children with cerebral palsy are provided in Table 2.3. The topographical and Swedish classification lack adequate psychometric properties for use in clinical practice at the present time. In contrast, based on the criteria proposed earlier, the SCPE<sup>18, 19</sup>, and the BFMF<sup>19, 24</sup> classification has good reliability and validity. The GMFCS has good reliability,<sup>53</sup> validity<sup>54</sup> and stability over time<sup>55</sup> serving as a standard classification system that can be used to classify children with CP. The MACS is analogous to the GMFCS and also has good reliability,<sup>12</sup> validity,<sup>27</sup> and stability.<sup>29</sup> The CFCS is a relatively new classification for which only preliminary evidence on psychometric properties has been published.<sup>13</sup>

**Table 2.3: Psychometric properties of classification systems in children with cerebral palsy**

<b>Classification systems</b>	<b>Reliability</b>	<b>Validity</b>
Topographical classification <sup>7</sup>	Inter-rater reliability: K = -0.01 to 0.59	
Surveillance of Cerebral Palsy in Europe <sup>8, 18, 19</sup>	Inter-rater reliability: K= 0.85 Inter-rater reliability: K= 0.84	
Swedish classification <sup>9</sup>	-	
Gross Motor Function Classification System <sup>10,20,21,22,53-55</sup>	Inter-rater reliability: ICC = 0.96 Inter-rater reliability for children older than 2 years of age K=0.75 Inter-rater reliability for children under 2 years of age K=0.55	Correlation with Gross Motor Function Measure scores r=-0.91

	Test retest reliability: G=0.79	
Bimanual Fine Motor Function <sup>11, 19, 24</sup>	Inter-rater reliability: K = 0.98	Correlation with Manual Ability Classification System <sup>24</sup>
Manual Ability Classification System <sup>12,26-29</sup>	Inter-rater reliability: ICC = 0.97 Test retest reliability: ICC = 0.97	Correlation with Functional independence measure for children r=-0.78 <sup>27</sup> Correlation with Pediatric Evaluation of Disability Inventory r=-0.72 <sup>28</sup>
Communication Function Classification System <sup>13</sup>	Intra-rater reliability: K = 0.82 Inter-rater reliability: 0.66	

K=Kappa statistics, ICC=Intraclass Correlation Co-efficient, G=Generalisability co-efficient

The clinical utility of the GMFCS has been studied by various groups of researchers<sup>56, 57</sup> and they found that the GMFCS has good international uptake and is widely used in clinical practice and research by various health professionals. Our group conducted a scoping review on the use and dissemination of the MACS<sup>58</sup> and the results of our study found that the MACS is used worldwide in wide variety of research contexts; however, its clinical utility is yet to be described. The clinical utility of the CFCS has not yet been studied. All of these classifications (i.e the GMFCS, the MACS, and the CFCS) can be administered by both parents and health care professionals and enhance communication between health care professionals and family members to assist in decision making in all aspects of rehabilitation.

The topographical classification and classifications based on motor disorders primarily serves the purpose of “examination”. In addition to the use in examination, the GMFCS currently provides evidence of prognostic properties. The Ontario Motor Growth curves for each GMFCS level predicts the functional mobility of children with CP at 12 years of age.<sup>59</sup> For example, a child classified with GMFCS level III at the age of 3 has a high probability of being able to walk with assistive devices indoors and outdoors as they

grow older. This peaks at 7 to 9 years of age and plateaus thereafter when children typically rely on wheeled mobility due to various reasons such as fatigue, personal preference, and consideration of energy expenditure and time. This information can be used to determine realistic goals in intervention planning. For example, with the above described example, rather than focusing on maintaining community ambulation, intervention planning should focus on maintaining health status and selecting appropriate assistive devices when moving around the home, school, and community at large. The recent evidence on stability of the MACS also provides some evidence of prognosis on manual ability of children with CP in addition to the use in examination.<sup>29</sup> For example, it could be predicted that a child categorized as MACS level III who, at the age of 3, requires assistance and preparation including mounting a sheet of paper on a table to do colouring activities may require similar assistance at the age of 18 to perform manual activities including painting. The CFCS, with the available evidence, only serves in identifying a subgroup and does not provide information on prognosis at this time.

With respect to the focus of classification systems on developmental characteristics, the GMFCS has specific age bands and descriptions with a major focus on specific developmental aspects. In contrast, both the MACS and the CFCS are classified in the context of age-appropriate developmental activities. Classifications of limb distribution, type of motor disorder, and the BFMF do not incorporate developmental characteristics. Development and prognosis are linked with each other. A stable classification with specific focus on developmental characteristics has the potential to predict prognosis. Prognosis plays a major role in predicting the course of a health

condition and its impact on developmental patterns of a child, and thus is useful for both realistic goal setting and intervention planning.

All of the classification systems of CP including topographical, motor disorder subtyping, and functional classification systems are easy and quick classifications as they take only minimal time to determine. For example, using the five-level classification systems takes five minutes if the assessor is familiar with the child or may take one half hour of exposure to the child, combined with conversations with a parent, to establish a level. All of these classification systems are non-commercial and complete descriptions are available free online making these classifications accessible and feasible.

## Developmental Co-ordination Disorder

The subgroups of DCD are determined by a combination of different measures and or assessment batteries. Synthesized findings of the psychometric properties of the measures used in classifying children with developmental co-ordination disorder are provided in Table 2.4 and detailed descriptions of the psychometric properties of the same measures are provided in Appendix 2.2. As the optimal indicator for reliability, the ICC was used in only a few measures and the values differ with various measures and or batteries. With the proposed criteria for determining reliability, some measures (i.e approximately two thirds) have good reliability and reliability of some measures (i.e one third) ranges from moderate to good. Reliability of other measures was explored using reliability co-efficients which could be either the Pearson's or Spearman's correlation coefficient, which inflates values in the presence of systematic differences within and between raters and overtime. In terms of validity, the correlation coefficient of tests

differs based on the tests/versions of tests with which they are correlated and the results present with a mixed picture, with only about 14% having good or strong evidence.

DCD is primarily diagnosed using a combination of the DSM and the ICD. The classification systems derived by the researchers in the identified studies contribute to the "examination" aspect. A citation search for utility of the classification systems on the identified studies did not provide information on usefulness in other elements of rehabilitation practice. Specifically, no information of the stability of systems has been reported. Green et al. reported the limited predictive validity of the classification system developed by their team.<sup>40</sup>

**Table 2.4: Summary of reliability and validity of measures used in classification of children with developmental coordination disorder**

Findings	Measures	
	Reliability	Validity
Good/Strong	<ul style="list-style-type: none"> <li>• Test re-test reliability of KAT</li> <li>• Test re-test and inter-rater reliability of MVPT</li> <li>• Inter-rater reliability of MABC</li> <li>• Test re-test and inter-rater reliability of COMPS</li> <li>• Inter-rater and Intrarater reliability of Rey-Osterreith Complex Figure Test</li> <li>• Test re-test reliability of Bell Crossing test</li> <li>• Test re-test and inter-rater reliability of DTVP</li> <li>• Test re-test reliability of Visually Evoked Potentials</li> </ul>	<ul style="list-style-type: none"> <li>• Correlations between BOT-2 and BOTMP total composite score</li> <li>• Correlation between MABC and PDMS</li> <li>• Correlation between WISC-IV and Perceptual Reasoning Index</li> </ul>
Moderate	<ul style="list-style-type: none"> <li>• Inter-rater reliability of DT-VMI</li> <li>• Test re-test and inter-rater reliability of BOTMP</li> <li>• Test re-test reliability of MABC</li> <li>• Test re-test reliability of TLT</li> </ul>	<ul style="list-style-type: none"> <li>• Correlation between the MVPT and the Spatial awareness subscale of the Rivermead Perceptual Assessment Battery</li> <li>• Correlation between Beery VMI and Comprehensive test of Basic Skills</li> <li>• Correlation between berry VMI and wide range assessment of Visual Motor Abilities</li> <li>• Correlation between BOT-2 and PDMS-2</li> <li>• Correlation between MABC and BOTMP</li> <li>• Correlation between total COMPS score and BOTMP Battery composite</li> </ul>

		<ul style="list-style-type: none"> <li>• Correlation between Binet IQ and Block design</li> <li>• Correlation between Catherine Bergego Scale and bells test</li> <li>• Correlation between the Porteus Vineland series and the Porteus Extension Series</li> <li>• Correlations between the TLT, and WAIS-R Digit Span</li> <li>• Correlations between the TLT, and Raven progressive matrices</li> <li>• Correlations between the TLT, and Test of Divided Attention</li> <li>• Correlation co-efficient with Lincoln-Oseretsky Motor Development Scale</li> </ul>
Weak/poor		<ul style="list-style-type: none"> <li>• Correlations between the MVPT-3 and the DTVP - 2</li> <li>• Correlation between the MVPT-3 and the DTVP</li> <li>• Correlation between Beery VMI and Bender-Gestalt</li> <li>• Correlation between MABC and Berry-VMI</li> <li>• Correlation between DTVP-2 and WISC-R</li> </ul>

KAT = Kinaesthetic Acuity Test, MVPT = Motor Free Visual-Perception Test, MABC = Movement Assessment Battery for Children, COMPS = The Clinical Observations of Motor and Postural Skills, DTVP = Developmental Test of Visual Perception, DT-VMI = Developmental Test of Visual Motor Integration, BOTMP = Bruininks Oseretsky Test of Motor Proficiency, TLT = Tower of London Test, WISC = Wechsler Intelligence Scale for Children, WAIS – R = Wechsler Adult Intelligence Scale Revised. IQ = Intelligence Quotient.

With regard to the emphasis on development, the Movement Assessment Battery for Children consists of specific age bands; the majority of the other measures and test scores are compared with the age equivalent scores except the visual evoked potential and bell crossing test in which the interpretation of the score is made within the context of other clinical findings and tests. For the most part, the criterion of development seems to be considered in classifying DCD.

The identified studies have used multiple measures/assessment batteries followed by cluster analysis. Also the majority of the measures used are commercial products and the duration required to administer the particular test/measure differs among the test/measures. I believe these measures require training to administer and score and it takes considerable time to determine a subgroup. Classification of children with DCD is determined using sophisticated analysis and a clear estimation of time required to administer a measure/assessment battery and perform the sophisticated analysis to determine the classification system was not provided in the identified studies, but can be concluded to be lengthy.

## Autism Spectrum Disorder

Different ways of classifying children with ASD has been provided by many researchers. Appendix 2.3 provides a detailed summary of the psychometric properties of the measures and a synthesized summary of psychometric properties of the measures used in classifying children with ASD are provided in Table 2.5. Reliability of the measures used by various researchers in deriving classifications for children with ASD was explored using three types of statistical techniques: the ICC, a reliability co-efficient



**Table 2.5: Reliability and validity of measures used in classification of children with autism spectrum disorder**

Findings	Measures	
	Reliability	Validity
Good/Strong	<ul style="list-style-type: none"> <li>•Reliability of the Stanford-Binet Intelligence Scale</li> <li>•Test-retest coefficients of PPVT</li> <li>•Test retest reliability co-efficient of VABS (survey form)</li> <li>•Test retest reliability co-efficient of VABS (expanded form)</li> <li>•Inter-rater reliability of ADI-R</li> <li>•Interrater reliability co-efficient of head circumference</li> <li>•Reliability of multicentre MRI</li> <li>•Test-retest reliability of EEG</li> <li>•Inter-rater and test re-test reliability of DSM-IV checklist</li> <li>•Inter-rater reliability of the yale-brown obsessive compulsive scale</li> </ul>	<ul style="list-style-type: none"> <li>•Correlation between the Stanford-Binet Intelligence Scale and Leiter International performance scale</li> <li>•Correlation between the PPVT-3 and the Wechsler Intelligence Scale for Children - 3</li> <li>•Correlation between the PPVT-3 and the Kaufman Adolescent and Adult Intelligence Test</li> <li>•Correlation between revised and original Vineland</li> <li>•Correlation between fetal brain volume and head circumference</li> <li>•Correlation between CELF-4 and CELF-3</li> <li>•Test-retest reliability co-efficient of Leiter international performance scale</li> <li>•Correlation between Leiter international performance scale and Stanford-Binet</li> <li>•Correlation between WISC-IV and Perceptual Reasoning Index</li> </ul>
Moderate	<ul style="list-style-type: none"> <li>•Reliability of Bayley Scales of Infant Developmental</li> <li>•Inter-rater reliability co-efficient of VABS (survey form)</li> <li>•Test-retest reliability of structural brain networks from diffusion MRI</li> <li>•Test re-test reliability of the yale-brown obsessive compulsive scale</li> </ul>	<ul style="list-style-type: none"> <li>•Correlations between Bayley and Griffiths scales</li> <li>•Correlation between the PPVT-3 and the Kaufman Brief Intelligence test</li> <li>•Correlation between the Adaptive Behavior Composite and the original Vineland unadjusted Social Quotient Correlation between the Adaptive Behavior Composite and Silverstein’s Deviation Social Quotient</li> <li>•Correlation between VABS and the Adaptive Behavior Inventory for Children</li> <li>•Correlation between Leiter international performance scale and WISC-R</li> </ul>
Weak/Poor	<ul style="list-style-type: none"> <li>•Inter-rater reliability of brain MRI</li> </ul>	<ul style="list-style-type: none"> <li>•Correlation between VABS and PPVT-R</li> <li>•Correlation between DTVP-2 and WISC-R</li> <li>•Correlation with Behavioral Avoidance Test</li> <li>•Correlation with Mandsley Obsessional Compulsive Inventory</li> </ul>

PPVT = Peabody Picture Vocabulary Test, VABS = Vineland Adaptive Behavior Scales, ADI-R= Autism Diagnostic Interview Revised, MRI = Magnetic Resonance Imaging, EEG= Electroencephalogram, CELF = Clinical Evaluation of language fundamentals, WISC = Wechsler Intelligence scale for children, DSM = Diagnostic Statistical Manual of Mental Disorders. DTVP - Developmental test of visual perception

(either Pearson's  $r$  or Spearman's  $\rho$ ), or the Kappa statistic. Similar to the situation in DCD, the findings show that two thirds of the measures have good or strong reliability. Based on the proposed criteria, validity of different tests/measures differs depending on the versions and the tests with which they are correlated. Results were mixed, with only 50% of the measures reporting good or strong validity.

All of the classifications serve the purpose of "examination" and few classification systems have prognostic implications. No information on the stability of classifications has been reported.

In terms of attention to the criterion of development, the measures used in the classification of ASD do not contain specific age groups; rather, scores of the majority of measures are converted into standardized scores or percentiles and compared with available normative data. In contrast, interpretation of scores of the Autism Diagnostic Interview Revised is done in comparison with DSM-IV and ICD-10 and interpretation of MRI and EEG are done in relation to the clinical findings, therefore these later methods do not incorporate developmental considerations.

Children with ASD are classified using sophisticated techniques such as cluster analysis or factor analysis on items of a particular measure or among different measures. The majority of the measures are commercial products and require training to administer and score. They also likely take substantial time to determine the subgroup.

## Summary of utility of the classifications

In summary, utility of the classifications of CP, DCD, and ASD differs based on the proposed criteria. A synthesized picture of the characteristics addressed by the existing classifications of the three selected neurodisabilities is provided in Table 2.6.

**Table 2.6: Characteristics of existing classifications of three selected neurodisabilities**

Criteria for optimal classification	Components of individual criterion	Classifications mapping to ICF components	CP	DCD	ASD
Mapping to the ICF	Body structure/function		√	√	√
	Capacity		√	√	√
	Performance		√	X	√
Psychometric properties	Reliable	Body structure/function	±	±	±
		Capacity	√	±	±
		Performance	√	X	±
	Valid	Body structure/function	±	±	±
		Capacity	√	±	√
		Performance	√	X	±
	Stable	Body structure/function	X	X	X
		Capacity	X	X	X
		Performance	√	X	X
Purpose	Examination		√	√	√
	Prognosis		√	X	√
Development		Body structure/function	X	±	±
		Capacity	X	±	√
		Performance	√	X	±
Feasibility			√	X	X

√ – Present, X – Absent, ± – Partially present, ASD - Autism spectrum disorder, CP – Cerebral Palsy, DCD – Developmental co-ordination disorder, ICF – International Classification of Functioning, Disability And Health

Of all the classifications used for classifying children with CP, the GMFCS and the MACS are standard, reliable, valid, stable, and feasible classifications that serve multiple purposes. A unique feature of the GMFCS and the MACS is that they are stable classifications and therefore helpful in predicting the prognosis of a child with CP, as he or she develops. Interestingly, the topographical and the SCPE classifications are also

used widely although they lack psychometric properties and serve only a limited purpose of examination. Classification systems in DCD and ASD demonstrate variable psychometric properties, require many measures/assessment batteries and sophisticated techniques, serve limited purposes (classification systems in DCD serve only “examination” whereas classification systems in ASD serve “examination” and “prognosis”), have limited attention to the criterion of development, and therefore the utility of these classification systems have not yet been fully elucidated.

## Recommendations

### Characteristics of optimal classification systems

This work on perspectives of classification of three neurodisabilities resulting in variable clinical utility caused me to propose more questions than answers in recommending classification systems: Which ICF constructs lend themselves best to useful classifications? How important is it for a classification system to be useful for multiple purposes and to cover aspects of development? Is there a classification that could be considered ideal and serve as a template to be followed or applied in other health conditions?

Ideally the answers to the questions above would be in the public domain; however, with the present state of knowledge I conclude that the ICF could be considered as a standard classification but at this point I cannot determine which of the three constructs (i.e body structure/function, capacity, and performance) are important to consider and whether the ICF is an optimal classification on its own. At this time, the ICF

doesn't provide any prognostic implications; however, future studies on natural history may shed light on functional prognosis.<sup>60</sup>

From my perspective, a classification should address the key features of a health condition. In addition to having the characteristics of strong reliability and validity, attention to development, timely completion, and being readily available as well as the key features of the condition should also be considered. Greater emphasis should be placed on the multiple purposes of classification in rehabilitation practice (i.e system should go beyond the examination element and include prognosis and intervention planning). At minimum, a stable classification has the potential to determine the prognosis of a disorder which helps in effective and efficient intervention planning.

As stated above, inclusion of the key features of conditions are one of the important characteristics that a classification should possess. In this section, the ability of available classifications to address the features of the corresponding health conditions are discussed.

CP is a disorder of movement and posture that occurs due to a non-progressive defect or a lesion in the developing brain. Children with CP often have comorbidities including disturbances in sensation, perception, cognition, communication, behaviour, epilepsy, and secondary musculoskeletal disorders.<sup>4</sup> Of the various features listed in the definition, the identified classifications subgroup children with CP based specifically on any one feature (for example, motor disorders, or distribution of involvement, or gross motor function, or manual ability, or communication). Notably, none of the systems

incorporate postural control or disturbances of sensation, perception, cognition or behavior or epilepsy or secondary impairments.

The key feature of DCD is a disturbance in development of co-ordination to the extent that it affects the academic performance. The prevailing classifications address the key feature of DCD in terms of coordination; however, the fact that there are no measures that capture performance in the academic setting precludes establishment of inclusion of all key features of the diagnosis.

The key features of the ASD according to the DSM V are disturbance in social communication and/or have repetitive behaviour. Of the existing classifications of ASD, the one proposed by Stevens and colleagues<sup>42</sup> addresses both the components of DSM V with the remaining classifications addressing only one of the two components of the DSM V criteria.

Based on the findings, it is clear that none of the classification systems meet all of the criteria for clinical utility. Specifically, none of the classifications addressed all of the key features and all classifications presented with variable psychometric properties. With regard to the purpose, classification systems in CP and ASD served more than examination and shed light on prognosis of the condition. There is considerable variability of classifications addressing the developmental aspects and at this time only classification systems of CP are feasible to administer.

Classifications that address the key features of a health condition, incorporate key constructs of the ICF relating to the outcomes of interest, have sound psychometric

properties, focus on development, include key elements of rehabilitation practice, as well as being feasible to administer are required for classifying children with the three selected neurodisabilities. The underlying framework used to classify a disorder plays a major role for developing a comprehensive rehabilitation program. At this point, I conclude that the heterogeneity associated with the selected neurodisabilities pose major challenges.

Although further work on classification is warranted for all neurodisability groups, the focus of this thesis is on CP. I believe that this approach to classification might be useful in future in planning rehabilitation services to people with complex, chronic, and heterogeneous conditions across the life span.

Classification systems in DCD and ASD were developed using cluster/factor analysis. Factor analysis is primarily used to identify groups of variables and cluster analysis is used to subgroup people or objects or data. Therefore cluster analysis is more relevant in identifying subgroups. In addition, I was interested in exploring a simple additive model, which is more clinically feasible than cluster analysis. In this thesis, relative weighting of various measures was not considered, beyond the scaling offered by individual measures, as used in the model testing of the Move & PLAY study.<sup>61, 62</sup> The comparability of results using a simple additive model compared to a more sophisticated cluster analysis has not yet been investigated. In addition to identifying subgroups of interest, many researchers also analyzed the relationship between the subgroups and specific outcomes.<sup>40, 42, 44, 48</sup>

Based on the findings of this preliminary study of DCD and ASD, and considering the heterogeneity and the complexity of the features in children with CP, the

primary purpose of this thesis is to present the results of two separate studies to explore different methods i.e summing technique (which is simple to create and clinically easy to replicate) and cluster analysis (which is analytically complex and difficult to apply clinically) of developing a holistic classification system in children with CP using a variety of measures across the ICF and investigating the association between the derived classifications and classification based on the participation-level child factors using Gross Motor Function Classification System and their respective associations with magnitude of change in motor function over a one-year period among children aged 18 months to 5 years.



## Chapter 3: Development of holistic classifications for children with cerebral palsy

### Introduction

According to the international consensus definition,<sup>4</sup> children with cerebral palsy (CP) present with multiple features; however, none of the prevailing classification systems classify children with CP holistically. The importance of describing and classifying as a whole and/or from a broader perspective could be further explained by a poem written by John Godfrey Saxe: Blind men and the Elephant, where 6 blind men were asked to describe the elephant by only touching one part of the elephant. One blind man touched only the leg and said the elephant is like a tree trunk, one touched only the tail and said the elephant is like a rope, one touched the ear and said the elephant is flat like a fan, one touched the tusk and said the elephant is sharp like a spear, and one touched the trunk and said the elephant is like a snake.<sup>63</sup> All the descriptions should be put together to avoid misinterpretations. This recommendation also applies to CP.

The prevailing classification systems including the Gross Motor Function Classification System (GMFCS),<sup>10</sup> the Manual Ability Classification System (MACS),<sup>12</sup> the Communication Function Classification System (CFCS),<sup>13</sup> and so on, are all excellent classification systems which have good reliability and validity. All of these classification systems are specifically designed to categorize respective areas of functioning in children with CP. Researchers have also attempted to relate various areas of functioning to one

another. All of these classification systems were developed through specific methodological steps.

The results of the study on the classification systems in DCD and ASD,<sup>64</sup> described in the previous chapter, demonstrated that cluster analysis is used in subgrouping. Therefore, in this thesis, children with CP were sub grouped using cluster analysis on multiple measures addressing the majority of the key features of CP. Children with CP present with complex features and subgrouping children with CP using cluster analysis may be an alternative and a clinically useful method. In addition, it is also of interest to explore the possibility of subgrouping children with CP using a simple summing technique of multiple measures.

The main focus of this chapter is to develop two versions of a more holistic classification system called the “Body Function Index in Cerebral Palsy – versions I & II” using 6 assessments discussed in detail in this chapter using two different methods i.e. a summing technique and cluster analysis. The new indices are named as body function index because they describe the neuro-musculo skeletal status and the extent of its influence on children’s body function.

The BFI-CP is a condition-specific index designed to measure the body function status of children with CP. The definition of functional index used to construct the BFI-CP is “neuro-musculoskeletal status and associated co-morbid health conditions, comprising the extent of the influence on a child’s body function in children with CP”. Associated co-morbid health conditions (framed in the context of function<sup>65,66</sup>) were

included as these comprise key features of children with CP. Comparison of the CSI and BFI-CP is provided in Table 3.1.

The two versions of the BFI-CP were informed by the Comprehensive Severity Index (CSI) developed by Dr. Susan Horn and her colleagues in “Practice-Based Evidence”.<sup>67</sup> The CSI embraces the traditional medical model (i.e International Classification of Diseases - ICD)<sup>5</sup> and uses disease-specific physiologic data. The CSI may be useful for analyzing length of hospital stay and severity of illness to achieve specific medical outcomes from a biomedical perspective; however, from a biopsychosocial perspective, assessment of disease-specific physiologic measures provides only limited information. Functional state, potentially including body function indices as outlined in the ICF, might provide a useful framework for classification to assist in developing specific goals and planning rehabilitation strategies.

**Table 3.1: Similarities & differences between the Comprehensive Severity Index and Body Function Index in Cerebral Palsy**

<b>Comprehensive Severity Index<sup>67</sup></b>	<b>Body Function Index in Cerebral Palsy</b>
Disease specific: informed by the ICD	Condition specific: informed by the ICF
Measures 'Severity' (Negative focus)	Measures 'Function' (Positive focus)
Stratifies patients	Stratifies children with cerebral palsy
Severity levels are determined based on physiological signs and symptoms	Functional levels are determined based on neuro-musculoskeletal functions and functional manifestation of associated co-morbid health conditions
Criteria set is developed by expert clinician panels for each ICD-9 CM codes	Constructs (primary impairment, secondary impairment, and associated health condition) are developed from literature survey, consensus with physical therapists and input from parents <sup>65,66</sup>
Criteria include laboratory measures	Constructs includes clinical measures

ICD – International Classification of Diseases, ICF – International Classification of Functioning, Disability and Health

The BFI-CP version I was developed using a simple summing technique on a range of clinical scores and dividing the total score into quintiles based on percentile ranking. The BFI-CP II was developed using cluster analysis. This study is an initial investigation into determining which of the two new indices is superior. Therefore the purpose of this study is (i) to thoroughly describe each of the new indices, (ii) to investigate relationships of each of the new indices to the Gross Motor Function Classification System (GMFCS) (acknowledged international gold standard), as well as to each other (providing evidence for construct validity) and (iii) to explore the extent to which the classifications differ from the GMFCS (further exploring the construct validity) and understanding in greater detail how different they are. Therefore the specific objectives of study are:

## Objectives

Objective 1: To develop the Body Function Index in Cerebral Palsy Version I (BFI-CP I), using indicators such as spasticity, balance, and distribution of involvement (i.e. primary impairments), limitation of range of motion, strength, and endurance (i.e. secondary impairments), as well as associated co-morbid conditions, by summing the values of the measures of these indicators and dividing them into quintiles.

Objective 2: To develop the Body Function Index in Cerebral Palsy Version II (BFI-CP II), by conducting a cluster analysis on the measures of the following indicators: spasticity, balance, distribution of involvement, limitations of range of motion, strength, endurance, and associated co-morbid health conditions.

Objective 3: To explore how the Body Function Index in Cerebral Palsy Versions I and II relate to the Gross Motor Function Classification System (GMFCS) and to each other and to explore differences among them.

Objective 4: To explore the differences among subsets of classifications of the BFI-CP I that do not correspond to the ordinal levels of the GMFCS in body structure and function measures (i.e primary and secondary impairments) and health conditions of those with higher levels of quintiles than the corresponding GMFCS levels and lower levels of quintiles than the corresponding GMFCS levels.

Objective 5: To explore the differences among subsets of the BFI-CP II classifications that do not correspond to the ordinal levels of the GMFCS in body structure and function measures (i.e primary and secondary impairments) and health conditions of those with higher levels of clusters than the corresponding GMFCS levels and lower levels of clusters than the corresponding GMFCS levels.

## Methods

### Design

This study is a secondary analysis for which data were extracted from an existing database from the Canadian Institutes of Health Research (CIHR)- funded Move & PLAY study (MOP 81107). Permission to use the existing database from the Move & PLAY study was obtained from the investigators.

## Participants

The original Move & PLAY study database had 430 children. Data from 25 children were excluded due to various reasons summarized in Appendix 3.1. Data from 405 children with cerebral palsy (CP) between the ages 18 months and 5 years who were enrolled in the Move & PLAY study were used for this study. The data were collected between July 2007 and March 2010 three times over a period of one year. Only data collected at time 1 were used in this study. The Move & PLAY study was approved by the Health Sciences Research Ethics Board at Western University and 20 other agencies (all participating sites) prior to data being collected. A signed informed consent was obtained from parents of each participant before initiating data collection. Detailed descriptions of the child and parent demographics are provided in Table 3.2.

## Measures

The following tests were used in the Move & PLAY study to measure the indicators of primary impairments (spasticity, balance, and distribution of involvement), secondary impairments (limitation of range of motion, muscle strength, and endurance), and associated co-morbid health conditions. These indicators were identified as the potential determinants of change in basic motor abilities of young children with CP through development of a theory and evidence-based conceptual model<sup>65</sup> and subsequent consensus process with physical therapists in the province of Ontario.<sup>66</sup> The model was subsequently refined,<sup>68</sup> leading to refinements of the measurement model.<sup>61</sup>

**Table 3.2: Demographics**

<b>Child characteristics</b>		<b>Mean (SD) (N=405)</b>
Age in months		38±11
		<b>Frequency (Proportion)</b>
Child's gender	Boys	230 (57)
	Girls	175 (43)
Child's race	African American or Black (not of Hispanic origin)	29 (7)
	Asian or Pacific Islander	17 (4)
	Hispanic/Latino	17 (4)
	Native American/North American Indian/Metis/Inuit	11 (3)
	White (not of Hispanic origin)	284 (70)
	Bi-racial & Others	47 (12)
Distribution of involvement	Monoplegia	9 (2)
	Hemiplegia	90 (22)
	Diplegia	95 (24)
	Triplegia	24 (6)
	Quadriplegia	187 (46)
GMFCS level	GMFCS I	136 (34)
	GMFCS II	48 (12)
	GMFCS III	51 (12)
	GMFCS IV	75 (18)
	GMFCS V	95 (24)
<b>Parent characteristics</b>		<b>Frequency (Proportion)</b>
Relationship with the child	Mother	351(87)
	Father	20 (5)
	Other	34 (8)
Parental education	Less than high school	10 (3)
	High school or GED	120 (30)
	Community college diploma; Technical degree/ Associates degree	107(26)
	Bachelors degree	94 (23)
	Masters degree	62 (15)
	Doctoral degree	12 (3)
Household income (N=391)	less than \$15,000	38 (10)
	\$15,000 - \$29,999	45 (12)
	\$30,000 - \$44,999	53 (13)
	\$45,000 - \$59,999	57 (14)
	\$60,000 - \$74,999	47 (12)
	\$75,000 or more	151 (39)

GMFCS = Gross Motor Function Classification System, GED = General equivalency diploma

Table 3.3 contains the details of the psychometric properties of the measures of these indicators<sup>62</sup> as well as the International Classification of Functioning, Disability and Health (ICF) constructs. Time to complete each measure is also provided in this table as an estimate of feasibility. All measures, except endurance and health conditions were collected by trained therapists. The scores in a few measures were rescaled and/or recoded such that higher scores indicate better performance for use in the development of “Body Function Index in Cerebral Palsy – Version I”. The rescaled scores were used in the development of “Body Function Index in Cerebral Palsy – Version II” and scores were not recoded. Both rescaling and recoding of each measure are described in the next section.

The Modified Ashworth Scale (MAS)<sup>69,70</sup> was used to measure spasticity. Bilateral hamstrings and elbow flexor muscles were tested by performing three repetitions. The first recorded score for each of four items were used. Ratings on resistance were done using a 6-point scale, the scores of which were rescaled from 0 to 5, where 0 denotes “no increase in tone” and 5 denotes “rigid”. The scores were recoded such that 5 indicates “no increase in tone” and 0 indicates “rigid”. The average of these four items was used.



**Table 3.3 – Psychometric properties of the measures ( Adapted with permission from Bartlett DJ, Chiarello LA, McCoy SW, et al. Determinants of gross motor function of young children with cerebral palsy: A prospective cohort study. Dev Med Child Neurol. 2014; 56: 275-282. Copyright © 2014. John Wiley And Sons)**

Key aspects of CP (Indicators)	Measure	ICF construct	Reliability	Validity	Time to Complete
Spasticity	Modified Ashworth Scale <sup>69</sup>	Body function	Inter-rater reliability : ICC = 0.79	Convergent validity with Tardieu scale, myotonometer and isokinetic dynamometer <sup>70</sup>	5 minutes
Balance	Early Clinical Assessment of Balance <sup>71</sup>	Capacity	Inter-rater: ICC = 0.99 Test-retest: ICC = 0.99	Known groups validity: - Significantly different among all GMFCS levels - Younger children lower scores than older children Convergent validity: r = 0.95 with the GMFM	10 – 15 minutes
Distribution of involvement	Monoplegia, Hemiplegia, Diplegia, Triplegia, Quadriplegia <sup>7, 14</sup>	Body function	-	-	5 minutes
Muscle Strength	Functional Strength Assessment (Neck, trunk, shoulders, lower extremity major muscle groups) <sup>72</sup>	Capacity	Test-retest reliability: ICC = 0.97 Cronbach's alpha* = 0.93	Known groups validity: - Significantly different among all GMFCS levels except II and III	10 minutes
Range of Motion	Spinal Alignment and Range of Motion Measure <sup>73</sup>	Body structure	Inter-rater and test-retest reliabilities (ICC): > 0.80 Cronbach's alpha = 0.95	Known groups validity: - Significantly different among all GMFCS levels	15 minutes
Endurance	Early Activity Scale for Endurance <sup>74</sup>	Performance	Test-retest reliability: ICC = 0.95 Cronbach's alpha = 0.83	Known groups validity: - Significantly different among all GMFCS levels but II and III and III and IV Construct validity – Spearman's rho = 0.52 with 6 Minute Walk Test	5 minutes
Associated co-morbidity	Health Conditions Questionnaire <sup>75</sup>	Body function	Test-retest reliability: ICC = 0.85	Known groups validity - Significantly different among all GMFCS levels Content validity: designed from international definition of cerebral palsy	5 minutes

CP = Cerebral Palsy; GMFCS = Gross Motor Function Classification System; GMFM = Gross Motor Function Measure; ICF = International Classification of Functioning, Disability and Health, ICC= Intraclass correlation co-efficient.

A newly developed measure, the Early Clinical Assessment of Balance (ECAB),<sup>71</sup> was used to measure balance. It was developed based on a combination of two measures: the Movement Assessment of Infants – Automatic Reactions section (MAI-AR),<sup>76</sup> and the Pediatric Balance Scale (PBS).<sup>77</sup> Accordingly, the ECAB consists of two parts: Part I (adapted from MAI-AR) and Part II (adapted from the PBS). Part I includes all MAI items except forward protective extension and was measured on a four-point scale rescaled to 0 to 3. Part II includes 6 items from the PBS. The items were first scored on a 5-point scale and were rescaled based on weighting for difficulty. Higher scores indicate better performance. The total score (0-100) was rescaled to 0-10. The rescaled score was used for analysis in the development of both BFI-CP I and II.

Distribution of involvement<sup>7, 14</sup> was measured using the five-point scale. Ratings were done based on the limb involvement – monoplegia was scored as 1; quadriplegia was scored 5. The scores were used for the development of BFI-CP II. The scores were recoded such that 1 indicates “quadriplegia” and 5 indicates “monoplegia” and the recoded score was used for the development of BFI-CP I.

Muscle strength was assessed for major muscle groups (neck and trunk flexors and extensors, and hip extensors, knee extensors, and shoulder flexors) to obtain an overall estimate. The measure is called the “Functional Strength Assessment” (FSA).<sup>72</sup> Muscle groups were assessed bilaterally on a 5-point ordinal scale (0 to 4) where 0 indicates “no initiation of movement against gravity” and 4 indicates “full available range against gravity and some or strong resistance”. The scores were rescaled to 1 to 5. The

mean of all the items was used to obtain an overall estimate of strength. The mean score was used for analysis in both BFI-CP I & II.

The Spinal Alignment and Range of Motion Measure (SAROMM)<sup>73</sup> was used to measure range of motion. The SAROMM consists of two subscales. The Spinal Alignment subscale consists of four items and the Range of Motion and Extensibility subscale comprises of twenty two items. The items were scored on a 5-point scale (0 to 4) where 0 indicates “normal alignment and range of motion” and 4 indicates “fixed deformity”. Average score across all the items were used in BFI-CP II. Scores were recoded such that 0 indicates “fixed deformity” and 4 indicates “normal alignment and range of motion”. The average of all of the items (equal weighting) was used. The recoded scores were used for BFI-CP I.

The “Early Activity Scale for Endurance” (EASE),<sup>74</sup> a newly developed parent-rated questionnaire, was used to measure endurance. The original 11-item questionnaire was reduced to 4 items (Items - 1,2,3,5). The parent/caregiver rated their child’s perceived level of energy, fatigue, and overall ability to sustain active movement without getting tired on a 5-point scale (1 to 5) where 1 indicates “never” and 5 indicates “always”. The average of four items was considered for analysis in both the versions of the BFI-CP.

Associated health conditions and co-morbidities were measured using the parent-rated scale the “Child Health Conditions Questionnaire”.<sup>75</sup> The health conditions measure consists of two parts: prevalence of conditions and impact of each on daily life. The scale contains 16 items. For each of 16 items, parents were first asked if their child had

problems (eg. problem with seeing, hearing etc.). If the item was answered “NO”, then it was recoded as 0 for impact. If the item was answered “YES”, then the score from impact was used which was scaled on a 7-point scale (1 - 7) where 1 indicates “not at all” and 7 indicates “to a very great extent”. Average impact across all the 16 items was used in the development of BFI-CP II. Scores were recoded such that if the item was answered “NO”, then it was recoded as 7 for impact. If the item was answered “YES”, then the score from impact was recoded on a 7-point scale where 0 indicates “to a very great extent” and 6 indicates “not at all”. “Average impact” (recoded) was calculated for all 16 health conditions and was used for the development of the BFI-CP I.

The detailed description, psychometric properties and the purposes of the GMFCS are discussed in chapters 1 and 2 of this thesis. All of the measures serve the purpose of examination except the GMFCS which serves multiple purposes. Work is in progress in the On Track study to investigate changes over time to monitor development using the ECAB, SAROMM, FSA, EASE, and Child Health Conditions Questionnaire. Many of these measures developed in the Move & PLAY study are available on the *CanChild* website: <http://canchild.ca/en/ourresearch/moveplay.asp> .

## Data collection procedures

All of the children were assessed three times i.e. at the beginning (time 1), middle (6 months later), and at the end (12 months after visit 1) of the Move & PLAY study. Data collected at time 1 were used to develop the two versions of the BFI-CP. All therapist assessors in the Move & PLAY study participated in rater training and criterion

testing to ensure reliable data collection. All assessors achieved more than 80% item agreement for all of the measures that they were responsible for completing. All data at time 1 were collected within one to one and half hours.

## Data analyses

### Development of Body Function Index in Cerebral Palsy: Background

The rescaled/recoded scores of all the measures as described earlier (MAS, ECAB, distribution of involvement, FSA, SAROMM, EASE and Child Health Conditions Questionnaire) were summed. The summed values were rank ordered and divided into percentiles to develop the BFI-CP I (Objective 1). The range of summed scores between 100<sup>th</sup> and 80<sup>th</sup> percentile were grouped as Quintile 1, between <80<sup>th</sup> percentile and 60<sup>th</sup> percentile were grouped as Quintile 2, between <60<sup>th</sup> percentile and 40<sup>th</sup> percentile were grouped as Quintile 3, between <40<sup>th</sup> percentile and 20<sup>th</sup> percentile were grouped as Quintile 4, and <20<sup>th</sup> were grouped as Quintile 5. The BFI-CP II was developed using cluster analysis. As a preliminary step, hierarchical cluster analysis was used to identify the number of clusters. The analysis yielded 2 clusters of children with CP. The prevailing functional classifications are five group classification systems. Therefore, K means cluster analysis using 5 cluster solution was selected to develop BFI-CP II to enable comparison to the GMFCS to be conducted (objective 2). The Spearman's correlation co-efficient was used to explore the relationship between the different versions of the BFI-CP and the GMFCS and to each other (objective 3). Chi-Square test

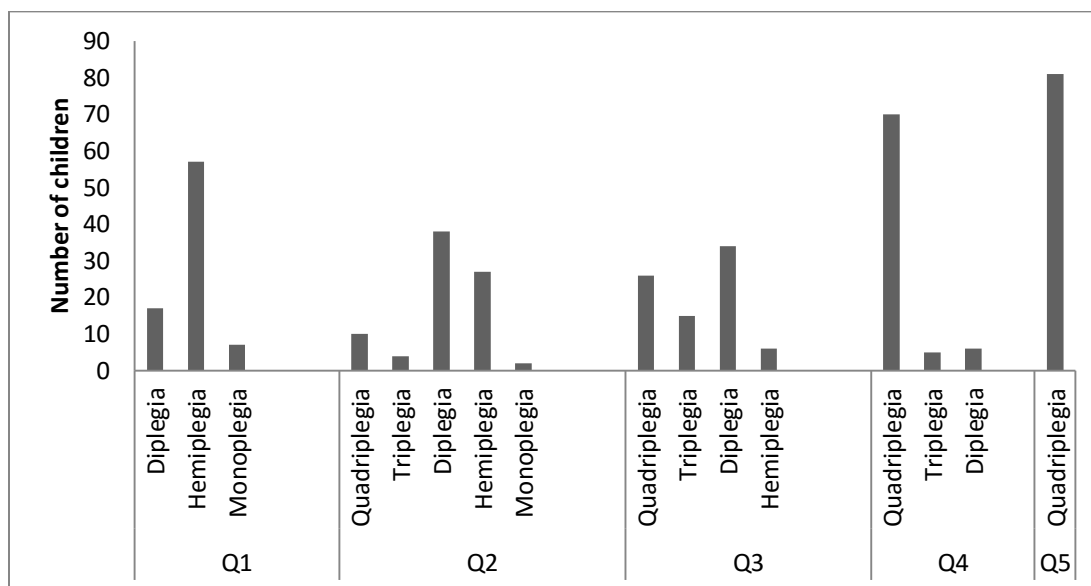
was used to explore the difference between the different versions of the BFI-CP and the GMFCS (objective 3).

The subset of children in BFI-CP I who did not align with the GMFCS in the cross tabulation between the BFI-CP I and the GMFCS were divided into two groups. The group of children above the diagonal were grouped as high quintile and the group of children below the diagonal were grouped as low quintile. Using a similar method, the subset of children in BFI-CP II who did not align with the GMFCS in the cross tabulation between the BFI-CP II and the GMFCS were divided into high cluster and low cluster groups. Mann Whitney U tests were used to explore differences between individual measures of primary and secondary impairments and health conditions between high and low quintile as well as high and low cluster groups (objectives 4 and 5). For distribution of involvement, Chi-Square tests were used to explore differences between the high and low quintile groups. To partially account for the inflated alpha level with multiple comparisons, a more conservative p value of  $<0.01$  was selected for objectives 4 and 5.

## Results

Four hundred and five children with CP were involved in this study. Figure 3.1 contains frequency distributions of limb distribution by quintiles.

Figure 3.1 Frequency of distribution of involvement - Quintiles (Body Function Index in Cerebral Palsy I)



Q - Quintiles

Table 3.4 provides a description on how the unit weighted summed values were divided into quintiles based on percentile ranking for the development of the BFI-CP I. It also provides details on descriptive statistics of individual measures in each quintile of the BFI-CP I. The descriptive statistics for distribution of involvement are not included in the individual quintiles although it is included in the BFI-CP I development. Refer to Appendix 3.2 (Figures 1-6) for boxplots for all variables except distribution of involvement.

Table 3.4: Descriptives for Body Function Index in Cerebral Palsy - I

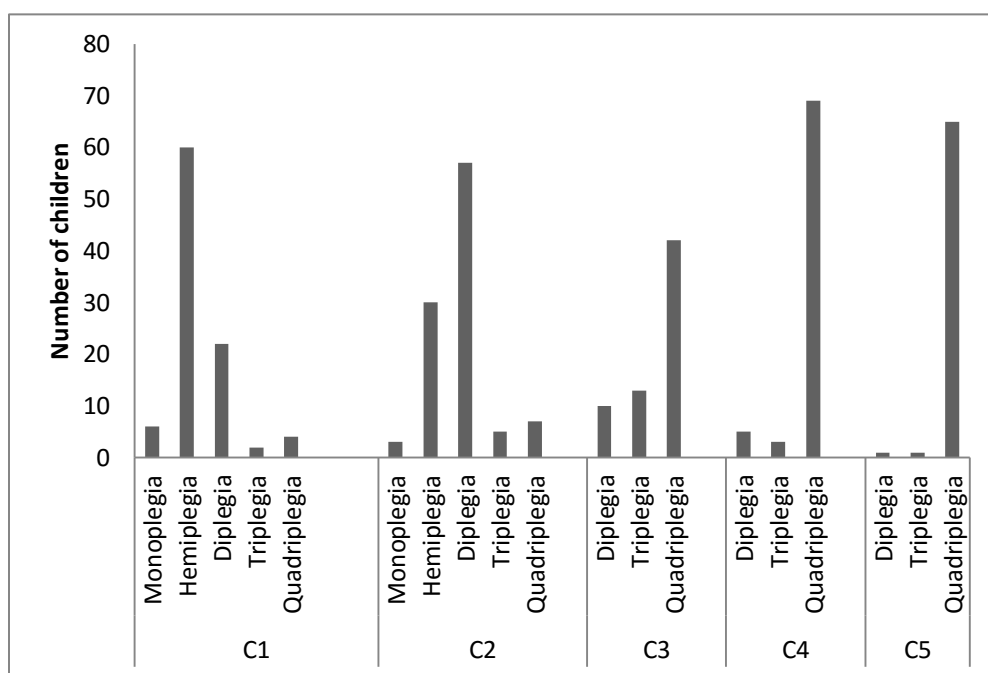
	Range	Variables	Mean	Standard Deviation	Median	Range	Skewness
Q1- (>80 - 100%)	33.87 - 39.69	ECAB	9.1	1.0	9.2	4.65, 10	-1.67
		Spasticity	4.5	0.5	4.5	3.3, 5	-0.66
		Strength	4.6	0.3	4.6	3.8, 5	-0.44
		SAROMM	3.7	0.2	3.8	3, 4	-1.07
		Endurance	4.2	0.6	4.3	2.3, 5	-.94
		Health condition	6.6	0.4	6.8	5.4, 7	-1.24
Q2- (>60 - 80%)	28.17 - 33.86	ECAB	5.7	1.5	5.6	2.7, 9.6	0.57
		Spasticity	4.3	0.6	4.5	3, 5	-.42
		Strength	4.2	0.38	4.1	3.25, 5	0.29
		SAROMM	3.5	0.4	3.6	2.1, 4	-1.20
		Endurance	3.6	0.7	3.5	2, 5	-.12
		Health condition	6.2	0.7	6.3	3.7, 7	-1.27
Q3- (>40 - 60%)	22.93 - 28.16	ECAB	3.6	1.2	3.6	1.2, 7.3	0.20
		Spasticity	3.6	0.8	3.5	1.5, 5	-.16
		Strength	3.7	0.5	3.7	2.3, 5	0.35
		SAROMM	3.2	0.5	3.2	1.8, 4	-.32
		Endurance	3.2	0.9	3.3	1, 5	-.01
		Health condition	6.0	0.8	6.3	2.7, 7	-1.46
Q4- (>20 - 40%)	17.89 - 22.92	ECAB	1.6	0.5	1.6	0.4, 3.3	2.96
		Spasticity	3.0	0.9	3.0	1, 5	0.11
		Strength	3.1	0.8	3.3	1.6, 4.3	0.14
		SAROMM	2.9	0.5	2.8	1.2, 3.8	0.02
		Endurance	2.7	0.8	2.8	1, 5	-.39
		Health condition	5.7	0.8	5.7	3.3, 7	0.01
Q5- (0 - 20%)	≤17.88	ECAB	0.68	0.5	0.6	0, 2.1	0.95
		Spasticity	2.3	0.9	2.3	1, 5	0.65
		Strength	2	0.5	2	1, 3.1	0.01
		SAROMM	2.4	0.5	2.5	0.5, 3.7	-0.97
		Endurance	1.7	0.7	1.5	1, 3.3	0.83
		Health condition	4.9	0.9	4.9	2.4, 6.5	-0.40

Q- Quintiles, ECAB- Early Clinical Assessment of Balance, SAROMM – Spinal Alignment and Range of Motion Measure

Figure 3.2 contains frequency of limb distributions by clusters. Table 3.5 contains the descriptive statistics for BFI-CP II cluster analysis. The descriptive statistics for distribution of involvement is not included in the table for the BFI-CP II; however, it is used in the development of the BFI-CP II. Refer to Appendix 3.3 (figures 1 - 6) for boxplots of all variables except distribution of involvement.



Figure 3.2: Frequency of distribution of involvement – Cluster (Body Function Index in Cerebral Palsy - II



C- Cluster, ECAB- Early Clinical Assessment of Balance, SAROMM – Spinal Alignment and Range of Motion Measure

Table 3.5: Descriptives for Body Function Index in Cerebral Palsy - II

	Variables	Mean	Standard Deviation	Median	Range	Skewness
C1	ECAB	9.0	0.9	9.2	6.7, 10	-0.77
	Spasticity	0.5	0.5	0.5	0, 2	0.72
	Strength	4.5	0.4	4.5	3.5, 5	-0.38
	SAROMM	0.3	0.2	0.2	0, 1.2	1.28
	Endurance	4.1	0.6	4.3	2.3, 5	-0.77
	Health Condition	0.5	0.5	0.3	0, 2.1	1.39
C2	ECAB	5.0	1.0	4.8	2.7, 7.3	0.34
	Spasticity	1.0	0.8	1.0	0, 2.8	0.42
	Strength	4.0	0.5	4.0	2.9, 5	-0.05
	SAROMM	0.6	0.5	0.5	0, 2.2	0.79
	Endurance	3.5	0.8	3.5	1, 5	-0.15
	Health condition	0.8	0.8	0.7	0, 4.3	1.59

C3	ECAB	2.7	0.9	2.7	1.1, 4.9	0.31
	Spasticity	1.1	0.8	1.0	0, 3	0.27
	Strength	3.6	0.6	3.6	2.1, 5	-0.16
	SAROMM	0.7	0.4	0.7	0, 1.73	0.51
	Endurance	3.1	1.0	3.0	1.3, 5	0.19
	Health condition	1.1	0.7	0.9	0, 2.7	0.65
C4	ECAB	1.4	0.5	1.4	0.4, 2.8	0.44
	Spasticity	2.7	0.8	2.8	0.8, 4	-0.04
	Strength	3.0	0.6	3.0	1.4, 4.4	-0.24
	SAROMM	1.3	0.5	1.3	0.4, 2.9	0.62
	Endurance	2.8	0.7	2.8	1, 4.8	0.01
	Health condition	1.2	0.6	1.3	0.1, 2.8	0.09
C5	ECAB	0.6	0.5	0.5	0, 2.4	1.44
	Spasticity	2.4	1.0	2.5	0, 4	-0.45
	Strength	2.0	0.7	2.0	1, 4	0.56
	SAROMM	1.5	0.5	1.5	0.3, 3.5	0.80
	Endurance	1.5	0.5	1.3	1, 3	1
	Health condition	2.3	0.9	2.3	0.5, 4.6	0.15

C - Cluster, ECAB- Early Clinical Assessment of Balance, SAROMM – Spinal Alignment and Range of Motion Measure

Table 3.6 contains the relationship and difference between the BFI-CP I and the GMFCS, BFI-CP II and the GMFCS and BFI-CP I & II.

**Table 3.6: Relationship and difference between Body Function Index in Cerebral Palsy -I and Gross Motor Function Classification System, Body Function Index in Cerebral Palsy -II and Gross Motor Function Classification System and Body Function Index in Cerebral Palsy – I and II**

Relationship and differences		Value	Significance
BFI-CP I and GMFCS	Spearman's rho	0.92 (95% CI - 0.88 - 0.96)	p<0.001
	Pearson Chi-Square	670.489	p<0.001
BFI-CP II and GMFCS	Spearman's rho	0.93 (95% CI - 0.89 - 0.96)	p<0.001
	Pearson Chi-Square	685.574	p<0.001
BFI-CP I and BFI-CP II	Spearman's rho	0.95	p<0.001

BFI-CP – Body Function Index in Cerebral Palsy, GMFCS – Gross Motor Function Classification System

All correlations are greater than or equal to  $r_s = 0.92$  ( $p < 0.001$ ) and all classifications were statistically significantly different ( $p < 0.001$ ).

Tables 3.7 and 3.8 contains the cross tabulations between the GMFCS and BFI-CP I and BFI-CP II, respectively. Tables 3.9 and 3.10 contains the results of the Mann Whitney U test for high quintile and low quintile, and high cluster and low cluster groups, respectively. The Chi-square test for distribution of involvement for high versus low quintile groups is 23.74 ( $p < 0.001$ ) and the chi-square test for distribution of involvement for high versus low cluster groups is 27.46 ( $p < 0.001$ ).

**Table 3.7: Cross tabulation between Body Function Index in Cerebral Palsy -I and Gross Motor Function Classification System**

	Q1	Q2	Q3	Q4	Q5	Total
GMFCS I	80	51	5	0	0	136
GMFCS II	0	23	25	0	0	48
GMFCS III	1	7	35	8	0	51
GMFCS IV	0	0	15	51	9	75
GMFCS V	0	0	1	22	72	95
Total	81	81	81	81	81	405

GMFCS – Gross Motor Function Classification System

Group A (n=98)

Group B (n=46)

**Table 3.8: Cross tabulation between Body Function Index in Cerebral Palsy -II and Gross Motor Function Classification System**

	Cluster1	Cluster 2	Cluster 3	Cluster 4	Cluster 5	Total
GMFCS I	93	42	1	0	0	136
GMFCS II	1	42	5	0	0	48
GMFCS III	0	18	29	4	0	51
GMFCS IV	0	0	26	44	5	75
GMFCS V	0	0	4	29	62	95
Total	94	102	65	77	67	405

GMFCS – Gross Motor Function Classification System

Group C (n=57)

Group D (n=78)

**Table 3.9: Descriptives for all variables for high quintile versus low quintile groups and results of statistical comparisons**

Measures	High quintile (group A)			Low quintile (group B)			p
	Median	Range	Inter quartile range	Median	Range	Inter quartile range	
ECAB	5.15	0.5, 9.6	2.40	1.85	0.4,5.4	1.45	<b>0.001</b>
Spasticity	3.75	1,5	1.75	3.5	1,5	1.56	0.232
Strength	4	1.88, 5	0.78	3.5	1.63, 5	0.91	<b>0.001</b>
SAROMM	3.42	2.08, 4	0.89	3.17	2.19, 4	0.78	0.014
Endurance	3.25	1,5	1.25	3.12	1,5	1.5	0.410
Health conditions	6.12	2.69, 7	1.11	6.15	3.63, 7	1.06	0.424

ECAB- Early Clinical Assessment of Balance, SAROMM – Spinal Alignment and Range of Motion Measure, p- probability value for Mann Whitney U test

**Table 3.10: Descriptives for all variables for high cluster versus low cluster groups and results of statistical comparisons**

Measures	High cluster (group C)			Low cluster (group D)			<i>p</i>
	Median	Range	Inter quartile Range	Median	Range	Inter quartile Range	
ECAB	5.7	0.6, 7.3	2.03	1.8	0.4, 8.3	2.13	<b>0.001</b>
Spasticity	0.75	0,4	1.25	1.5	0, 4	1.81	<b>0.001</b>
Strength	4	2.13, 5	0.63	3.37	1.38, 5	1.13	<b>0.001</b>
SAROMM	0.42	0, 1.73	0.88	0.96	0, 2.92	0.78	<b>0.001</b>
Endurance	3.5	1,5	1.5	3	1, 5	1.5	0.019
Health conditions	0.87	0.06, 4.31	1.16	1.06	0, 2.75	0.95	0.649

ECAB- Early Clinical Assessment of Balance, SAROMM – Spinal Alignment and Range of Motion Measure, *p*- probability value for Mann Whitney U test

Table 3.11 contains a summary of the comparison of the two versions of the BFI-CP.

### 3.11: Summary of comparison between two versions of the Body Function Index in Cerebral Palsy

BFI-CP I	BFI-CP II
Developed using a unit-weighted summing technique	Developed using cluster analysis
Scores of a few measures were rescaled and/or recoded	Scores of a few measures were rescaled but not recoded
Measured on an ordinal level	Measured on an ordinal level
5-level classification	5-level classification
Correlated significantly with GMFCS ( $r_s=0.92$ )	Correlated significantly with GMFCS ( $r_s=0.93$ )
Significant differences between high and low quintile groups in ECAB, strength and distribution of involvement	Significant differences between high and low cluster groups in ECAB, spasticity, strength, distribution of involvement and SAROMM scores.

BFI-CP – Body Function Index in Cerebral Palsy, GMFCS – Gross Motor Function Classification System,  $r_s$  = Spearman's correlation co-efficient, ECAB- Early Clinical Assessment of Balance, SAROMM – Spinal Alignment and Range of Motion Measure

## Discussion

To my knowledge, this is the first study exploring holistic classification systems in children with CP. As described earlier, the GMFCS, the Manual Ability Classification System (MACS), the Surveillance of Cerebral Palsy in Europe (SCPE), the Communication Function Classification System (CFCS), Swedish classification, and distribution of involvement were widely used in literature to subgroup children with CP; however, it is obvious that they do not classify children with CP in all of their complexity. The international definition of CP and the notion of the 'comprehensive severity index' triggered the quest for developing a holistic classification system. Children with CP are extremely heterogenous. Bearing this in mind, in this study multiple measures that examine different features of children with CP were used.

A disadvantage of not considering weighting in the simple summing technique is that the relative influence of various measures is not taken into account. Nonetheless, this is considered to be a moot point, considering the heterogeneity of children with CP. Table 3.4 shows that the mean value of each variable decreases stepwise from Quintile 1 to Quintile 5; however, the median value of spasticity for quintile 1 and 2 as well as the median value for Health condition of quintile 2 and 3 are the same. By definition, the ranges of summed scores of quintiles did not overlap but there is a considerable overlap between ranges of individual variables among quintiles. With regard to skewness, all of the variables in quintile 1 are negatively skewed with variability in the skewness of all other quintiles. However, the variable balance, measured using the ECAB, in Quintile 4 shows a marked skewness compared to others.

The BFI-CP II was developed using cluster analysis which is an appropriate technique for classifying groups of individuals. A 5-cluster solution was selected because the majority of the classification systems in CP had 5 levels and this enabled close examination of correspondence with the GMFCS. The major limitation in this version is that a complex analysis (cluster analysis) was used. Interestingly, the clusters were ranked in order of high to low functional abilities similar to the simple summing technique. Table 3.5, Figure 3.2 and box plots in Appendix 3.3 all show a systematic variation in mean and median values across all the clusters, except for the median value of spasticity in clusters 2 and 3, which are identical. The results for the cluster analysis also showed overlap of value in ranges of individual variables among clusters. Interestingly, the upper-limit (range) of the variable strength is "5" in clusters 1 through 4 and the upper-limit of strength is "4" in cluster 5. The upper-limit (range) of endurance is also up to "5" in clusters 1, 2 and 3 and "4.8" in cluster 4. In addition, the lower limit of the health condition variable is down to "0" in clusters 1, 2, and 3, and only 0.1 in cluster 4 and 0.5 in cluster 5. There is a considerable variability in the upper limit of the health condition variable among the clusters. With regard to skewness, the majority of the variables are within  $\pm 2$  with the highest being "1.59" for the health condition variable in Cluster 2.

Both of the new indices (i.e. BFI-CP I and II) are strongly and significantly correlated with the GMFCS, (with non significant differences in the magnitude of the correlation between indices) each accounting for approximately 85% of the variance in the GMFCS level. In addition, because of the large sample size, statistically significant differences between the two versions of the BFI-CP and the GMFCS were also found.

The BFI-CP versions are entirely different from other classifications (eg. the MACS the CFCS, and the motor disorder subtypes) as the BFI-CP versions represent overall features of children with CP. Based on these results, both versions of the BFI-CP address the majority of the features of CP. Although the GMFCS is quick, easy to administer, and is a standard classification system, the BFI-CP versions are more comprehensive in addressing and classifying overall health and body function status of children with CP. Therefore the BFI-CP versions are complementary to the GMFCS and can be used to describe CP more comprehensively. The results also indicate a strong correlation between both versions of the BFI-CP, which is not entirely surprising as both the versions of the BFI-CP were derived from the same sample using the same measures, albeit different methods.

Regardless of the method used to develop the two new indices, all of the indicators contributed to both versions of the BFI-CP with rank order contributions. Although both the BFI-CP I and II are highly correlated with the GMFCS and with each other, the results of this study also show a statistically significant difference between high quintile and low quintile in three variables (i.e balance, distribution of involvement and strength). The results also show a statistically significant difference between low cluster and high cluster in 5 variables (i.e balance, distribution of involvement, spasticity, strength, and range of motion) summarized in table 3.10. No differences in endurance or impact of health conditions were detected for either group. The results of this study indicate that some children with CP have a great impact of health conditions regardless of the functional level. Wong et al. compared the prevalence and impact of health problems in preschool children with and without CP, stratified by the GMFCS, and found both a



higher prevalence and a significant impact of health problems in children with CP, even those in GMFCS level I compared with typically developing children.<sup>75</sup>

The BFI-CP versions are more comprehensive than the GMFCS. Gross motor function is only one piece of a complex puzzle and therefore classifying children with CP only based on one indicator has a disadvantage of missing the totality of CP. Therefore examining children with CP from a broader perspective helps in establishing a potentially comprehensive classification system. In addition, the BFI-CP versions are constructed using a set of clinically feasible measures rated by both assessors and parents. Therefore the BFI-CP versions are integrated classification systems that incorporate both clinicians' and parents' perspectives. At this point, it is not clear whether the simpler BFI-CP I (with 3 differences from the GMFCS) is preferred over BFI-CP II (with 5 differences from the GMFCS).

Although the results of this study found no to minimal overlap of mean/ median values in both versions of the BFI-CP, the overlap between the ranges of variables in both of the new indices across the 5 respective levels may be attributed to the individual differences among children with CP. Therefore it is important to be mindful of the fact that two children with same mean values may demonstrate different features, again highlighting the fact that children with CP are heterogeneous.

The reliability of the new tools are assumed based on the reliability of the individual measures. As stated earlier, all of the measures used in the development of the two indices are reliable and valid measures.

In summary, both versions of the BFI-CP are 5-level classifications measured on an ordinal scale. The BFI-CP I was developed using a unit weighted summing technique and scores of a few measures were rescaled and/or recoded. The BFI-CP II was developed using a common exploratory data analysis tool (i.e cluster analysis) and the scores of a few measures were rescaled. With regard to comparison between the GMFCS and the new indices, 36% of children in version I and 33% of children in version II are different. This also explains the importance of considering the individual differences in each children.

A limitation of this study is the generalizability of the study's results. Although children with CP from thirteen centres representing urban, suburban, and rural areas across Canada and US were recruited, the majority of the children were white (70%) and 46% of the children had quadriplegia. The majority of the parents were highly educated (41%) and had high socio-economic status (39%). Therefore the results of this study are generalizable only to this sample. A second limitation of this study is the lack of succinct description of each levels of the BFI-CP unlike the GMFCS, MACS and CFCS. Finally, although the MACS was reviewed in Chapter 1, it was not included in both of the new indices as data on manual abilities were not collected in the Move & PLAY study. Similarly although the CFCS was reviewed earlier, it had not been published at the time the Move & PLAY study was planned. This is a constraint of using secondary data.

In conclusion, the BFI-CP versions are an aggregate account of key features of children with CP, measured through multiple domains and/or constructs of the ICF (i.e body function/structure, activity and participation). The measures used in the

development of the new indices can be administered easily and together take less than an hour to administer, therefore could be considered feasible in clinical practice. The strong, but not perfect, correlations between the GMFCS and the two versions of the BFI-CP indicate that they could be used as complementary methods in describing children with CP. The next step in this line of inquiry is to determine the ability of the two versions of the BFI-CP to predict the prognosis of gross motor function.

## Chapter - 4 - Prognostic implications of the holistic classifications in children with cerebral palsy

### Introduction

In Chapter 3, two indices were developed using body function measures. The usefulness of a classification is highly dependent on its prognostic implications, as described in Chapter 2. This next study is an important part in exploring the utility of the holistic classifications developed in Chapter 3 of this thesis.

Prognosis refers to the probable course of a health condition and/or change in function or development over a specific period of time.<sup>78</sup> Understanding and interpreting prognosis of a specific outcome is an important aspect in clinical decision making.<sup>38</sup> Although CP is a non-progressive disorder, the clinical manifestations change over time which increases the complexity associated with understanding and interpreting prognosis. As reviewed in Chapter 2, the prognostic implications of the Gross Motor Function Classification System (GMFCS) and the Manual Ability Classification System (MACS) in predicting gross motor function and manual function in broad brush strokes are well discussed in the literature; however, considering the diversity and the complexity of the features of children with CP, there is a gap in the literature in addressing the prognosis of holistic classification system in children with CP. The main objective of determining prognosis is to enhance clinical decision making in selecting appropriate interventions and/or environmental modifications. Therefore, it is important to determine the prognosis

of holistic classification systems in children with CP in an effort to enhance decision making in this population.

Hanna and colleagues<sup>79</sup> proposed a method to enhance the utility of the GMFCS for understanding and interpreting the meaning of the magnitude of change in gross motor function over time as measured by the Gross Motor Function Measure (GMFM-66).<sup>80</sup> They developed reference percentile curves using 2 time points of data on GMFM-66 scores at a one-year interval from a sample of a previous study by Rosenbaum et al.<sup>59</sup> Reference percentile curves were created for each GMFCS level plotted at the 3<sup>rd</sup>, 5<sup>th</sup>, 10<sup>th</sup>, 25<sup>th</sup>, 50<sup>th</sup>, 75<sup>th</sup>, 90<sup>th</sup>, 95<sup>th</sup>, and 97<sup>th</sup> percentiles. Based on the means and the standard deviations of the changes in percentiles in GMFM-66 by GMFCS levels, they were also able to establish an expected interval of change in percentiles between two assessments corresponding to 20%, 50%, and 80% probabilities. For the purpose of this study, only 50% probability values were used because this permitted a greater proportion of children to be in the categories of developing ‘better than expected’ and ‘more poorly than expected’ than if the 80% probability values were used. It also ensured that 50% of children were developing ‘as expected’ rather than 20% or 80% if those values had been selected. Table 4.1 describes the expected interval of change in percentiles between repeat assessments corresponding to 50% probability over a period of one year.<sup>79</sup>

**Table 4.1: Expected interval of change in percentiles between assessments over a one year interval<sup>79</sup>**

Probability	GMFCS I	GMFCS II	GMFCS III	GMFCS IV	GMFCS V
50%	±10.5	± 10.5	±8.4	±8.0	±8.9

GMFCS – Gross Motor Function Classification System

Children with change in the percentiles between assessments within the expected interval of one year described in Table 4.1 are considered “developing as expected”.

Children with time 2 percentile ranks greater than 10.5, 10.5, 8.4, 8, and 8.9 points above the time 1 percentiles for GMFCS levels I through V respectively are interpreted as “children developing better than expected”. Conversely, children with time 2 percentile ranks less than -10.5, -10.5, -8.4, -8, and -8.9 points below the time 1 percentile for GMFCS level I through V respectively are interpreted as “children developing more poorly than expected”.

Three case examples were selected from the Move & PLAY study data set to explain the classification of outcome of motor function. Details of the characteristics of the children selected, with their names changed are provided in Table 4.2. The GMFM-66-B & C score of Jessica with GMFCS level V, changed, from 8.12 to 18.01 with a change of 9.89 points. The percentile ranking of Jessica changed from 1 (at time 1) to 16 (at time 2) with a change of 15 percentile points. Based on the expected interval of change in percentiles between assessments (i.e.  $\pm 8.9$  for GMFCS level V) Jessica is developing better than expected as her change in percentile rank is above the expected interval. The GMFM-66-B & C score of Noah who has GMFCS level I changed from 84.05 to 87.99 with a change score of 3.94. The percentile ranks of Noah decreased from 84<sup>th</sup> percentile to 81<sup>st</sup> percentile with a percentile difference of -3. Although there is a decrease in the percentile rank, the values are within the expected interval of change in percentile ranking (i.e.  $\pm 10.5$  for GMFCS level I) and therefore, Noah is developing as expected. Conversely, the GMFM-66-B & C score of Catherine with GMFCS level III, changed from 44.97 to 42.61 with a change score of -2.36, and the percentile ranks

decreased from 26<sup>th</sup> percentile to 10<sup>th</sup> percentile with a difference of -16. Catherine is developing more poorly than expected, as the change in percentile rank is below the expected interval (i.e.  $\pm 8.4$  for GMFCS level III).

**Table 4.2: Case examples**

<b>Case examples and parameter</b>	<b>Time 1</b>	<b>Time2</b>	<b>Percentile difference</b>	<b>Classification of outcome of motor function</b>
Jessica (Level V)				
Age, Y	4.3	5.3		
GMFM – 66- B & C score	8.12	18.01		
Percentile	1	16	15	Developing better than expected
Noah (Level 1)				
Age, Y	4.8	5.8		
GMFM – 66- B & C score	84.05	87.99		
Percentile	84	81	-3	Developing as expected
Catherine (Level III)				
Age, Y	3.5	4.5		
GMFM – 66- B & C score	44.97	42.61		
Percentile	26	10	-16	Developing more poorly than expected

GMFM – 66 – B & C – Gross Motor Function Measure – 66 – Basal & Ceiling, Y - Years

This method of classifying children with CP as “developing better than expected”, “developing as expected” and “developing more poorly than expected” using the reference percentile curves helps therapists interpret change over time and understand and compare each child’s capacity with the development of children with CP with same functional level.<sup>79</sup> The main focus of this Chapter is to explore the prognostic implications of the BFI-CP I and II and the GMFCS for change in motor function using the reference percentile method proposed by Hanna and colleagues.

## Objectives

Objective 1: To explore the relationship between the Body Function Index in Cerebral Palsy Version I (BFI-CP I) and outcome of change in motor function based on 50% probability that children are developing “better than expected”, “as expected”, or “more poorly than expected”.

Objective 2: To explore the relationship between the Body Function Index in Cerebral Palsy Version II (BFI-CP II) and outcome of change in motor function based on 50% probability that children are developing “better than expected”, “as expected”, or “more poorly than expected”.

Objective 3: To explore the relationship between the Gross Motor Function Classification System and outcome of change in motor function based on 50% probability that children are developing “better than expected”, “as expected”, or “more poorly than expected”.

## Methods

### Design

This study is a secondary analysis. Data for this part of this thesis was also extracted from an existing data base from the Canadian Institutes of Health Research (CIHR-) funded multi-site longitudinal cohort study "Move & PLAY". Permission to use the data was obtained from the Move & PLAY study team.



## Participants

The Move & PLAY study database had 430 children included in the study.

Twenty five children were excluded due to various reasons detailed in Appendix 3.1.

Forty children were further excluded due to missing time 1 or time 2 Gross Motor

Function Measure scores. Finally, 365 children between the ages 18 months and 5 years were included for the purpose of this study. Detailed description of the characteristics of the children and the parents participants of this study are provided in Table 4.3.

**Table 4.3: Demographics**

<b>Child characteristics</b>		<b>Mean (SD) (N=365)</b>
Age in months		38±11
		<b>Frequency (Proportion)</b>
Child's gender	Boys	204(56)
	Girls	161 (44)
Child's race	African American or Black (not of Hispanic origin)	23 (6)
	Asian or Pacific Islander	15 (4)
	Hispanic/Latino	14 (4)
	Native American/North American Indian/Metis/Inuit	9 (3)
	White (not of Hispanic origin)	257 (70)
	Bi-racial &Others	47 (13)
Distribution of involvement	Monoplegia	8 (2)
	Hemiplegia	88 (24)
	Diplegia	84 (23)
	Triplegia	23 (6)
	Quadriplegia	162 (45)
GMFCS level	GMFCS I	129 (35)
	GMFCS II	45 (12)
	GMFCS III	47 (13)
	GMFCS IV	65 (18)
	GMFCS V	79 (22)

Parent characteristics		Frequency (Proportion)
Relationship with the child	Mother	314 (86)
	Father	20 (6)
	Other	31 (8)
Parental education	Less than high school	9 (3)
	High school or GED	102 (28)
	Community college diploma; Technical degree/ Associates degree	94 (26)
	Bachelors degree	88 (24)
	Masters degree	60 (16)
	Doctoral degree	12 (3)
Household income (N=353)	less than \$15,000	30 (9)
	\$15,000 - \$29,999	39 (11)
	\$30,000 - \$44,999	44 (12)
	\$45,000 - \$59,999	50 (14)
	\$60,000 - \$74,999	46 (13)
	\$75,000 or more	144 (41)

GMFCS = Gross Motor Function Classification System, GED = General equivalency diploma

## Measures

The Modified Ashworth Scale,<sup>69</sup> Early Clinical Assessment of Balance,<sup>71</sup> distribution of involvement<sup>7</sup>, Functional Strength Assessment,<sup>72</sup> Spinal Alignment and Range of Motion Measure,<sup>73</sup> Early Activity Scale for Endurance,<sup>74</sup> and Health Conditions Questionnaire<sup>75</sup> were used to derive two versions of the BFI-CP. The detailed description of the measures including the psychometric properties are presented in Chapter 3.

Gross motor function was measured using the basal and ceiling approach of the Gross Motor Function Measure (GMFM-66-B & C).<sup>81</sup> The GMFM-66-B & C<sup>81</sup> is a reliable (ICC = 0.99) and a valid (ICC = 0.98) measure in which original GMFM-66<sup>80</sup> items are arranged in increasing difficulty order. This adapted measure is administered

with a basal score of three consecutive scores of 3 (completes) for three items through to a consecutive three scores of 0 (does not initiate) with at least 15 items between the basal and the ceiling scores.

## Data Collection Procedures

As described earlier, the data were collected at three time points for each child from six provinces in Canada and four regions in United States between July 2007 and March 2010. Data collected at time one were used to develop the new indices. At time 1, the following measures completed by both parents and assessors were used for the purpose of this study: Gross Motor Function Classification System (GMFCS), Gross Motor Function Measure-66 B&C<sup>81</sup> (GMFM-66-B&C), Modified Ashworth Scale,<sup>69</sup> Early Clinical Assessment of Balance,<sup>71</sup> distribution of involvement<sup>7</sup>, Functional Strength Assessment,<sup>72</sup> Spinal Alignment and Range of Motion Measure,<sup>73</sup> Early Activity Scale for Endurance,<sup>74</sup> and the Health Conditions Questionnaire.<sup>75</sup> At the end of the study (one year from the initial visit - time 3) data from a therapist-completed measure (GMFM-66-B&C)<sup>81</sup> was also used for the purpose of this study. Data collected at time 1 and time 3 were used to explore the objectives of this study. As described earlier, all the assessors involved in the Move & PLAY study were trained and criterion tested.

## Data analyses

The GMFM-66-B & C data were collected at two different times, at an average of a one-year interval. The total scores were converted into percentile scores and the difference between the percentile scores was calculated. The change in the percentile

scores was classified into developing "better than expected", "as expected" or "more poorly than expected" for each GMFCS level based on 50% probability explained by Hanna et al<sup>79</sup> as described in the introduction.

The BFI-CP I was developed using a simple summing technique and quintile approach. The BFI-CP II was developed using cluster analysis. Please refer to Chapter 3 for detailed descriptions. Spearman's correlation co-efficient is a non- parametric statistical method of assessing the possible association between two variables. The values of the correlation co-efficient can be anywhere between +1 and -1. Values closer to  $\pm 1$  indicates a strong relationship and the values closer to 0 indicates a weak relationship. Spearman's correlation co-efficient is an appropriate technique when one or both the variables are skewed or rank ordered. In addition, the Spearman's correlation co-efficient is robust to outliers.<sup>36</sup> Therefore Spearman's correlation co-efficient was used to explore the relationship between the two versions of the BFI-CP and the GMFCS and the change in outcome of motor function. The significance level was set as 0.05.

## Results

Tables 4.4 to 4.6 contain the cross tabulations between the BFI-CP I, BFI-CP II, and GMFCS and GMFM. Table 4.7 describes statistically significant but weak correlations between the BFI-CP I, BFI-CP II and the GMFCS and outcome of change in motor function based on the 50% probability that children are developing 'better than expected', 'as expected', or 'more poorly than expected'.

**Table 4.4: Cross tabulation of the distribution of frequencies between the Body Function Index in Cerebral Palsy version I and motor outcome classification**

Classification of outcome of motor function based on 50% probability	BFI-CP I				
	I	II	III	IV	V
Developing better than expected	23	24	41	41	25
Developing as expected	34	36	24	26	23
Developing more poorly than expected	21	14	9	7	17

BFI-CP I – Body function index in cerebral palsy version I

**Table 4.5: Cross tabulation of the distribution of frequencies between Body Function Index in Cerebral Palsy version II and motor outcome classification**

Classification of outcome of motor function based on 50% probability	BFI-CP II				
	I	II	III	IV	V
Developing better than expected	23	39	38	30	24
Developing as expected	44	36	19	28	23
Developing more poorly than expected	23	17	4	8	16

BFI-CP II – Body function index in cerebral palsy version II

**Table 4.6: Cross tabulation of the distribution of frequencies between the Gross Motor Function Classification System and motor outcome classification**

Classification of outcome of motor function based on 50% probability	GMFCS				
	I	II	III	IV	V
Developing better than expected	33	22	29	40	30
Developing as expected	63	15	15	22	28
Developing more poorly than expected	33	8	3	3	21

GMFCS – Gross Motor Function Classification System

**Table 4.7: Relationships between Body Function Index in Cerebral Palsy version I and motor outcome classification, Body Function Index in Cerebral Palsy version II and motor outcome classification, and Gross Motor Function Classification System and motor outcome classification**

Relationships	Spearman's rho	Approx. Sig
BFI-CP I and outcome classification	0.12	<b>0.02</b>
BFI-CP II and outcome classification	0.16	<b>0.02</b>
GMFCS and outcome classification	0.15	<b>0.005</b>

BFI-CP - Body function index in children with cerebral palsy, GMFCS - Gross Motor Function Classification System

## Discussion

The overall findings of this study indicate that it is challenging to predict change in motor function (outcome classification) using either a holistic classification system or

the international gold standard classification (i.e. the GMFCS) in children with CP. There were statistically significant, but weak, correlations between two versions of the holistic classifications and the GMFCS and change in gross motor function measured using the GMFM and classified according to the 50% probability method proposed by Hanna et al.<sup>79</sup> The results are statistically significant in spite of weak correlations because of the large sample size. Only 1.4% of variance in change in gross motor function was explained by the BFI-CP I, 1.6% of variance in change in gross motor function was explained by the BFI-CP II, and 2.2% of variance in change in gross motor function was explained by the GMFCS. Scatter plots in Appendix 4.1, in combination with the cross tabulations in tables 4.4 to 4.6, do not show non-linear relationships.

The unanticipated findings of this study could be attributed (at least in part) to the heterogeneity of children with CP. Two case examples (with names changed) extracted from the Move & PLAY study data are provided in Table 4.8. These examples are framed in the context of the scaling of scores used to construct the BFI-CP version I classification (quintile approach) as described in the previous chapter (i.e. higher scores represent “better performance”). The case examples show the relative difference in strengths of each variable, although both of the children have similar BFI-CP I scores. In this case example, Lucas has a more functional GMFCS level (level III) with higher balance scores, and slightly less impact of health conditions than William, but Lucas also has poorer strength and endurance scores and more spasticity and range of motion restrictions than William who has a less functional GMFCS level (level IV) with lower balance scores and quadriplegia but higher strength, spasticity, and endurance scores and less range of motion restrictions than Lucas.

**Table 4.8: Case example**

<b>Case example and parameters</b>	<b>Lucas</b>	<b>William</b>
BFI-CP I score	27.55	27.56
Age	56 months	31 months
GMFCS level	III	IV
Distribution of involvement	Diplegia	Quadriplegia
Balance score	4.1	2.3
Spasticity score	4.25	5
Strength score	3.25	4.13
SAROMM score	2.58	3.88
Endurance score	3.5	5
Health score	6.88	6.25

BFI-CP I – Body function index in cerebral palsy version I,  
 GMFCS – Gross Motor Function Classification System,  
 SAROMM – Spinal Alignment and Range of Motion Measure

In addition to the description of scores, it is useful to explore and compare the health conditions of Lucas and William. The health conditions scores for Lucas is 6.88 and that for William is 6.25. Both the children have problems seeing; however, it doesn't have any impact on Lucas' activities of daily living. In contrast, problems with seeing affects William "to a moderate extent" possibly, in part, explaining lower balance scores. In addition, William also has problems with learning and understanding, which affects him to a small extent. Lucas has problems involving his mouth but it doesn't affect his daily activities at all. William has problems with digestion as well, which affects him "to a small extent". Furthermore, William also has problems with growth and his heart



however, they don't affect his daily activities. Although the difference between the health conditions raw scores is only 0.63, there is a marked heterogeneity between these two children with regard to the presentation and the impact of each health conditions on their lives. This heterogeneity thus likely explains part of the challenges associated with predicting change in gross motor function in children with CP who exhibit diverse features and comorbidities.

A second issue related to predicting change in children with CP may be attributed to the concepts of dynamic systems theory. Children with CP demonstrate inter-individual variation and developmental change cannot be generalized.<sup>82</sup> As speculated in dynamic systems theory,<sup>82</sup> development is non-linear and child development proceeds in spurts and plateaus over time. Developmental change is the result of the interaction of multiple systems.<sup>82</sup> Qualitative change reflects the emergence of new behaviour which occurs when there is a change in the state of attractor well.<sup>82</sup> The primary impairments in CP lead the emergence of secondary impairments. As stated in the literature, a unit change in a determinant may not necessarily result in a unit change in outcome.<sup>82,68</sup> A substantial improvement in range of motion may result in very small improvement in gross motor function and vice versa. In addition, children with CP demonstrate a wide variation in the rate of development,<sup>79</sup> the functional level of individual features, and impact of associated health conditions. The complexity and heterogeneity of the health condition, uniqueness of each child with CP, and speculations of dynamic systems theory, all challenge the prediction of change in motor function.

The findings of this study, in the context of related literature, also support the importance of examining children with CP from a broader perspective, providing a comprehensive holistic picture. Therefore each child with CP needs a comprehensive assessment of balance, distribution of involvement, spasticity, strength, range of motion, endurance, and presence of co-morbidities. Furthermore, each feature needs to be considered and interpreted separately in the context of the whole child, due to the non-linearity associated with the progression of each feature overtime.<sup>82</sup>

Although it is feasible to obtain a comprehensive picture of children with CP using the new indices, this study has several limitations in exploring their clinical utility. The psychometric properties of the two new indices were not determined (although they are based on measures with good psychometric properties themselves). Although other features of utility described in Chapter 2 were also not explored in this Chapter, the lack of association between classification and prognostic course limit their use as comprehensive indices for clinical decision making.

In summary, the heterogeneity of the health condition of CP increases the complexity and difficulty in predicting change in gross motor function (using outcome classification) using two holistic classification systems. Each feature of the child has to be observed, and interpreted, separately and it is important to understand individual childrens' strengths and weaknesses in order to plan treatment to support motor function. Case examples of this direction are provided in the final chapter.

## Chapter 5: Summary, implications, and conclusion

The work in this thesis was informed by the international consensus definition<sup>4</sup> which describes the clinical features of children with cerebral palsy (CP). The broader definition emphasizes the importance of a more inclusive classification. Therefore it was felt to be important to develop holistic classifications for describing and classifying children with CP.

Before working on the development of a holistic classification for children with CP, an effort was made to understand the strategies used in developing classification systems in several childhood disorders. This contributed to Chapter 2 of this thesis in which the prevailing classification systems in selected childhood disorders (i.e. developmental co-ordination disorder (DCD) and autism spectrum disorder (ASD)) in addition to CP (Chapter 1) were reviewed. The findings of this preliminary work demonstrated that there is a gap in the literature with regard to classifying children with CP addressing the key features of the health condition. This work suggested methods (i.e. cluster analysis) that could be used to develop a holistic classification in children with CP.

Chapter 3 of this thesis focused on development of holistic classification systems in children with CP addressing the majority of the key features of the international consensus definition of CP that were available in a pre-existing database using both a simple summing technique and cluster analysis. The overlap of the ranges of the values in both of the new indices demonstrated the individualized presentation of children with CP.

The findings of this study also demonstrated a significant correlation of the two new indices with the international gold standard classification system (Gross Motor Function Classification System [GMFCS]<sup>10,20</sup>). The cross tabulations between the two new indices and the GMFCS indicates that one third of the children with CP are different (Table 3.7 and 3.8). Therefore it is important to consider this heterogeneity while understanding individual children with CP.

The results of cluster analysis in this study are different from the cluster analyses of children with DCD and ASD (Chapter 2). In DCD and ASD, the clusters described were discrete (eg. ideomotor dyspraxia, visual spatial and visual constructional dyspraxia, and mix dyspraxia; essential autism and complex autism). In contrast, the clusters derived on a sample of children with CP in this study were in rank order with similar results to the simple summing technique.

A second motivation to explore a more holistic classification was to conduct a study parallel to the Comprehensive Severity Index (CSI) (informed by the International Classification of Diseases) but for rehabilitation professionals informed by the ICF. There is a difference between the CSI and the Body Function Index in Cerebral Palsy (BFI-CP) as described in Chapter 3. The CSI is used for analyzing the severity of illness and is used in predicting mortality, morbidity, cost, and length of hospital stay.<sup>67</sup> The CSI is calculated based on the physiologic measures such as laboratory measures.<sup>67</sup> The BFI-CP stratifies children with CP based on neuro-musculoskeletal functions and functional manifestation of associated co-morbid health conditions. A description of differences between the CSI and the BFI-CP is provided in Table 3.3.

The primary usefulness of a classification is based on prognostic implications. This led to the study described in Chapter 4 which focused on the prognostic implications of the two new indices. The findings indicated weak correlations between the two new indices and the GMFCS and the outcome of change in motor function. The variability and the complexity of the presentation of children with CP pose a major challenge in predicting change in motor function using either of the two new indices or the GMFCS based on the outcome classification system used in this study.

This chapter (Chapter 5) is focused on the clinical implications and application of the results of this thesis in administration, teaching, and research.

## Clinical implications

The clinical implications of the findings of this thesis are explained using case examples. Three case examples are children who were “developing as expected” in motor function and GMFCS level III, with an age range from 38 to 40 months of age. The Move & PLAY study results for children in GMFCS level III indicates strong relationships between primary impairments such as balance, spasticity, quality of movement and distribution of involvement and motor abilities and modest relationships of strength, range of motion, endurance, and adaptive behaviour with motor abilities.<sup>62</sup> Quality of movement and adaptive behaviour parameters were not used in this thesis and therefore, their associations with motor outcome are not discussed. The raw scores and the percentiles of the parameters of three children with CP whose names are changed are provided in Table 5.1. The percentiles presented in Table 5.1 were extrapolated

**Table 5.1: Case examples of determinants of motor function of children with Gross Motor Function Classification System III who are developing as expected**

Names changed	Age (months)	Distribution of involvement	ECAB		Strength		SAROMM		Endurance		Health conditions	
			Score	Percentile	Score	Percentile	Score	Percentile	Score	Percentile	Score	Percentile
Lisa	38	quadriplegia	3.6	70 <sup>th</sup>	3.75	50 <sup>th</sup>	1	70 <sup>th</sup>	4.75	95 <sup>th</sup>	.13	5 <sup>th</sup>
Chloe	39	Diplegia	2.85	40 <sup>th</sup>	5.0	99 <sup>th</sup>	.54	30 <sup>th</sup>	2.5	20 <sup>th</sup>	.13	5 <sup>th</sup>
Mathew	40	Diplegia	4.15	80 <sup>th</sup>	3.25	20 <sup>th</sup>	.85	65 <sup>th</sup>	2.5	20 <sup>th</sup>	1.75	99 <sup>th</sup>

ECAB – Early Clinical Assessment of Balance, SAROMM – Spinal Alignment and Range of Motion Measure.

approximately from the boxplots of the Move & PLAY model testing power point summary created using cross sectional data on children between 18 months and 5 years of age.<sup>83</sup>

Recall that the scores of the early clinical assessment of balance (ECAB),<sup>71</sup> Functional Assessment of Strength (FSA),<sup>72</sup> and Early Activity Scale for Endurance” (EASE),<sup>74</sup> were scaled such that higher percentiles represent strong balance, strong strength and greater endurance respectively. The scores of the Spinal Alignment and Range of Motion Measure (SAROMM)<sup>73</sup> were scaled such that higher percentiles represent more limitations. Scores on the Child Health Conditions Questionnaire”,<sup>75</sup> which measures associated health conditions were scaled such that higher percentiles represent greater impact of health conditions on daily activities of living.

Lisa has quadriplegia, strong balance, moderate strength, significant range of motion restriction, strong endurance, and low impact of health condition on daily activities. The percentile ranking of Lisa’s motor function changed from 47<sup>th</sup> to 47.5<sup>th</sup> percentile (difference of 0.5) between the two assessments. Chloe has diplegia, moderate balance, strong strength, less range of motion restriction, poor endurance, and low impact of health conditions on daily activities. The GMFM percentile ranking of Chloe changed from 32<sup>nd</sup> percentile to 34<sup>th</sup> percentile between the two assessments. Mathew has diplegia, strong balance, poor strength, moderate range of motion restriction, poor endurance, and high impact of health conditions on daily activities. Mathew’s GMFM percentile changed from 41<sup>st</sup> percentile to 44<sup>th</sup> percentile over the one year interval.

Although all the three children are described as developing as expected, the strengths and weaknesses of each child are different.

As stated earlier, children with CP have variations with multiple interacting systems. Identifying each child's strengths and limitations are important in planning intervention. In these case examples, although all three children are developing as expected, while planning intervention, therapists should identify the areas for improvement, areas for maintenance, and requirements for environmental modifications.

Lisa has room for improvement in strength and range of motion. Lisa's strengths are good balance, and endurance, and little impact of health conditions on daily activities. For Lisa, the therapist's plan for intervention might focus on improving strength and range of motion and maintaining balance and endurance to support motor function.

For Chloe, there is room for improvement in balance and endurance. The therapist might focus on maintaining strength and range of motion and analyze the components of balance and focus on improving balance and endurance to support motor function. With regard to improving endurance, the therapist might also analyze the requirements for provision or modification of assistive devices which might improve her endurance.

Mathew has room for improvement in strength, range of motion, and endurance. The therapist might focus on maintaining balance. Also, the therapist should review the health conditions questionnaire in detail for Mathew as the health conditions affect his activities of daily living to a greater extent. In this case example, Mathew has problems



seeing and problems with digestion, which affect his daily activities to a small extent. He has problems communicating, controlling emotion, and pain which affect his activities to a moderate extent. He also has problems in learning and understanding which affects his daily activities to a very small extent. He has problems with sleeping that affects his daily activities to a great extent. Mathew should be referred to appropriate health care professionals related to the health conditions that affect Mathew's daily activities. Specifically, Mathew might benefit from having a sleep study, as well as a referral to a psychologist for emotion control and initiating a detailed assessment of pain by his developmental pediatrician.

The intervention plan for each child will differ based on the individual child's strengths and limitations. It is beyond the scope of this thesis to review the effectiveness of various interventions. Although research evidence is important, the uniqueness of children with CP increases the challenges in applying evidence into practice. Palisano and colleagues<sup>84</sup> proposed recommendations for optimal pediatric rehabilitation services for children with CP. They propose that multiple sources of knowledge (i.e. research evidence, theory-based knowledge and practice-based evidence) should be considered in selecting services for children with CP.<sup>84</sup>

Therapists should identify the best research evidence and use their expertise to tailor the intervention that fits the child's strengths and needs.<sup>85</sup> Therapists must critically analyze internal and external validity of studies before making decisions.

The activity-focused intervention model proposed by Valvano<sup>86</sup> provides guidance and theoretical rationale for therapists in selecting intervention services for

children with developmental disabilities addressing the child's individual needs. Activity-focused intervention emphasizes practice and repetition of functional activities to improve the child's participation in daily activities. This model involves a therapist developing activity-related goals to increase participation, planning activity-focused interventions to provide opportunities for practicing functional activities by adapting the principles of motor learning and motor development to meet the child's strengths and limitations, and integrating impairment focused intervention with activity-focused intervention.<sup>86</sup>

The needs identified based on a comprehensive assessment could be used to develop activity-focused intervention strategies. It is beyond the scope of this thesis to discuss in detail the intervention plans for every single component of movement system for these case examples. Therefore the strategy for planning activity-focused intervention is explained for one component for one case example.

For example, Mathew's endurance is at the 20<sup>th</sup> percentile; his physical activity level is not similar to other children of his age. Let us assume that the family's desired outcome for Mathew is to move in and out of a chair on his own. This activity requires a lot of balance, strength, and endurance. Mathew's balance is at 80<sup>th</sup> percentile, his strength is at 20<sup>th</sup> percentile and his endurance is at the 20<sup>th</sup> percentile. The therapist could plan intervention to use his balance to improve his strength and endurance. Mathew could be provided with opportunities to push through his hands on varied tasks. Mathew could be encouraged to push through his hands during his daily activities such as moving in and out of the bath tub, moving between the floor and a low level bench, and so on.

Practice-based evidence (PBE) also serves as a good starting point for services that do not have research evidence.<sup>87</sup> PBE is considered as an alternative method for the randomized controlled trial (RCT).<sup>88</sup> PBE study designs take into account client and treatment differences and provide information on what happens during the usual care process (natural setting).<sup>87,88</sup> In addition, PBE studies have a low risk of bias<sup>88</sup> and are one of the best sources of information that could be used in intervention planning. This method of inquiry holds promise in heterogeneous conditions such as CP.

The BFI-CP versions might serve the purpose of examination based on the framework proposed by the APTA in Figure 2.1 in identifying the subgroup in which the child best fits, in evaluating the results based on the examination. The BFI-CP versions may also be used in selecting intervention according to the needs of the individual child as described above. The On Track study, which is in progress, is focusing on developing reference percentile and longitudinal growth curves to monitor many characteristics of children with CP as they age (<https://www.canchild.ca/en/research-in-practice/current-studies/on-track>). The results of the On Track study might shed light on the prognostic implications of the BFI-CP.

## Implications for administration

In terms of administration, managers should ensure that clinicians are provided time to acquire knowledge and have access to new measures and learn about clinical decision-making tools such as that offered through the Move & PLAY study. Managers should recognize the challenges faced by clinicians in dealing with heterogeneity and

provide appropriate mentorship to clinicians to gain expertise and knowledge in understanding children with CP holistically. Managers should encourage clinicians to administer, score, and interpret psychometrically sound measures, such as those described in this study, to describe children with CP more holistically. Each child presents uniquely and the rate of progression of all determinants of motor function does not occur at a steady pace.<sup>62</sup> Policies are the pathways to bring about change; therefore, managers should ensure that policies are in place to mandate regular comprehensive assessment to obtain a comprehensive picture of each child with CP.

## Implications for teaching

The overall findings could be used in physical therapy curricula. It is necessary to educate physiotherapy students about the importance of doing an ongoing comprehensive assessment of children with CP using psychometrically sound and clinically feasible measures such as those used in this dissertation.<sup>89</sup> The knowledge derived from this dissertation might help physical therapy students to understand the inter-individual variability of children with CP. The product of this work could be used to educate physiotherapy students about the challenges associated with predicting change in motor function. The findings could also be used as an example of comprehensive assessment to describe the uniqueness and identify the strengths and needs of each child with CP and enable development of attainable goals and plan intervention to address multiple features. Instructors should emphasize the importance of parents describing the overall health status of their children with CP and involving parents in developing family centered goals and in intervention decision making. Educating students about the challenges associated

with the heterogeneity of children with CP, the necessity to do comprehensive assessments for children with CP, and identifying the strengths and limitations will enable students to develop realistic goals and offer specific interventions to improve their selected goals during their professional practice.

## Implications for research

Although the results of this dissertation demonstrated that holistic classifications are not useful, the importance of doing a comprehensive assessment to understand the strengths and limitations of each child with CP has been elucidated. The box plots used in this thesis to determine percentiles for various measures were derived from the Move & PLAY study

(<http://canchild.ocean.factory.ca/system/tenon/assets/attachments/000/000/314/original/KeyFindingsMovePLAY.pdf>). These box plots were developed using cross sectional data on children between 18 months and 5 years of age. These box plots are only useful to interpret the meaning of various measures at a particular point of time and are not useful in understanding change over time. To interpret change over time, reference percentiles are required. The results of the On Track study may be of value in tracking the development of each characteristic of children with CP as they will provide information on change over a period of time.

The knowledge obtained from a comprehensive assessment, together with the results of the On Track study, may be useful in making decisions on selecting appropriate services for children with CP.<sup>67</sup> With regard to selecting intervention programs, RCTs are

regarded as the highest level of evidence. Nevertheless, there are many limitations of RCTs. RCTs do not reflect real world clinical settings. The RCTs determine group differences and eliminate individual differences using randomization.<sup>61,90</sup> In real world settings, children with CP are heterogeneous and present with multiple co-morbidities. RCTs best address body structure and function and are appropriate to test single interventions.<sup>61</sup> Therapists working with children with CP are not only interested in body structure and function, but also in activity and performance. RCTs are provided under standardized and controlled environments and eliminate interaction of the intervention with personal or environmental factors. Children with CP are greatly influenced by personal and environmental factors. In clinical settings, children with CP are provided multiple interventions and it may not be feasible to create a standardized condition. In regular clinical practice individuals are treated, not groups. Although, RCTs provide establishment of causal inferences, the results are not applicable for use in regular clinical practice.

Single subject designs are alternative experimental designs that explore the causal inferences at an individual level. Threats to internal validity are addressed through within subject and between subject comparisons. Threats to external validity are addressed by replicating the investigation by systematically changing one or more aspects of the intervention (systematic replication).<sup>36, 90, 91</sup> In single subject designs the participants serve as their own controls throughout the experiment. A treatment is considered effective if the effectiveness is demonstrated repeatedly and reliably within a single participant or across different participants.<sup>36, 90, 91</sup> It is beyond the scope of this thesis to describe in detail the types of single subject designs. Single subject designs can be used

to explore the effects of intervention programs and the environmental variables on performance at the individual level.<sup>91</sup> Therefore single subject experimental studies addressing the uniqueness of children with CP is warranted for making decisions on selecting appropriate services.

## Conclusion

The developed classification systems are of limited use in classifying children with CP holistically. However, a comprehensive assessment using multiple measures might complement the functional classification systems including the GMFCS, the Manual Ability Classification System, and the Communication Function Classification System in describing and understanding children with CP. It is also clear from the results of this thesis that it is challenging to predict gross motor function (using outcome classification) using the two new indices. However, the relative strengths and weaknesses of each determinant of motor function should be monitored individually. Therapists should use results of individual measures, extract information from multiple sources of knowledge, and use their critical thinking in making decisions and select interventions that fits the child's and families' goals.

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## Appendix 2.1

## Details of database search

### Methods

#### Database search

A literature search on PubMed, CINAHL, SCOPUS, Cochrane Library, Embase, and Proquest databases and Google was conducted using the combination of terms including “*classification*” or “*subtypes*” or “*subgroups*” and “*Developmental Coordination Disorder*” or “*Autism Spectrum Disorder*” with the goal of identifying ways of classifying children with these selected neuro-disabilities. The search was restricted from the year 2000 to June 2013 in order to focus on currently used classification systems. Articles were included if they provided information or focused on subgrouping of the DCD or ASD and if a specific classification system of any of the three selected neuro-disabilities was used in a study. Articles published in languages other than English and articles that focused on assessment, screening, or treatment and did not provide any information on subtyping of any of the three selected neuro-disabilities were excluded.

#### Procedure

Different ways of classifying the two selected neuro-disability disorders were identified through a thorough review of the identified relevant literature. Next, the measures used to describe the constructs of each classification system were identified. The contents of the individual measures were analyzed and the overall contents of each

measure were mapped to the ICF constructs. Specifically I am interested in differentiating between capacity and performance, therefore the overall contents were mapped to the qualifier, (i.e. capacity and performance) and/or body function components wherever applicable.

## Appendix 2.2

### Psychometric properties of measures used in studies to identify subgroups of children with Developmental Coordination Disorder

KAT <sup>2.2.1</sup>	Test-retest $r = 0.90$
MVPT <sup>2.2.2</sup>	Internal consistency: $r = 0.81$ to $r = 0.84$ Test-retest reliability: $r = 0.77$ to $r = 0.83$ Correlation between the MVPT and the Spatial awareness subscale of the Rivermead Perceptual Assessment Battery was $r = 0.72$ . Correlations between the MVPT-3 and the Developmental Test of Visual Perception - 2 was $r = 0.27$ to $r = 0.82$ Correlation between the MVPT-3 and the Developmental Test of Visual Perception was $r = 0.38$ to $r = 0.73$
DT-VMI <sup>2.2.3</sup>	Split half correlation across age groups was 0.95 Inter-rater reliability was 0.73 to 0.99 Correlation between Beery VMI and Comprehensive test of Basic Skills was 0.63 Correlation between Beery VMI and Bender-Gestalt ranged from 0.29 to 0.93 Correlation with Wide Range Assessment of Visual Motor Abilities was 0.52
BOTMP <sup>2.2.4</sup>	Test-retest reliability: ICC=0.58 to 0.89 Test-retest reliability: $r = 0.69$ to 0.80 Interrater reliability $r = 0.63$ to 0.97 Correlations between BOT-2 and BOTMP correlation on total composite was adj $r = .80$ . Correlation between BOT-2 and PDMS-2 was $r = 0.73$
MABC <sup>2.2.5</sup>	Test-retest reliability: ICC = 0.62-0.92 Interrater reliability: ICC = 0.92 to 1.00 Correlation coefficient with BOTMP ( $r = 0.53$ to 0.79) Correlation with Berry-VMI (0.31 to 0.35) Correlation with PDMS ( $r = 0.76$ )

COMPS <sup>2.2.6</sup>	Test-retest reliability: ICC = 0.93 Interrater reliability: ICC = 0.88 Correlation between total COMPS score and BOTMP Battery composite r=0.561
WISC <sup>2.2.7</sup>	Reliability co-efficient by split half technique r=0.92 to 0.95 (spearman's correlation co-efficient) Correlation between DTVP-2 and WISC-R ranged from 0.4 to 0.8 Correlation between WISC-IV and Perceptual Reasoning Index was r = 0.86
Block Design <sup>2.2.8</sup>	Close approximation between Binet and Block design medians Correlation between Binet IQ and Block design is 0.57 to 0.82
Rey-Osterreith Complex Figure Test 2.2.9,2.2.10	Inter rater reliability:0.88 to 0.97 Intrarater reliability: 0.93 to 0.98 Discriminate brain damaged and psychiatric individuals from normal individuals
Bell Crossing test <sup>2.2.11</sup>	Test-retest reliability: ICC = 0.84 Correlation between Catherine Bergego Scale and bells test ranged from r = 0.50 to 0.74
Porteus Labyrinth Test <sup>2.2.12</sup>	Internal consistency: Cronbach's alpha = 0.81 Correlation between the Porteus Vineland series and the Porte Us Extension Series range from r = 0.50 to 0.85
TLT <sup>2.2.13</sup>	Test-retest correlations for the TLT score ranged from r=0.58 to 0 .66 Internal consistency: Cronbach's alpha =0.30 Correlations between the TLT, and WAIS-R Digit Span (total score and backwards) ranged from r = 0.50 to 0.61 Correlations between the TLT, and Raven progressive matrices was r = 0.55 Correlations between the TLT, and Test of Divided Attention was r = 0.55
DTVP <sup>2.2.14</sup>	Test-retest Reliability Coefficients: r= 0.92 to 0.95 Inter rater reliability ranged from 0.93 to 0.99 Correlations between the MVPT-3 and the DTVP - 2 was r = 0.27 to r = 0.82 Correlations between the MVPT-3 and the DTVP was r = 0.38 to r = 0.73 Correlation between DTVP-2 and WISC-R ranged from 0.4 to 0.8
Hand writing scale	Not available

Language Screening battery	Not available
Kinaesthetic perception	Not available
Neuro-Psychomotor Test <sup>2.2.15</sup>	Correlation co-efficient with Lincoln-Oseretsky Motor Development Scale ranged from 0.72 to 0.84
Electroretinogram	Not available
Motor Electro-oculogram	Not available
Visually Evoked Potentials <sup>2.2.16</sup>	Test-retest reliability: ICC = 0.88

ICC = intraclass correlation co-efficient, r = reliability co-efficient, KAT = Kinaesthetic Acuity Test, MVPT = Motor Free Visual-Perception Test, DT-VMI = Developmental Test of Visual Motor Integration, BOTMP = Bruininks Oseretsky Test of Motor Proficiency, MABC = Movement Assessment Battery for Children, COMPS = The Clinical Observations of Motor and Postural Skills, WISC = Wechsler Intelligence Scale for Children, TLT = Tower of London Test, DTVP = Developmental Test of Visual Perception, WAIS – R = Wechsler Adult Intelligence Scale Revised.

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## Appendix 2.3

### Psychometric properties of measures used in studies to identify subgroups of children with Autism Spectrum Disorders

Measures	Psychometric Properties
Wing Autistic Disorder Interview Checklist <sup>2.3.1</sup>	Not Available
The Stanford-Binet Intelligence Scale <sup>2.3.2</sup>	Reliability co-efficients across ages ranged from 0.83 to 0.98 Reliability established using McNemar's analysis Correlation with Leiter International performance scale $r= 0.79$
Bayley Scales of Infant Developmental <sup>2.3.3</sup>	Test-retest reliability: range from 0.53 to 0.91 Correlations between Bayley and Griffiths scales ranged from $r=0.530$ to 0.83
PPVT <sup>2.3.4</sup>	Split half reliability: alpha co-efficients ranged from 0.93 to 0.98 Test-retest coefficients ranged from .92 to .96 Correlation between the PPVT-3 and the Wechsler Intelligence Scale for Children - 3 ranged from 0.82 to 0.92 Correlation between the PPVT-3 and the Kaufman Adolescent and Adult Intelligence Test ranged from 0.76 to 0.91 Correlation between the PPVT-3 and the Kaufman Brief Intelligence test range from 0.63 to 0.83
VABS <sup>2.3.5</sup>	Survey form: <ol style="list-style-type: none"> <li>1. Split half reliability coefficients for the Adaptive Behaviour composite: <math>r= 0.89</math> to 0.98</li> <li>2. Test retest reliability co-efficient <math>r = 0.77</math> to 0.93</li> <li>3. Inter rater reliability: <math>r=0.62</math> to 0.78</li> </ol> Expanded form <ol style="list-style-type: none"> <li>4. Split half reliability coefficients for the Adaptive Behaviour composite: <math>r= 0.94</math> to 0.99</li> <li>5. Test retest reliability co-efficient <math>r = 0.80</math> to 0.90</li> </ol> Correlation between the Adaptive Behavior Composite and the original Vineland unadjusted Social Quotient was 0.55 Correlation between the Adaptive Behavior Composite and Silverstein's Deviation Social Quotient was 0.55 Correlation between revised and original Vineland was 0.97 Correlation between VABS and the Adaptive Behavior Inventory for Children was 0.58 Correlation between VABS and PPVT-R was 0.28

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ADI-R <sup>2.3.6</sup>	Inter rater reliability: Weighted Kappa ranged from 0.62 to 0.89 ICC value range from 0.93 to 0.97 Cronbach's alpha: ranged from 0.69 to 0.95 ADI-R can discriminate autistic from mentally handicapped/language-impaired preschool children.
Head circumference <sup>2.3.7, 2.3.8</sup>	Interrater reliability: $r = 0.93$ ICC = 0.93 Correlation between fetal brain volume and head circumference was $r=0.97$
Brain MRI <sup>2.3.9 – 2.3.11</sup>	Test-retest reliability of structural brain networks from diffusion MRI ICC ranged from 0.62 to 0.76 Inter rater reliability kappa value ranged from 0.29 and 0.92 Reliability of multicentre MRI ICC was 0.96
Brain EEG <sup>2.3.12</sup>	Test-retest reliability ranged ICC = 0.8 to 0.95 Internal consistency alpha co-efficient range from 0.43 to 0.94
CELF <sup>2.3.13</sup>	Internal consistency reliability coefficients across ages ranged from $r= 0.87$ to 0.95 Internal consistency reliability coefficients across clinical groups ranged from $r= 0.83$ to 0.95 Correlation between CELF-4 and CELF-3 ranged from 0.80 to 0.87
Leiter International performance scale <sup>2.3.14, 2.3.15</sup>	Split half reliability: 0.91 to 0.94 Test-retest reliability co-efficient: 0.91 Correlation with WISC-R full scale IQ ( $r=0.74$ ) Correlation with Stanford-binet $r= 0.79$ Leiter has better discriminative value as determined by Arthur point Scale. Reliability Co-efficient as determined by split half method $0.91\pm 0.031$
WISC <sup>2.3.16</sup>	Reliability co-efficient by split half technique $r=0.92$ to 0.95 (spearman's correlation co-efficient) Correlation between DTVP-2 and WISC-R ranged from 0.4 to 0.8 Correlation between WISC-IV and Perceptual Reasoning Index was $r = 0.86$
Sensory Questionnaire	Not Available
Kinsbourne Overfocusing Scale	Not Available
DSM IV checklist <sup>2.3.17, 2.3.18</sup>	Interrater reliability kappa value $k=0.55$ Inter rater reliability was $r =0.89$ Test retest reliability was $r = 0.97$ Cronbach's alpha: 0.95
Photoarticulation Test	Not Available
The Short Sensory Profile <sup>2.3.19</sup>	Internal consistency Cronbach's co-efficient alpha = 0.47 to 0.91 Content validity was confirmed during test development and their results showed that 80% of the therapists agreed on 63% of the items on the category placement and new categories were developed for the remaining.

	Convergent and discriminant validity were established by correlating with School Function Assessment.
The Yale-Brown Obsessive Compulsive Scale <sup>2.3.20</sup>	Internal consistency: 0.69 Inter rater reliability (ICC) was 0.93 Test-retest reliability (ICC) was 0.61 Correlation with Behavioral Avoidance Test was 0.43 Correlation with Mandsley Obsessional Compulsive Inventory was 0.43
ICC = intraclass correlation co-efficient, r = reliability co-efficient, k=Kappa co-efficient, PPVT = Peabody Picture Vocabulary Test, VABS = Vineland Adaptive Behavior Scales, ADI-R= Autism Diagnostic Interview Revised, MRI = Magnetic Resonance Imaging, EEG= Electroencephalogram, CELF = Clinical Evaluation of language fundamentals, WISC = Wechsler Intelligence scale for children, DSM = Diagnostic Statistical Manual of Mental Disorders.	

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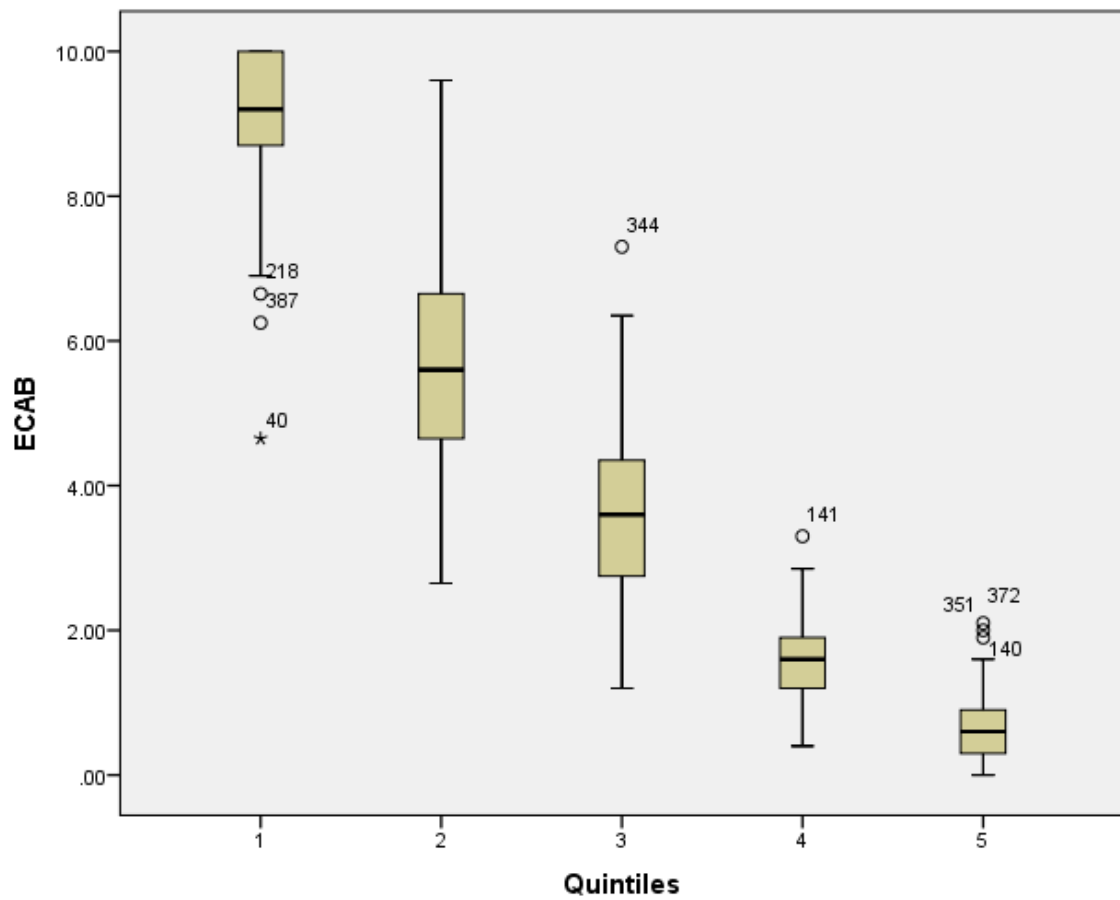
## Appendix 3.1

### Dealing with missing data

The Move & PLAY database originally had 430 cases. Twenty cases with missing Early Clinical Assessment of Balance scores were deleted as we couldn't recover the data. Two further cases were deleted due to missing distribution of involvement scores. Two more cases were further deleted due to missing Functional Strength Assessment (FSA) scores. One case was further deleted due to missing Endurance score. We kept one case with one missing item in FSA score and the FSA average for this case was calculated by adjusting the denominator. One case with four missing Spinal Alignment and Range of Motion Measure (SAROMM) items was also kept and the SAROMM mean score for this case was calculated by adjusting the denominator. Two cases in which parents rated "not applicable" for Endurance score were re-coded as "0" based on the On Track study criterion for this scoring pattern. Two cases with one missing Health conditions items were also kept and the average was calculated by adjusting the denominator.

## Appendix 3.2

Figure 1: Boxplots for Early Clinical Assessment of Balance in Quintiles (Body Function Index in Cerebral Palsy - I)



ECAB = Early Clinical Assessment of Balance

Figure 2: Boxplots for spasticity in Quintiles (Body Function Index in Cerebral Palsy - I)

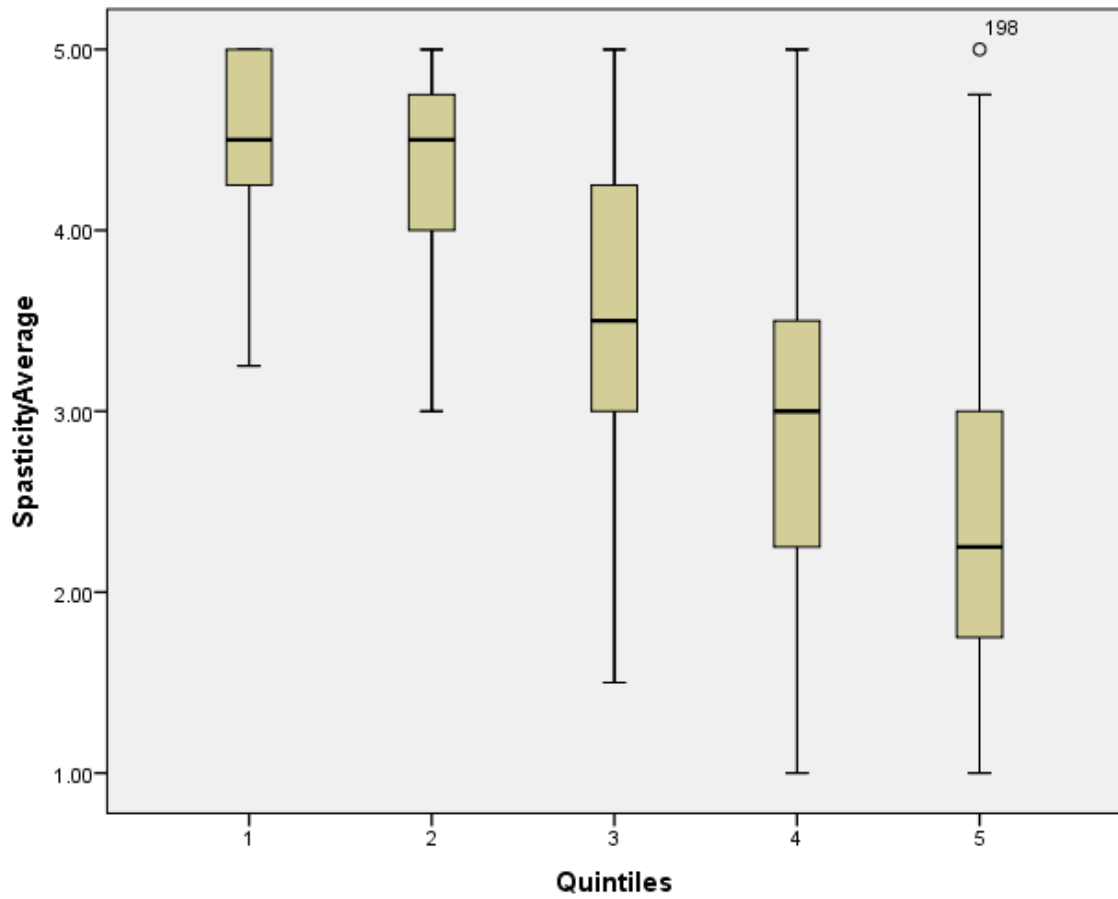
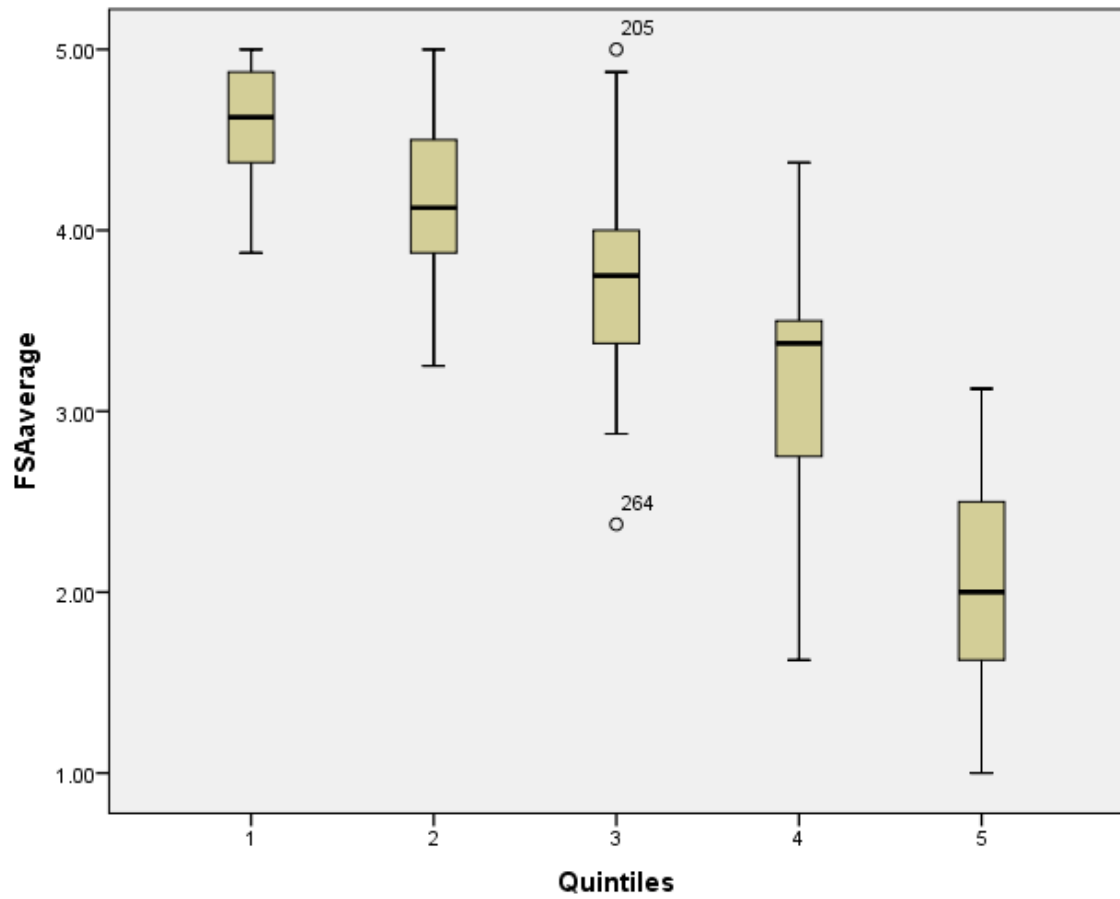


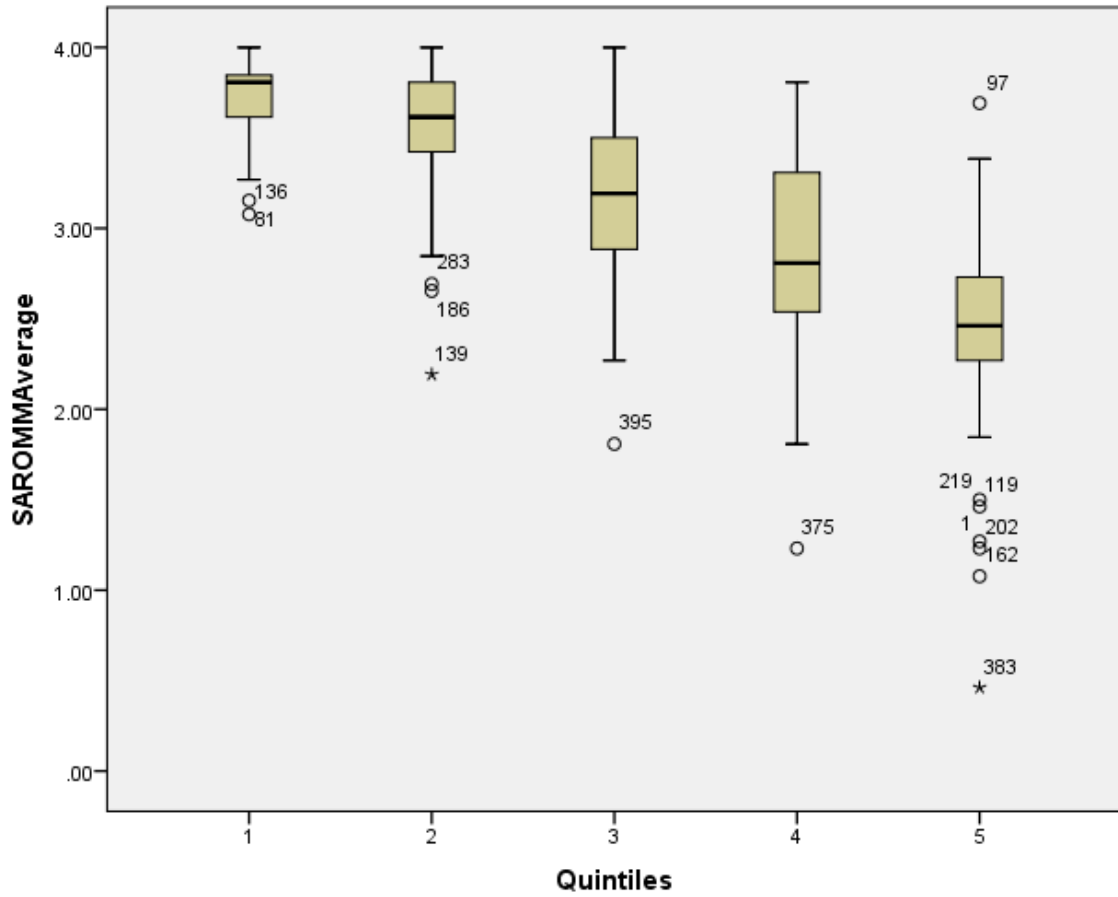


Figure 3: Boxplots for strength in quintiles (Body Function Index in Cerebral Palsy - I)



FSA = Functional Strength Assessment

Figure 4: Boxplots for Spinal Alignment and Range of Motion Measure in quintiles (Body Function Index in Cerebral Palsy – I)



SAROMM = Spinal Alignment and Range of Motion Measure

Figure 5: Boxplots for endurance in quintiles (Body Function Index in Cerebral Palsy - I)

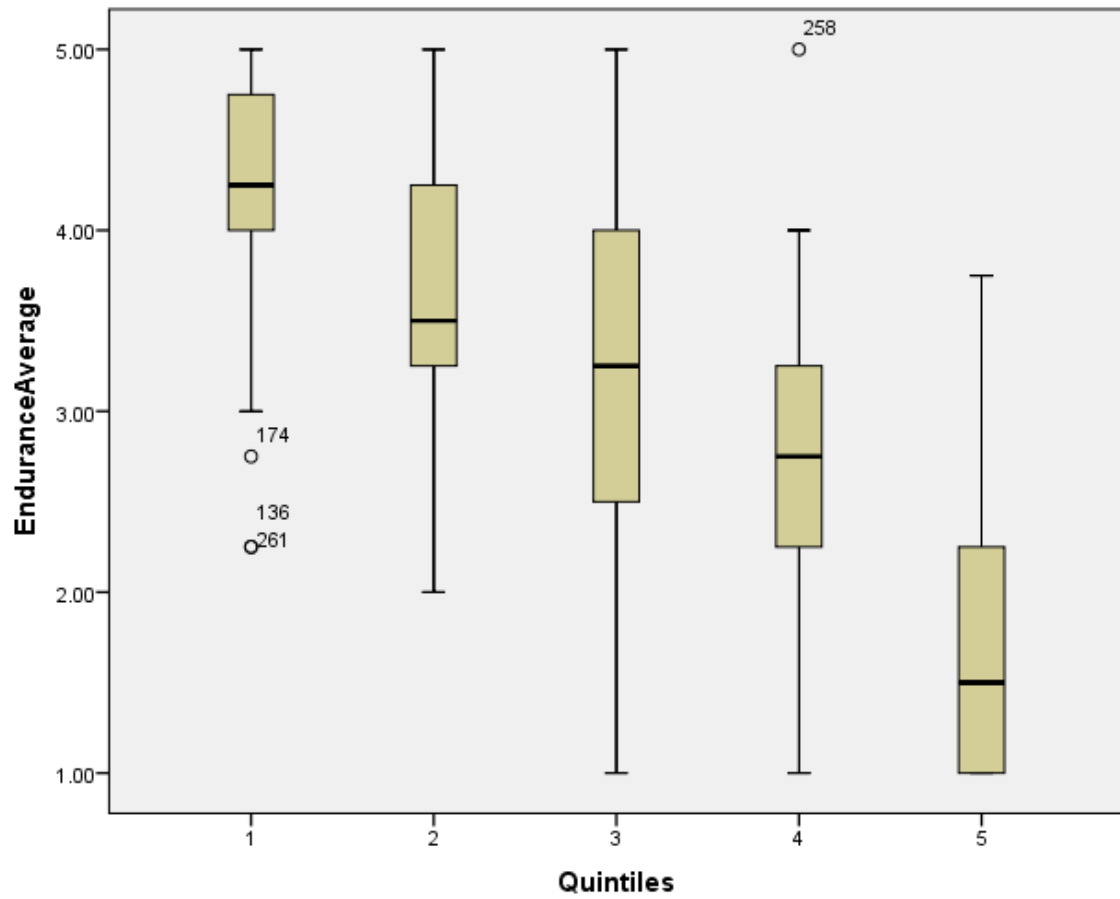
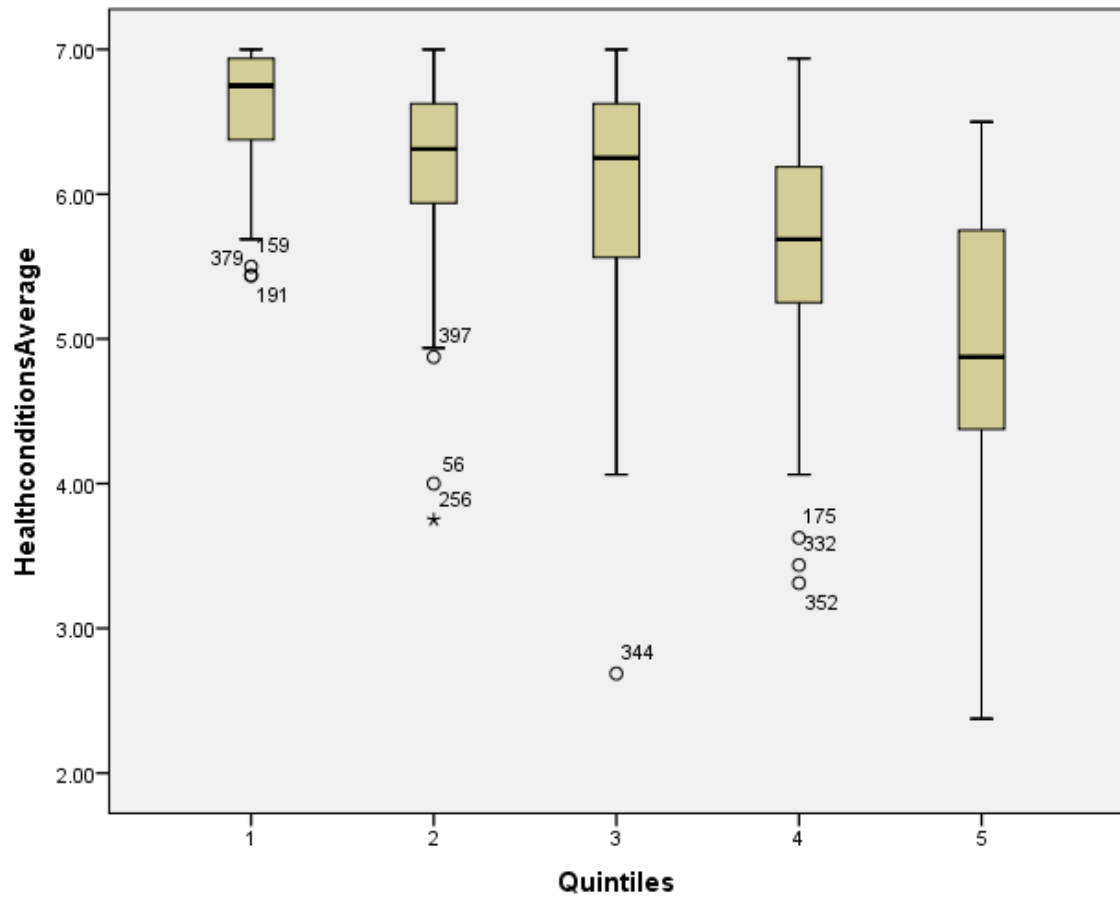
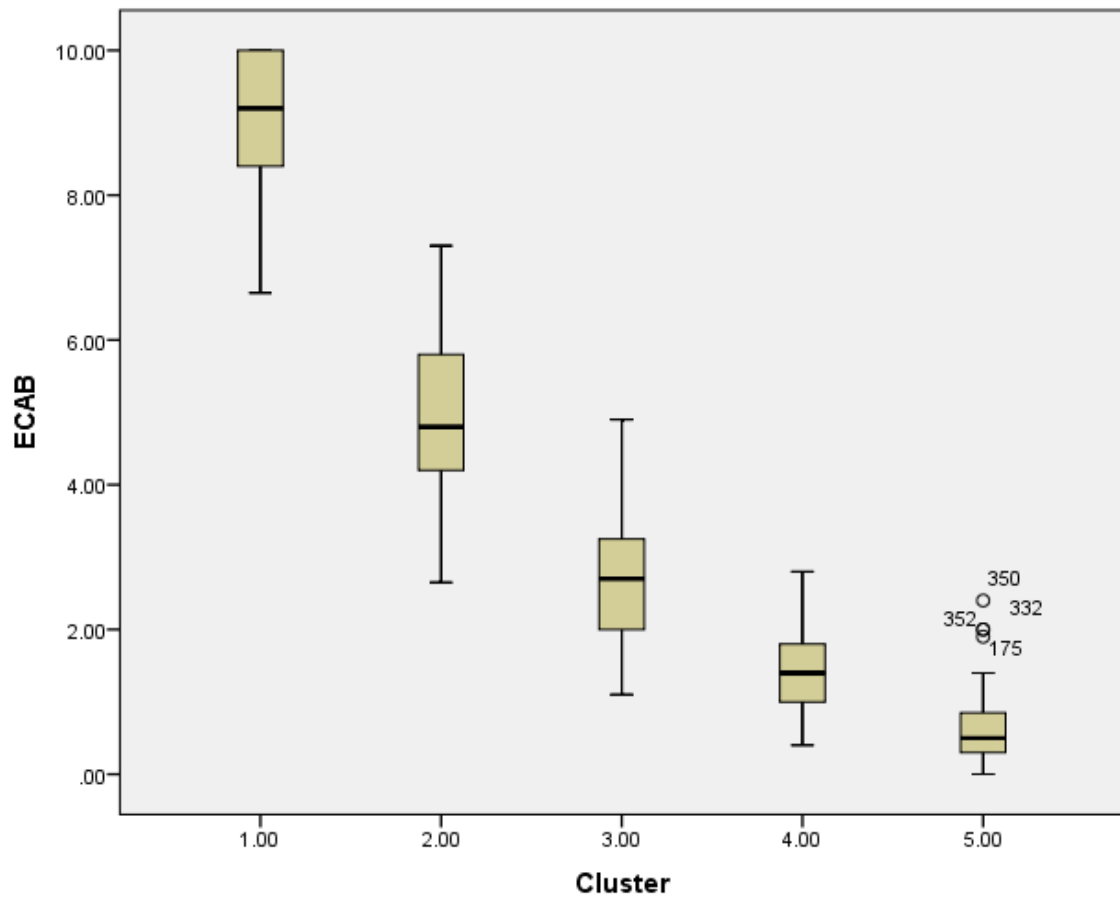


Figure 6: Boxplots for health conditions in quintiles (Body Function Index in Cerebral Palsy – I)



## Appendix 3.3

Figure 1: Boxplots for Early Clinical Assessment of Balance in cluster (Body Function Index in Cerebral Palsy - II)



ECAB = Early Clinical Assessment of Balance

Figure 2: Boxplots for spasticity in cluster (Body Function Index in Cerebral Palsy - II)

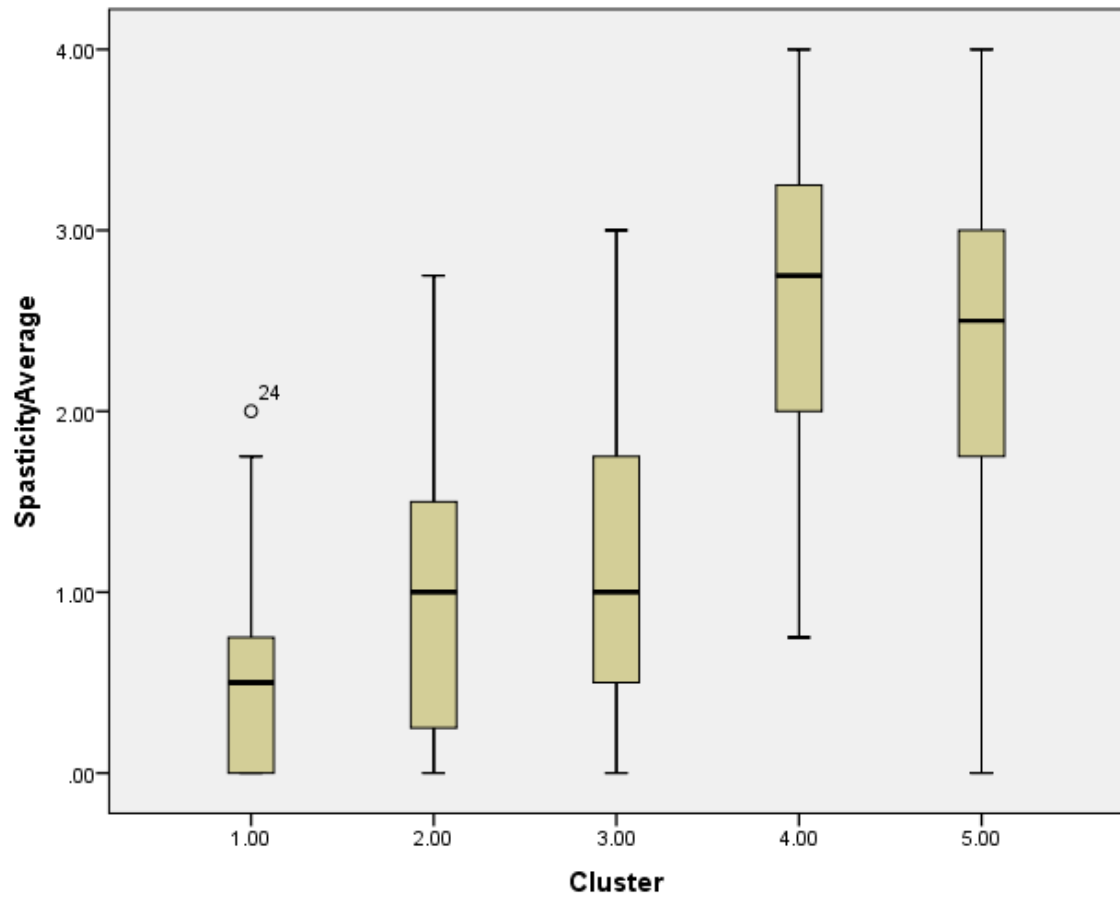
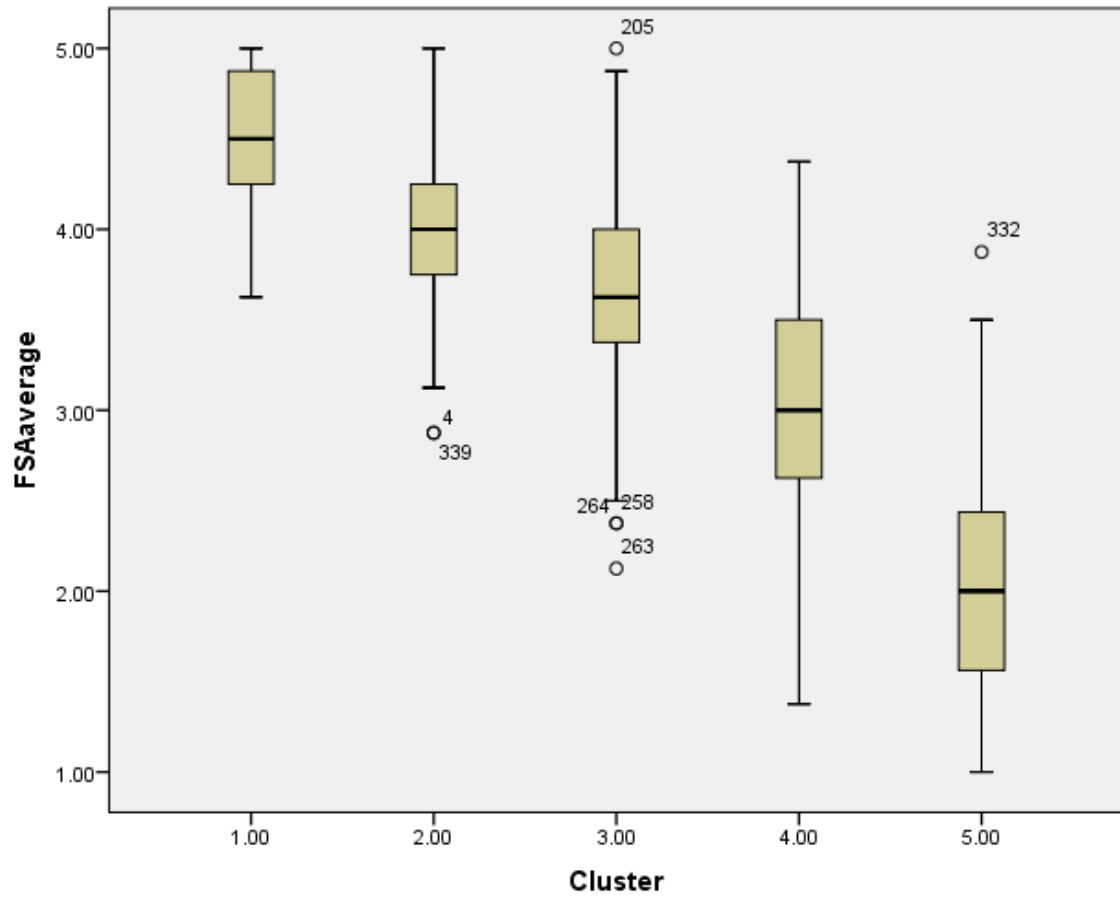
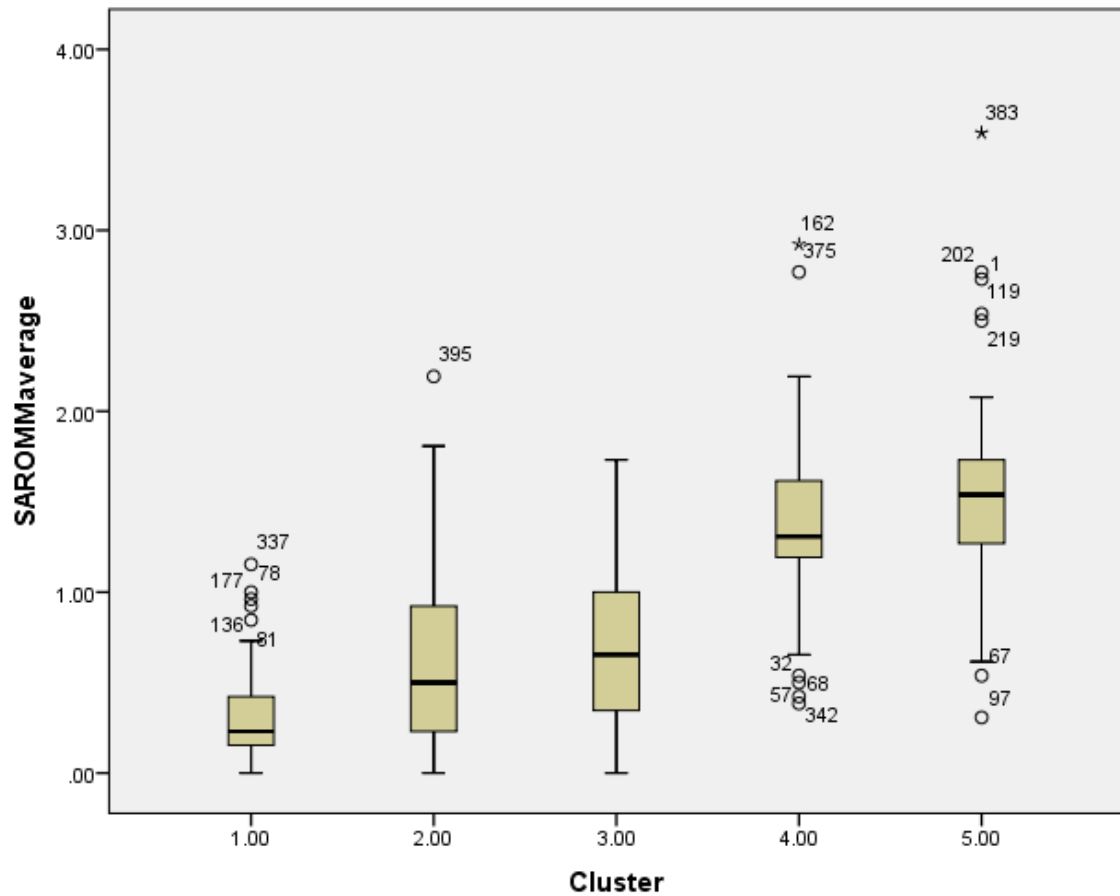


Figure 3: Boxplots for strength in cluster (Body Function Index in Cerebral Palsy - II)



FSA = Functional Strength Assessment

Figure 4: Boxplots for SAROMM in cluster (Body Function Index in Cerebral Palsy - II)



SAROMM = Spinal Alignment and Range of Motion Measure



Figure 5: Boxplots for endurance in cluster (Body Function Index in Cerebral Palsy - II)

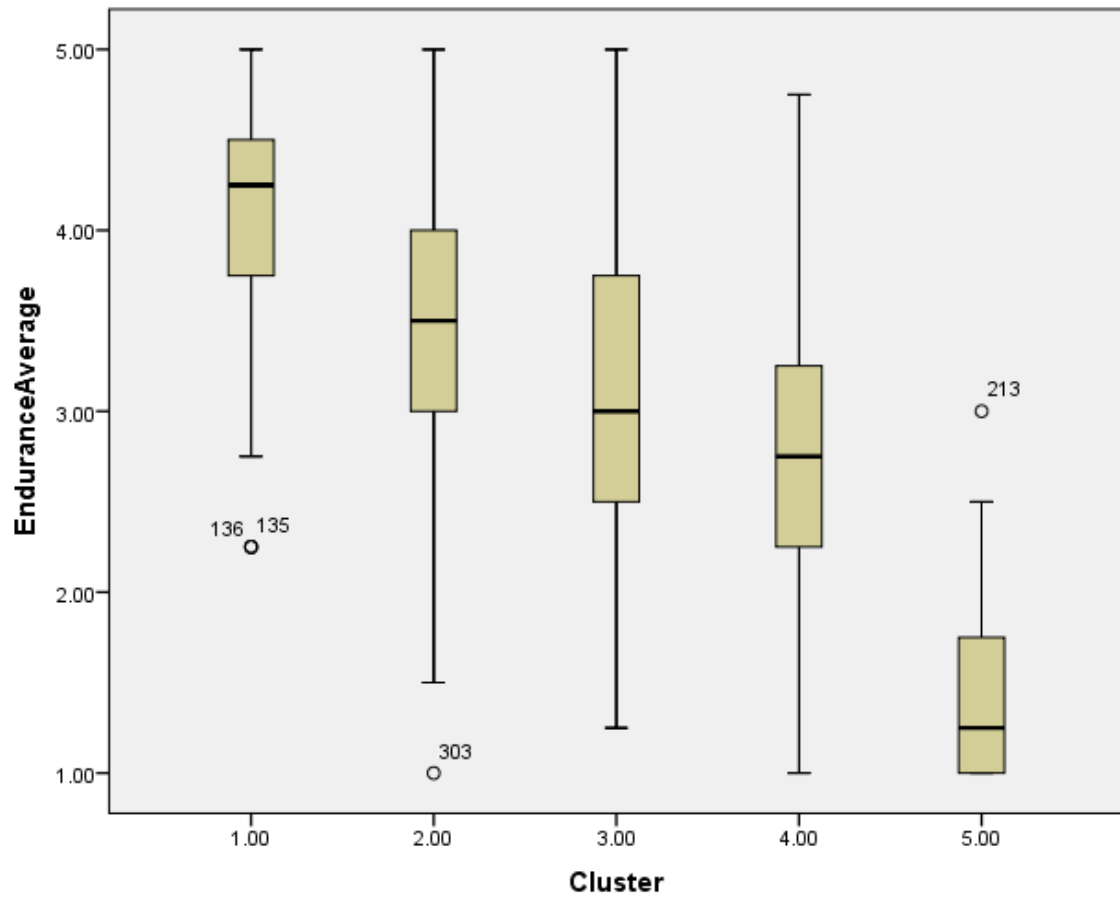
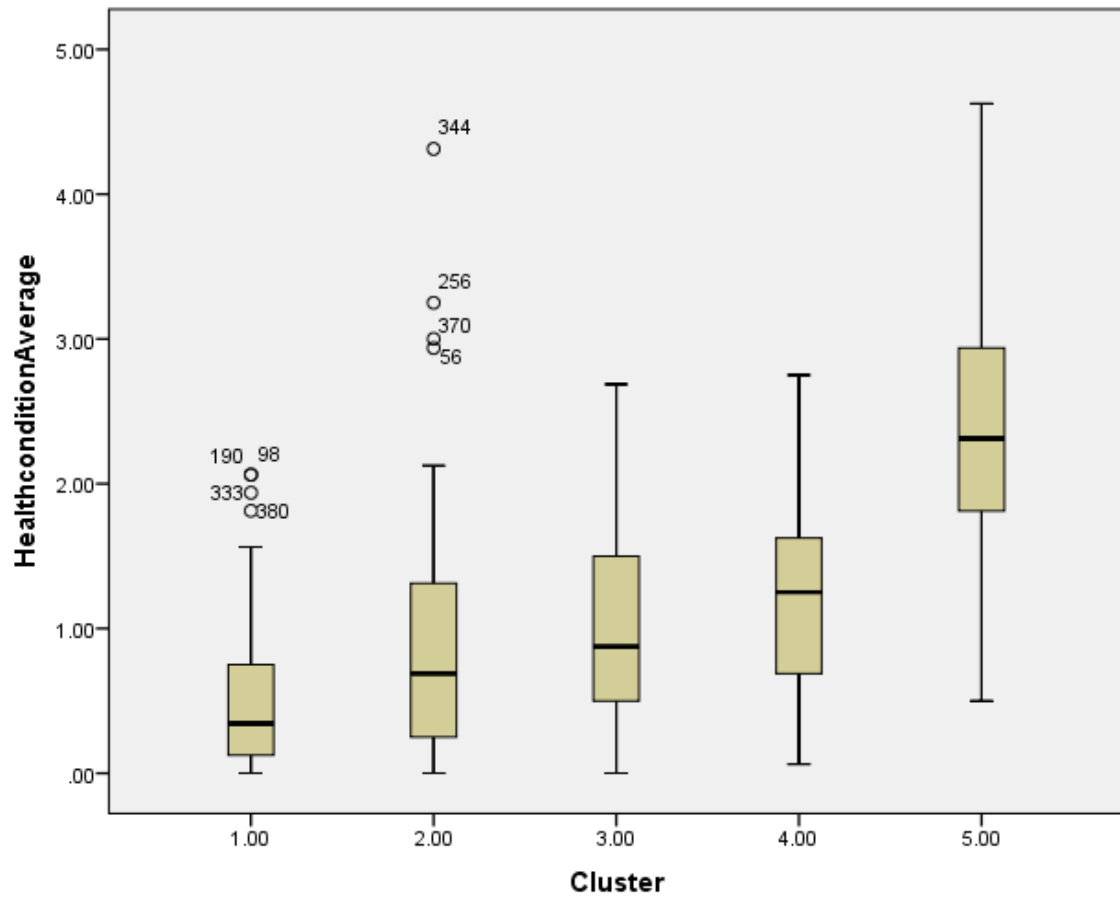


Figure 6: Boxplots for health conditions in cluster (Body Function Index in Cerebral Palsy - II)



## Appendix 4.1: Scatter plots

Figure 1: Scatter plot between quintiles (Body Function Index in Cerebral Palsy - I) and motor outcome classification based on 50% probability

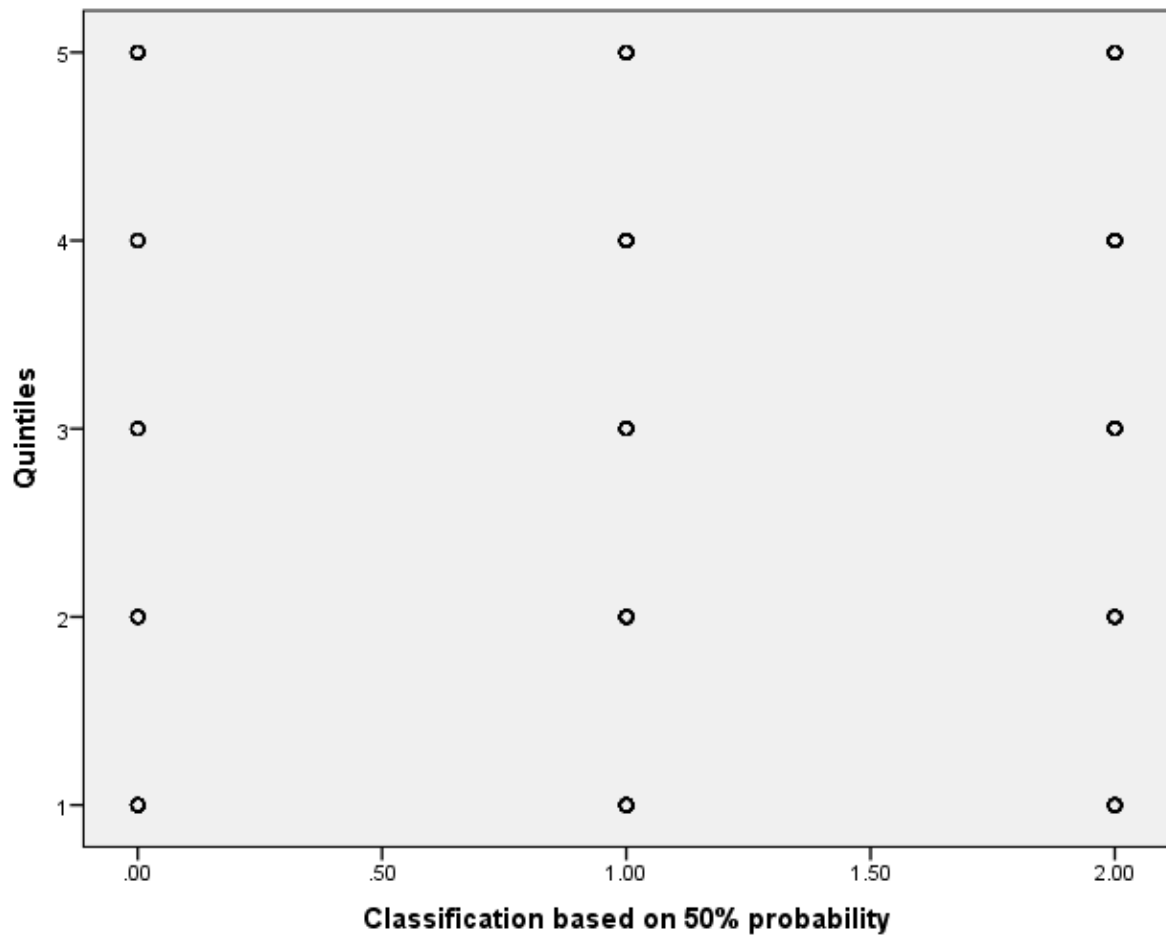


Figure 2: Scatter plot between cluster solution (Body Function Index in Cerebral Palsy - II) and motor outcome classification based on 50% probability

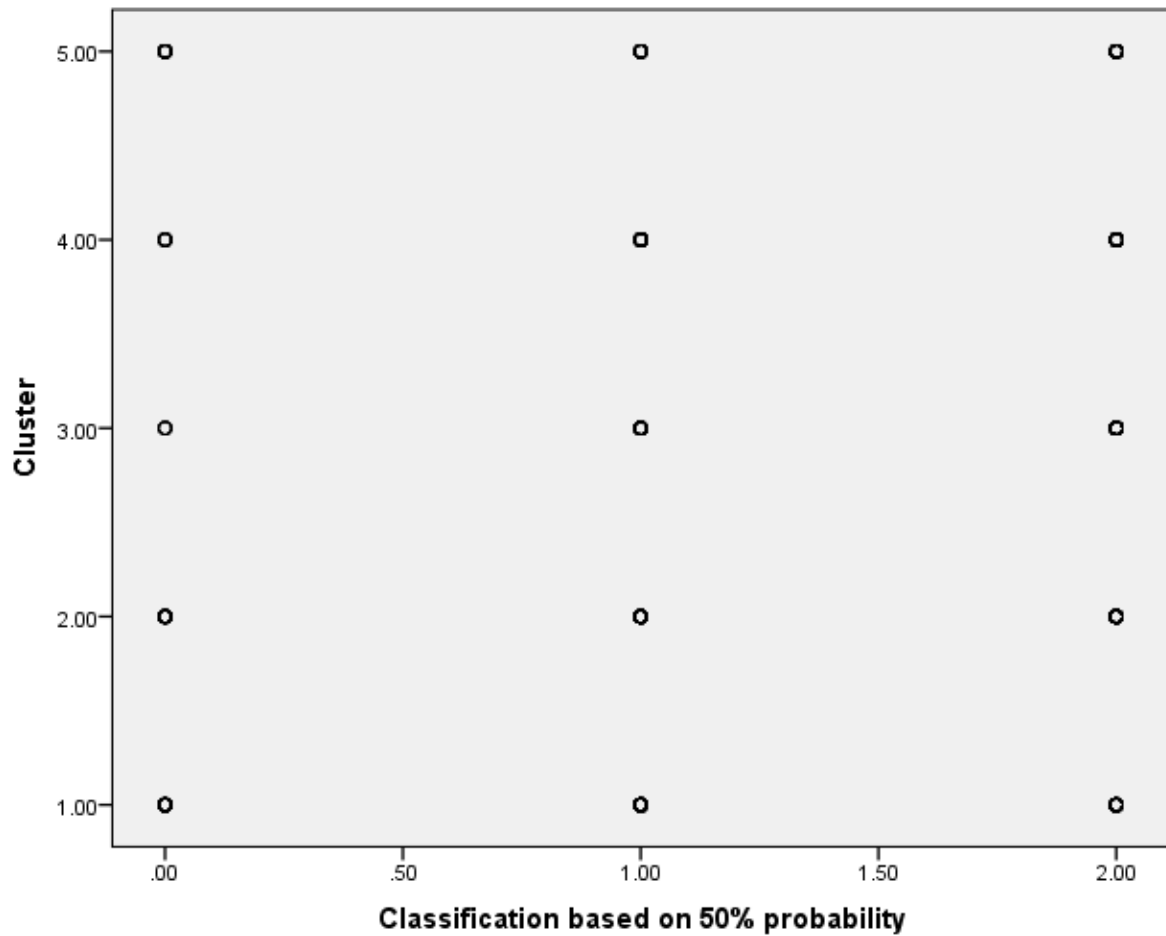
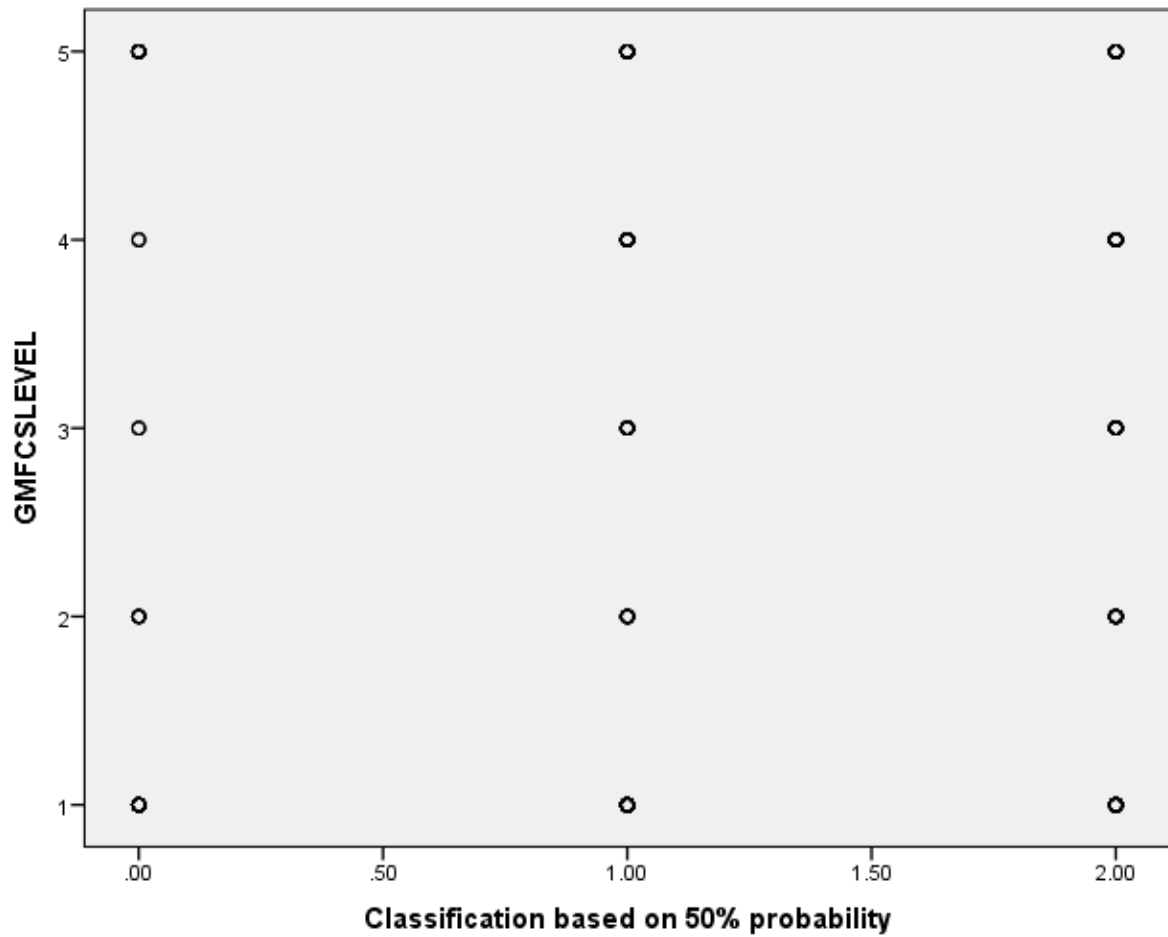


Figure 3: Scatter plot between Gross Motor Function Classification System and motor outcome classification based on 50% probability



GMFCS = Gross Motor Function Classification System

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## Curriculum Vitae

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### Peer Reviewed Publications:

1. **Jeevanantham D**, Bartlett D. Subgrouping children with cerebral palsy from a broader perspective using two methods. *Physiother Theory Pract.* Under Review. June 2016.
2. **Jeevanantham D**. Application of The International Classification Of Functioning, Disability And Health - Children And Youth In Children With Cerebral Palsy. *Indian Pediatr.* Indian Pediatr. 2016 [Epub ahead of print]
3. Rajendran V, **Jeevanantham D**. Assessment of Physical function in geriatric oncology based on International Classification of Functioning, Disability and Health (ICF) framework. *Curr Geri Rep.* 2016; 1-13.
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5. Rajendran V, **Jeevanantham D**. The Elderly Mobility Scale. *J Acute Care Phys Ther.* 2016;7:3-4.
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7. Rajendran V, Roy FG, **Jeevanantham D**. A preliminary randomized controlled study on the effectiveness of vestibular specific neuromuscular training in children with hearing impairment. *Clin Rehabil.* 2013;27: 459-67.
8. Rajendran V, Roy FG, **Jeevanantham D**. Effect of exercise intervention on vestibular related impairments in hearing-impaired children. *Alexandria J Med.* 2013;49: 7-12.

9. Rajendran V, Roy FG, **Jeevanantham D**. Postural control, motor skills, and health-related quality of life in children with hearing impairment: a systematic review. *Eur Arch Otorhinolaryngol* 2012; 269:1063-1071.
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11. **Jeevanantham D**, Pattnaik M, Mohanty PP, Rajendran V. Effect of 12 weeks weight bearing and non-weight bearing aerobic exercises on overweight and obese individuals. *Indian J Phys Occup Ther*. 2011; 5: 47-51.

#### **OTHER SCHOLARLY CONTRIBUTIONS**

1. **Jeevanantham D**. Classifying the manual abilities of young children with cerebral palsy. *Dev Med Child Neurol*. 2016 [Epub ahead of print]
2. **Jeevanantham D**. The Manual Ability Classification System: A scoping review. [http://download.lww.com/downloads/eJP/pedpt/podcasts/150621\\_Fall\\_PPT\\_Podcast\\_PRODUCTION\\_MASTER.mp3](http://download.lww.com/downloads/eJP/pedpt/podcasts/150621_Fall_PPT_Podcast_PRODUCTION_MASTER.mp3).

#### **PRESENTATIONS**

##### **POSTER PRESENTATIONS**

**Deepa Jeevanantham**, Doreen Bartlett. How to holistically subgroup children with cerebral palsy?

1. FHS Research Day, Western University, London, ON, March 2014.
2. Child Health Symposium, Joint symposium by Faculty of Health Sciences and Thames Valley Children's Centre, May 2015.

**Deepa Jeevanantham**, Emily Dyszuk, Doreen Bartlett. The Manual Ability Classification System: A scoping review.

1. Provincial: Ontario Association of Children's Rehabilitation Services, Toronto, Nov 2013.
2. Institutional: HRS Research Forum, Western University, London, ON, Feb 2014.
3. Institutional: FHS Research Day, Western University, London, ON, March 2014.

##### **ORAL PRESENTATIONS**

**Deepa Jeevanantham**, Emily Dyszuk, Doreen Bartlett. Functional classification systems in Children with Cerebral Palsy, Child Health Symposium, Joint symposium by Faculty of Health Sciences and Thames Valley Children's Centre, May 2014.

**Deepa Jeevanantham**, Doreen Bartlett. Development of Multivariate Classification Systems in Children with Cerebral Palsy. Oral presentation - Seed category - HRS Research Forum, Feb 2013.

**Deepa Jeevanantham**, Monalisa Pattnaik, Pathita Paban Mohanty. Effect of 12 weeks of weight bearing and non-weight bearing aerobic exercises on overweight and obese individuals. Indian Association of Physiotherapists, Dehradun, India, Jan 2008.

**Deepa Jeevanantham**, Monalisa Pattnaik, Pathita Paban Mohanty. Effect of aerobic exercise on sedentary female youth. Indian Association of Physiotherapists, Calcutta, India Jan 2007.