THE FUNCTIONAL MOBILITY SCALE FOR CHILDREN WITH CEREBRAL PALSY: RELIABILITY AND VALIDITY

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ABSTRACT

The purpose of this thesis was to investigate the psychometric properties and clinical utility of the Functional Mobility Scale (FMS) for children with cerebral palsy (CP). The FMS quantifies mobility according to the need for assistive devices in different environmental settings. Initially a systematic review was conducted on the psychometric properties and clinical utility of existing evaluative outcome measures that assessed activity limitation in children with CP. Good to excellent reliability was found for all tools. In contrast, the validity and responsiveness of many tools required further investigation. The FMS was the only tool to quantify activity with different assistive devices for a range of environmental settings. A key objective of this thesis was to investigate the reliability, construct, concurrent and discriminative validity, as well as the responsiveness to change of the FMS.

The inter-rater reliability of the FMS was investigated in 118 children with CP who were measured on two occasions, using 44 raters from different clinical professions. Substantial agreement was found for the three FMS subscales with unweighted kappa values of 0.66-0.71. There were no differences in agreement between raters for different age groups or different levels of severity of CP. There was no conclusive evidence of systematic bias between raters. Overall the FMS was found to be a reliable measure of activity in children with CP.

A subsequent investigation examined the responsiveness of the FMS following orthopaedic surgery and spasticity management in 84 children to determine if the FMS could show both change and stability in mobility status. This found the scale to be able to detect clinically important change following complex orthopaedic surgery. The FMS showed minimal change following botulinum toxin injections, where functional mobility was not expected to change. Further examination of responsiveness of the FMS and what constitutes a minimal clinically important difference for different interventions was considered to be warranted, given that responsiveness to clinically important change is an essential property of evaluative measures.
The construct validity of the FMS was examined in a sample of 18 children by comparing the FMS rating obtained by usual method of clinician interpretation of self-report with direct observation of the children in their home and school settings. Fair to moderate agreement between self-report and direct observation was found with kappa values of 0.27-0.45. Children with CP were observed to use a variety of mobility methods and often more than one method of mobility was used in each setting. The study highlighted the need for further investigation with larger studies of this important clinical issue of whether self-report tools measure performance.

The concurrent and discriminative validity of the FMS were investigated in 172 children with CP to determine if the FMS measures activity limitation and can distinguish between groups of children with CP based on severity of activity limitation. Concurrent validity was supported by high correlations of the FMS with the Gillette Functional Assessment Questionnaire with Spearman’s rho of 0.69-0.79, and with the Gross Motor Function Classification System (GMFCS) with Spearman’s rho of 0.74-0.91. The FMS was able to discriminate between children who walk independently (GMFCS levels I and II) from those who require assistive devices to walk (GMFCS levels III and IV).

To conclude, the FMS has unique features not found in other measures of activity limitation used for children with CP. It enables clinicians to assess mobility according to the need for assistive devices within the different environmental settings of home, school and the wider community. It was found to be clinically useful and feasible, with sound psychometric properties. The FMS can also be used to quantify mobility in children with CP in a range of clinical settings, such as gait laboratories as well as settings with restricted access to expensive technology. This thesis provides clinicians with a robust tool for documenting mobility status at one point in time and for assessing change over time and following therapeutic interventions.
DECLARATION

This is to certify that

i. This thesis comprises only my original work towards the PhD
ii. Due acknowledgement has been made in the text to all other material used
iii. The thesis is less than 100,000 words in length, exclusive of table, maps, bibliographies and appendices
iv. The thesis reflects work done during the period of candidature
v. I have used no part of this work for the award of another degree
vi. This thesis was conducted according to the Code of Conduct for Research
vii. Research data and records collected, used and maintained in the conduct of my research will be retained and accessible for five years from the point of thesis submission unless publication or public release of the work of research subsequently occurs, in which case the research data and records will then be retained for five years after publication, or public release, of the work of research

Adrienne Harvey

February 2008
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DOCTORAL CANDIDATURE

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Conference Presentations


**Seminar Presentations**


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National Health and Medical Research Council Postgraduate Scholarship (co-funded by the CP Institute) - 2007
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<td>AF</td>
<td>Adrienne Fosang</td>
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<tr>
<td>AIMS</td>
<td>Alberta Infant Motor Scale</td>
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<td>ANOVA</td>
<td>analysis of variance</td>
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<td>ASK</td>
<td>Activities Scale for Kids</td>
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<td>CAPE</td>
<td>Children’s Assessment of Participation and Enjoyment</td>
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<td>CHQ</td>
<td>Child Health Questionnaire</td>
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<tr>
<td>CHQ-CF</td>
<td>Child Health Questionnaire child form</td>
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<tr>
<td>CHQ-PF</td>
<td>Child Health Questionnaire parent form</td>
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<tr>
<td>CI</td>
<td>confidence interval</td>
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<tr>
<td>CONSORT</td>
<td>consolidated standards of reporting trials</td>
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<td>COPM</td>
<td>Canadian Occupational Performance Measure</td>
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<tr>
<td>CP</td>
<td>cerebral palsy</td>
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<td>EE</td>
<td>energy expenditure</td>
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<td>ES</td>
<td>effect size</td>
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<td>FMAS</td>
<td>Functional Motor Assessment Scale</td>
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<td>FMS</td>
<td>Functional Mobility Scale</td>
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<td>GAS</td>
<td>Goal Attainment Scale</td>
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<td>GMFCS</td>
<td>Gross Motor Function Classification System</td>
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<td>GMFM</td>
<td>Gross Motor Function Measure</td>
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<td>GMFM-66</td>
<td>Gross Motor Function Measure – 66 item version</td>
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<tr>
<td>GMFM-88</td>
<td>Gross Motor Function Measure – 88 item version</td>
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<tr>
<td>HRQOL</td>
<td>health related quality of life</td>
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<td>IC</td>
<td>internal consistency</td>
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<td>ICC</td>
<td>intraclass correlation coefficient</td>
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<tr>
<td>ICF</td>
<td>International Classification of Functioning, Disability and Health</td>
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<tr>
<td>JH</td>
<td>Janet Hough</td>
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<tr>
<td>JR</td>
<td>Jonathan Robin</td>
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<tr>
<td>K</td>
<td>kappa</td>
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<tr>
<td>MCID</td>
<td>minimal clinically important difference</td>
</tr>
<tr>
<td>MH</td>
<td>Marty Hughes</td>
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<tr>
<td>MM</td>
<td>Meg Morris</td>
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<tr>
<td>Abbreviation</td>
<td>Full Form</td>
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<td>--------------</td>
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<tr>
<td>MOOSE</td>
<td>meta-analysis of observational studies</td>
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<td>OR</td>
<td>odds ratios</td>
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<tr>
<td>PDMS</td>
<td>Peabody Developmental Motor Scale</td>
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<td>PEDI</td>
<td>Pediatric Evaluation of Disability Inventory</td>
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<td>PedsQL</td>
<td>Pediatric Quality of Life Inventory</td>
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<tr>
<td>PODCI</td>
<td>Pediatric Outcomes Data Collection Instrument</td>
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<tr>
<td>POSNA</td>
<td>Pediatric Orthopaedic Society of North America</td>
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<tr>
<td>QOL</td>
<td>quality of life</td>
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<tr>
<td>QUOROM</td>
<td>quality of reporting of meta-analyses</td>
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<tr>
<td>RCT</td>
<td>randomised controlled trial</td>
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<td>ROC</td>
<td>receiver operating characteristic</td>
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<tr>
<td>ROM</td>
<td>range of movement</td>
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<tr>
<td>SD</td>
<td>standard deviation</td>
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<tr>
<td>SDR</td>
<td>selective dorsal rhizotomy</td>
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<tr>
<td>SEMLS</td>
<td>single event multilevel surgery</td>
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<tr>
<td>SRM</td>
<td>standardized response means</td>
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<tr>
<td>STARD</td>
<td>standards for reporting of diagnostic accuracy</td>
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<tr>
<td>TUG</td>
<td>Timed Up and Go</td>
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<td>VDRO</td>
<td>varus derotation osteotomy</td>
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<tr>
<td>WeeFIM</td>
<td>Functional Independence Measure for Children</td>
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<tr>
<td>WHO</td>
<td>World Health Organisation</td>
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<tr>
<td>3DGA</td>
<td>three dimensional gait analysis</td>
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CHAPTER ONE: INTRODUCTION

1.1. The problem: mobility in children with cerebral palsy and how to measure it

This thesis investigates the psychometric properties and clinical utility of a recently developed scale, the Functional Mobility Scale (FMS) (Graham et al 2004). The FMS is an outcome measure used by physiotherapists and other health professionals. It rates mobility in children with cerebral palsy (CP) in different environmental settings according to their need for assistive devices. Although a range of modalities can be employed for mobility, the FMS focuses on walking. The mobility of children with CP can be limited by the physical manifestations resulting from the positive and negative features of the upper motor neurone syndrome, including spasticity, weakness, loss of selective motor control and deficits in balance and coordination (Mayer and Esquenazi 2003). The progressive neuromusculoskeletal pathology that occurs can include the development of contractures and torsion of long bones (Graham et al 2004). This can further limit mobility of children with CP and result in the need for assistive devices to enable them to move around at home, at school and in the community.

To be mobile is “the freedom to move from place to place” (Oxford English Dictionary 1994). This is synonymous with being movable, portable or transportable. “Mobility” falls into the ‘activities and participation’ component of the International Classification of Functioning, Disability and Health (ICF) (World Health Organisation 2001) that is explained further in Chapter Two. For children with CP, mobility enables movement from place to place, participation with their family and peers and the ability to function optimally in everyday life. The heterogeneity of the population of children with CP means that there is the potential for a very diverse and complex range of mobility states. For example, some children may have difficulty with balance and coordination on uneven surfaces as they walk from one place to another. Others who have more severe motor impairment may require the use of assistive devices to mobilize, ranging from crutches and walkers through to wheelchairs.
Cerebral palsy is the most common cause of physical disability affecting children in developing countries with an incidence of 2.0-2.5 per 1000 live births (Stanley et al 2000). Physiotherapists and other clinicians require tools to accurately measure and record changes in mobility across the spectrum of mobility disorders observed in children with CP. Chapter Three of this thesis will systematically examine the psychometric properties and clinical utility of the available evaluative outcome measures that assess activity limitations in children with CP. It will highlight that very few tools exist that measure mobility in isolation and that many measure it within the context of a larger inventory. It will also show that the FMS is the only tool to consider the different assistive devices used by children with CP within the different environmental settings of the home, school and wider community. The rationale behind the development of the FMS will be explored in Chapter Four.

A preliminary study examined some aspects of reliability and validity of the FMS (Graham et al 2004). Chapter Three will show that before the FMS can be considered to be a psychometrically sound outcome measure to quantify mobility in children with CP, further examination of its psychometric properties is required. As well as ensuring that the FMS is based on a valid theoretical construct, Chapters Five to Eight of the thesis investigate the inter-rater reliability and concurrent, construct, and discriminative validity and responsiveness to change of the scale. The extent to which the FMS can be used with confidence as a performance measure of mobility for children with CP is explored. The ultimate aim is to have a psychometrically sound and clinically useful measurement tool that quantifies mobility in children with CP.

The key research questions to be addressed in the thesis are:

i. What are the psychometric properties and clinical utility of the available evaluative measures of activity limitation for children with CP?

ii. What is the inter-rater agreement of the FMS when tested by a range of clinicians from different professional backgrounds?
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iii. What is the responsiveness to change of the FMS for detecting clinically significant change following orthopaedic surgery and spasticity management?

iv. Does the information obtained from the FMS via self-report of the assistance required in the different settings of the home, school and community accurately reflect the mobility status of children with CP?

v. What is the concurrent validity of the FMS with a similar measure of mobility and classification system of activity limitation as criterion measures?

vi. Is the FMS able to discriminate the severity of activity limitation in children with CP as classified by the Gross Motor Function Classification System (GMFCS)?

1.2. Aims and hypotheses

1.2.1. Aims

In response to the key research questions, the aims of this thesis were to:

i. critically evaluate the literature on existing evaluative outcome measures that assess activity limitation by systematically appraising the design of the studies and the psychometric properties and clinical utility of the measures;

ii. determine the inter-rater agreement of the FMS in children with CP using a large number of clinicians as raters from different clinical professions. This includes examining the agreement for different age groups and severity of CP of the children and the existence of any systematic bias of raters;

iii. examine the ability of the FMS to detect minimal clinically important differences in mobility status following orthopaedic surgery and spasticity management;

iv. examine one aspect of construct validity of the FMS by comparing FMS ratings performed by usual administration method with direct observation at home and school;
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v. determine the concurrent validity of the FMS by correlating it with the walking scale of the Gillette Functional Assessment Questionnaire (FAQ) and the GMFCS;

vi. examine the discriminative validity of the FMS by determining if it can discriminate severity of activity limitation of children with CP as classified by the GMFCS.

1.2.2. Hypotheses

The hypotheses were:

i. The clinical utility of the available evaluative measures used in children with CP that assess activity limitation will vary according to their proven psychometric properties, the dimensions of activity they measure and their purpose for application in CP.

ii. There will be a high level of inter-rater agreement for the FMS in children with CP using a large group of raters from various clinical backgrounds. The level of agreement will be different for children of different age groups and severity of CP.

iii. The FMS will be able to detect clinically important differences in functional mobility in children with CP where change in mobility occurs; it will show change in children having orthopaedic surgery and will show stability in children having botulinum toxin injections.

iv. The mobility status of children with CP observed within the environmental settings of home and school will have substantial agreement with the self-report ratings of the FMS.

v. The FMS will have a strong positive relationship with the FAQ and a strong negative relationship with GMFCS levels.

vi. The FMS will discriminate severity of activity limitation in children with CP using the GMFCS for children classified as GMFCS levels I-IV.
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| Chapter 1 | Introduction: the research question, synopsis, aims and significance of the studies in the thesis | There is a need for psychometrically robust outcome measurement tools to assess mobility in children with CP to document status, particularly after interventions |
| Chapter 2 | Outcome measurement for children with CP | Description and classification of CP within the ICF. Purpose and requirements for outcome measurement in CP. |
| Chapter 3 | Study 1: Systematic review | Critical appraisal of literature on evaluative activity limitation outcome measures used for children with CP; psychometrics, feasibility and clinical utility |
| Chapter 4 | Development of the FMS and Study 2: pilot study | Original development of the FMS and ability of the FMS to detect change after multilevel surgery |
| Chapter 5 | Study 3: Reliability of the FMS | Inter-rater reliability of the FMS using a range of clinicians |
| Chapter 6 | Study 4: Responsiveness to change of the FMS | Responsiveness to change of the FMS for children undergoing orthopaedic surgery and spasticity management |
| Chapter 7 | Study 5: Construct validity of the FMS | Variety of mobility methods used and agreement between self-report FMS and direct observation of children in their own environments |
| Chapter 8 | Study 6: Discriminative and concurrent validity of the FMS | Concurrent validity of the FMS by correlating it with a similar outcome measure and ability of the FMS to discriminate severity of CP as described by GMFCS |
| Chapter 9 | Grand discussion | Synthesis of findings, clinical implications, limitations, future directions and conclusions |

Figure 1.1 Concept map of thesis structure
1.3. Synopsis

The overall structure and conceptual basis of this thesis is summarised in the concept map shown in Figure 1.1. It shows that the concepts of measurement and the systematic review of activity limitation measures set the framework for the remaining studies exploring the psychometric properties of the FMS. Study 2 is a retrospective pilot study. Three separate samples of children were recruited prospectively for studies 3-6. The association between each of these samples with respect to the various studies is illustrated in Figure 1.2. This demonstrates that sample 2 and 3 were combined for data analysis in Study 6.

![Concept Map]

Sample 1
Retrospective sample of 66 children with CP

Study 2
Pilot study: change after surgery

Sample 2
118 children with CP

Study 3
Reliability

Sample 3
96 children with CP

Study 6
Concurrent and discriminative validity

Sample 4
18 children with CP recruited from one special school

Study 4
Responsiveness to change

Sample 1
Retrospective sample of 66 children with CP

Study 2
Pilot study: change after surgery

Figure 1.2 Samples recruited for overall thesis
1.4. Rationale of the thesis

The rationale for each study is outlined below.

1.4.1. Study 1: Systematic review of the activity limitation outcome measures used in children with cerebral palsy

Chapter Three presents the results from a systematic review of the evaluative activity limitation measures used in children with CP. It critically examines the psychometric properties of the available tools as well as their feasibility and clinical utility. A thorough understanding of the reliability, validity and responsiveness of existing tools and the dimensions of activity measured was considered to be a necessary prerequisite before further investigation of the validity of the FMS was implemented. The review was conducted with a focused literature search strategy and a precise review protocol for study selection, data extraction and quality assessment. The methodology utilised was based on guidelines for the scientific process behind conducting a systematic review. This allowed for decisions on the clinical utility of the available measures to be made based on the best possible evidence (Cook et al 1997, Verhagen et al 2001, Herbert et al 2005). The results from this review facilitated the structuring for the remaining studies investigating the FMS. This study has been accepted for publication in Developmental Medicine and Child Neurology.

1.4.2. Study 2: Development of the FMS and pilot study

The aim of Chapter Four is to provide the background to the initial development of the FMS. It includes the rationale of why the scale was developed, the target population and how it was developed. The administration method of the FMS and the clinical feasibility of the scale are also described. This chapter reports an initial pilot study of the FMS investigating its ability to detect change following single event multilevel surgery in children with CP, a population for which the scale was developed. This study has been published in Developmental Medicine and Child Neurology (2007) 49:603-607. A copy of the FMS is provided in Appendix A and a brochure is located inside the back cover of this thesis.
1.4.3. Study 3: Reliability of the Functional Mobility Scale

Chapter Five reports the results from an investigation into the inter-rater reliability of the FMS. The FMS was developed for use by clinicians from different professional backgrounds to assess mobility in children with CP. Inter-rater reliability is critical in the population of children with CP because they will be assessed by many different clinicians over their lifetime. It is imperative that consistency between clinicians be established before the FMS can be used reliably. Not only does this imply consistency between large numbers of clinicians, it also indicates a need to ensure consistency between clinicians from different professional backgrounds and settings. This study examines the inter-rater reliability using a large number of raters as clinicians and determines the agreement between clinicians from the hospital setting as well as those in the community setting. It also examines agreement using physiotherapists and orthopaedic surgeons. If agreement between raters was found to be high then the FMS could be considered a reliable measure of mobility for the population examined in this study.

1.4.4. Study 4: Responsiveness to change of the FMS

Chapter Six examines whether the FMS is able to detect clinically important change, that is, the responsiveness to change of the scale. Evaluative measures are required to have proven responsiveness to change as a prerequisite measurement property before they can be used confidently as outcome measures (Kirschner and Guyatt 1985). The FMS was developed to assess mobility and, in particular, to evaluate change in mobility following interventions such as orthopaedic surgery. It is therefore essential that the ability of the FMS to detect clinically important change in the population of children with CP undergoing these types of interventions is investigated. As well as detecting change, it is also important that the FMS can show stability where no change has occurred. This study examines change in mobility in children following single event multilevel surgery and Botulinum toxin injections. Change in mobility status from the pre-intervention baseline are analysed at regular post-intervention periods to determine if the FMS is able to detect change occurring. A comparison of change in mobility following SEMLS and Botulinum toxin injections are also examined. If the FMS is found to detect clinically important
change, it would support the use of the tool in children with CP to monitor change over time following such interventions.

1.4.5. Study 5: Construct validity of the FMS

Chapter Seven examines one aspect of construct validity of the FMS by comparing FMS ratings obtained by usual administration method with direct observation of the children at their schools and within their homes. The FMS is administered by self report from the child or parents. Like other self-report measures, there is an assumption that what is reported accurately reflects true performance. One way to ascertain this is to compare self-report with direct observation of performance. This is examined by comparing the clinical FMS ratings of a group of children with direct observation of their mobility methods at their school and home. This study was considered both a necessary and interesting component of this thesis to investigate the validity of the FMS. It was also considered to be a key concept to examine for the field of measurement in CP because the issue of whether any self report measure reflects performance has not been adequately investigated.

1.4.6. Study 6: Discriminative and concurrent validity of the FMS

Chapter Eight investigates the concurrent and discriminative validity of the FMS. It examines the correlation of the FMS with another measure of mobility, the Gillette Functional Assessment Questionnaire (FAQ), as well as the correlation of the FMS and the GMFCS, a descriptor of gross motor function. This was considered important to examine to ensure the FMS measures activity and physical function, yet is not so similar to existing measures that its use is not warranted. Discriminative validity was considered another factor to support the overall validity of the FMS. The scale’s ability to discriminate severity of motor impairment of CP as determined by the GMFCS level of the children was examined using the combined sample for Study 2 and Study 3. The results of these two investigations will provide further evidence to support the validity of the FMS as a measurement tool for children with CP.
1.5. Significance of the research

The results of this investigation have the potential to positively impact children with CP and their families, physiotherapists and other clinicians who work with children with CP, health care providers and the general community. Physiotherapists and other clinicians require valid outcome measures that quickly and easily measure mobility in children with CP. Not only do they have to be available and show clinical utility, they also have to be reliable and valid (Law et al 1999). Mobility is a key component of activity that enables children with CP to negotiate the relevant environments of home, school and community settings in order to participate in society with their family, peers and members of the community. This research aims to provide a reliable and valid tool to measure mobility for use by physiotherapists and other clinicians who manage children with CP. The FMS will then be available and suitable for use in both clinical and research settings.

The unique aspect of the FMS is its ability to quantify the variety of different assistive devices that children use in a range of environments. Within the clinical setting, it has the potential to document mobility status of children with CP at one point in time and to assess change in mobility status over time. The information obtained could be used to track improvement or deterioration in a child’s mobility over time and to help guide decision-making for further intervention or therapy. It also has a potential role in measuring change in mobility following costly interventions such as orthopaedic surgery and spasticity management. These interventions are implemented with the aim of maintaining or improving functional abilities, such as mobility, in children with CP (Graham and Harvey 2007). It is therefore essential that physiotherapists and other clinicians can measure and document changes in mobility following treatments. It can also be important to document both improvement and deterioration (Harvey et al 2007). By measuring the changes in mobility associated with interventions we can better gauge their effectiveness and improve decision-making for intervention strategies. This could benefit children and their families by ensuring that they receive the best possible care and obtain the optimal outcome. This in turn could benefit health care providers and the
general community with more targeted treatment and improved overall health of children with CP.

Within the research setting, the FMS could become a key outcome measure for epidemiological studies examining change in function in children with CP over time, longitudinal studies investigating changes in mobility throughout childhood into adulthood and trials into the effects of interventions. Because the FMS is specific to mobility, it will provide information on activity levels. It could be used in conjunction with other valid and reliable outcome measures that focus on different domains of disability and health, such as body structures and function and participation measures. This could enable a comprehensive picture of the health status of children with CP to be obtained for research and clinical purposes. Using the FMS within the research setting could add to the current body of knowledge of activity patterns in children with CP, particularly with respect to treatments. The results also have the potential to advance current clinical management of children with CP by tailoring treatment to ensure optimal mobility results from costly interventions, ensuring overall better health care. Optimal mobility allows children to participate fully within their relevant environments (Palisano et al 2003). By accurately measuring mobility with tools such as the FMS, and by ensuring our interventions are optimizing mobility, the participation of these children will also be optimized.

The unique features of the FMS with its focus on mobility and assistance required warrant its development and validation. The FMS is clinically feasible and can be used in many clinical settings, including those with limited resources. It requires no equipment and minimal training for administration and is not time consuming to complete. It therefore has the potential to be very useful clinically. Before it is utilized more widely, the reliability and validity of the FMS need to be thoroughly investigated to ensure that it is psychometrically sound. This thesis examines these psychometric properties to determine whether the FMS can be utilized more widely.
CHAPTER TWO: OUTCOME MEASUREMENT FOR CHILDREN WITH CEREBRAL PALSY

This chapter will explore CP within the context of the International Classification of Functioning, Disability and Health (ICF), with a focus on outcome measurement. Cerebral palsy will be introduced briefly, followed by discussion of how the clinical manifestations can be described, classified and measured with consideration of the ICF domains. Most emphasis will be directed towards activity limitation because this is the domain of the ICF that the FMS quantifies. This background will provide the basis for the systematic review on activity limitation outcome measures reported in Chapter Three. The concepts of capability and performance and the influence of the environment on activity will also be explored because these are key factors underpinning the development of the FMS (see Chapter Four). The purposes and prerequisites for outcome measurement using tools such as the FMS will be examined for children with CP. This will provide a background against which the results of the psychometric testing of the FMS can be interpreted.

2.1. Cerebral palsy: definition and incidence

The FMS was developed to quantify mobility in children with CP. Cerebral palsy is the most common cause of physical disability affecting children in developed countries with an incidence of 2.0-2.5 per 1000 live births (Stanley et al 2000). The classic definition of CP is “a disorder of movement and posture due to a defect or lesion of the immature brain” (Bax 1964). This definition was modified in 1992 to encapsulate the heterogeneity of the disorders covered by the term CP to; “an umbrella term covering a group of non-progressive, but often changing, motor impairment syndromes secondary to lesions or anomalies of the brain arising in the early stages of development” (Mutch et al 1992).

The definition of CP has been revised recently by an executive committee for a report on the Definition and Classification of Cerebral Palsy, April 2006, to incorporate concepts developed by the ICF (Rosenbaum et al 2007). The proposed definition is;
‘Cerebral palsy 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’ (Rosenbaum et al 2007).

2.2. Clinical presentation of cerebral palsy within the International Classification of Functioning, Disability and Health (ICF)

Cerebral palsy incorporates a wide variety of clinical presentations and the term “cerebral palsies” is now often used to indicate the diversity and heterogeneity of the disorder (Miller and Clark 1998, Stanley et al 2000). The ICF (World Health Organisation 2001) is considered in this section because it is a framework that can be utilised to describe the clinical manifestations of CP. The framework and how CP presents clinically within it are outlined below. Outcome measurement for children with CP can be tailored around the different domains of the ICF as will be presented in section 2.5.3. The focus of this thesis is mobility of children with CP as measured by the FMS. This corresponds to the activities and participation domain of the ICF. For this thesis, the ICF was used to assist with defining the features of CP and external influences that the FMS aims to quantify. It also enabled structuring the critical evaluation of available outcome measures used for children with CP.

2.2.1. What is the ICF

The ICF aims to provide a unified and standard language and framework for the description of health and health-related states (World Health Organisation 2001). It incorporates biological and social perspectives of disablement to represent more fully the impact of health conditions, such as CP, on an individual’s life, including participation in society (Beckung and Hagberg 2002). The domains represented in the ICF are described from the perspective of the body, the individual and society for body functions and structures, and activities and participation (World Health Organisation 2001). They are
described along with the components of contextual factors of environmental and personal factors which may impact a person’s health state (World Health Organisation 2001). An individual’s functioning in a specific domain is an interaction or complex relationship between the health condition and contextual factors, as shown in the ICF model in Figure 2.1.

![Figure 2.1 Interactions between the components of the ICF (WHO 2001)](image)

Having a unified language to describe health conditions, such as CP, and the influence the various domains play within the individual, can enhance communication between different clinicians involved in the care of people with such conditions (Goldstein et al 2004). Applications of the ICF include; ‘as a statistical tool to collect and record data, as a research tool to measure outcomes, quality of life or environmental factors, as a clinical tool to match treatments with specific conditions and outcome evaluation, as a social policy tool and as an educational tool’ (World Health Organisation 2001).

The advantage of the framework provided by the ICF is it provides a useful tool for communication between clinicians and for describing CP and to match outcome evaluation and treatment interventions to particular aspects of the condition. A limitation of using the ICF is that it does not consider quality of life as perceived by individuals with CP or their caregivers. It also does not consider the financial burden of health conditions such as CP. The Clinical Value Compass (Nelson et al 1996) is another framework for measuring and improving health care. It is based on concepts of measuring outcomes in CP.
Chapter 2 – Outcome Measurement in CP

(Goldberg 1991). It recognizes 4 domains; functional health status, costs, clinical outcomes and satisfaction. Although the compass does include costs, the ICF is more commonly used for describing health conditions, including CP, and is the framework used as the model for conceptualizing activity in this thesis.

2.2.2. Cerebral palsy within body functions and structure

Within the ICF, body functions are the physiological functions of body systems and body structures are anatomical parts of the body such as organs, limbs and their components (World Health Organisation 2001). Impairments are problems in body function or structure as a significant deviation or loss (World Health Organisation 2001). The primary injury in CP is the brain lesion resulting in an upper motor neurone lesion which is considered to have a number of positive and negative features (Graham and Selber 2003, Mayer and Esquenazi 2003). The positive features include spasticity, hyper-reflexia and co-contraction and the negative features include weakness, loss of selective motor control and deficits in balance and coordination (Gage and Novacheck 2001, Mayer and Esquenazi 2003). The interaction between spasticity and weakness leads to both neural and mechanical changes in muscle and progressive musculoskeletal pathology (Bache et al 2003). The changes in muscle length and structure that occur in the muscles and bones of the extremities are therefore secondary to the central nervous system lesion (Gage and Novacheck 2001). Overall, whilst the underlying brain lesion in CP is static, the musculoskeletal manifestations are progressive (Bache et al 2003).

The upper motor neurone lesion and the progressive musculoskeletal pathology in children with CP may produce gait deficits. Gait deficits in children with CP can be considered as primary, secondary or tertiary (Davids et al 2003). They are primary when they relate directly to the underlying disorder of the central nervous system, for example spasticity. Secondary deficits occur as a consequence of growth and development of the musculoskeletal system and are usually progressive over time, for example skeletal deformities and muscle contractures (Davids et al 2004). Tertiary coping responses are used to counteract the effects of primary and secondary deviations and can be compensatory (Bache et al 2003). Consideration and understanding of
progressive musculoskeletal changes are critical to the prevention and management of impairments and disability in children with CP (Graham 2006). The musculoskeletal pathology and resulting gait deficits can impact on activities, including mobility. This can in turn affect the children’s participation as will be described in the following section. Tools such as the FMS are required to measure the impact of pathology on activity and participation in children with CP.

2.2.3. Cerebral palsy within activities and participation

Within the ICF, activity is the execution of a task or action by an individual and participation is involvement in a life situation (World Health Organisation 2001). Activity limitations are difficulties an individual may have in executing activities and participation restrictions are problems an individual may experience in involvement in life situations (World Health Organisation 2001). In separating the two, activity limitation can be thought of as difficulty at the person level and participation restriction at the societal level (Schneidert et al 2003).

The FMS quantifies mobility and this can be classified within the activities and participation component of the ICF. Mobility involves “moving by changing body position or location or by transferring from one place to another, by carrying, moving or manipulating objects, by walking, running or climbing, and by using various forms of transportation” (World Health Organisation 2001). The FMS focuses on walking or wheelchair mobility in order for the child to get from one place to another. How well children mobilise can be associated with their levels of participation with their peers and family. Reduced mobility can limit their participation. Activities and participation therefore, is a key area of focus for outcome assessment and planning of interventions for children with CP.

The influence of mobility and physical function on activities and participation was examined in a study using family assessed instruments in 129 children with CP aged 6-12 years (Morris et al 2006). It reported that physical independence and mobility had the most influence on children’s activities and participation,
compared to clinical or economic burden and social integration. This finding is not surprising given that mobility is closely related to activities and participation, as found by the study. Another study examined participation levels in 427 children with physical limitations aged 6-14 years using the Children’s Assessment of Participation and Enjoyment (CAPE) (Law et al 2006). Children displayed a broad range of diversity and intensity of participation with greater involvement in informal activities than formal activities. Informal activities occur more spontaneously with less planning whereas formal activities are more planned and tend to involve organised sports. Children older than 12 years had lower overall participation levels than younger children. Children with physical disabilities participated in an average of only three out of 10 active physical activities. This descriptive study provided important information showing reduced activity levels of children with CP, particularly for older children. It also provided evidence that children with CP tend to participate in formal activities less often.

Other studies have also shown that activity and participation levels in children with CP are influenced by the severity of motor impairment, with more reduced activity and participation levels in those children with greater motor impairment (Beckung and Hagberg 2002, Schenker et al 2005). All of these studies suggest that children with CP who have limited mobility participate less in physical activities that might be important for both their physical and general well-being. The effectiveness of interventions and treatments for children with CP that aim to improve mobility and activity require examination. There is a need for reliable and valid measures to assess the effects of such interventions on activity and participation in children. This is the focus of the systematic review reported in Chapter Three.

2.2.4. Environmental factors and cerebral palsy

This section of the thesis discusses how mobility and activity in children with CP are influenced by environmental factors. When measuring activity, these environmental factors require consideration. These concepts were instrumental in the development of the FMS reported in Chapter Four.
Environmental factors are external to the individual and make up the physical, social and attitudinal environment in which people conduct their lives (World Health Organisation 2001). Awareness of the dynamic between the child and surroundings and how this affects performance is important when considering the range of interventions possible to increase participation in the home, school and community (Goldstein et al 2004). Children’s environments change across the stages of infancy, early childhood, middle childhood and adolescence and these changes can influence the child’s interactions (Simeonsson et al 2003). Outcome assessment for activity and participation in children with CP therefore needs to consider the environments that are relevant to the age group of the child being measured.

Participation in children with CP can be influenced by where they live (Hammal et al 2004) and by the socio-economic status of the family (Law et al 2006). Physical aspects of the environment that might affect mobility include accessibility, surfaces, obstacles, distances as well as time constraints. A child is more familiar with their home environment, which is likely to be more constant, than the community setting which also has longer distances to adapt to (Palisano et al 2003). The effect of the environmental setting on usual mobility methods in children with CP has been investigated (Palisano et al 2003, Tieman et al 2004). Both of these studies asked parents to report the usual methods their children used for mobility in the settings of home, school and the wider community.

The first study examined methods in a stratified sample of 636 children with CP aged 2-12 years, GMFCS levels I-IV (Palisano et al 2003). This found that children used a range of different methods in different settings, particularly those children classified GMFCS level II-IV. Children were more dependent on adult assistance outdoors and in the community compared with at school and less dependent at home. Children younger than 3 years were generally more dependent on adult assistance for mobility than those aged 4-12 years. The second study examined changes in methods over time and across settings in a smaller group of 62 children aged 6-14 years who were classified as GMFCS level II-IV (Tieman et al 2004). This group was studied because they had previously shown to display more variability in their mobility methods.
compared to children in levels I and V. At home the most common methods were floor mobility or walking. At school walking with a mobility aid or powered mobility was the most common method used. Wheelchairs were the most common method used in the outdoors/community. Across settings children used methods requiring less assistance at home than school and at school less assistive methods than in the community.

A limitation of these two key studies is that they did not distinguish the different types of assistive devices children with CP use, for example whether they use a walker or crutches. However they are important because they highlight the variety of mobility methods used by children with CP. They also demonstrate that mobility methods can change as a result of environmental setting. The results confirm that it is very important for clinicians to consider the environmental setting when assessing an activity such as mobility. They also emphasize the need to consider and quantify the range of mobility methods children use within each setting because the same child may use more than one method. The effect of environmental setting will also influence the mobility methods used as children grow and change over time and following treatments and interventions, such as orthopaedic surgery and spasticity management. Because the environmental setting may influence mobility methods children use, it is important that clinicians are able to measure this with reliable and valid tools. The FMS aims to do this. These have been key studies in the development of the FMS and will be explored further in Chapters Four and Seven.

2.2.5. The ICF qualifiers of capability and performance

It is important to consider whether an outcome tool is measuring capability or performance because this can influence the level of ability recorded and how that information is applied. Performance and capability are the two qualifiers within the activities and participation component of the ICF (World Health Organisation 2001). Performance describes what an individual does in his or her current environment and can also be understood as “involvement in life situation”. This includes the environmental factors. Capacity describes an individual’s ability to execute a task or action and indicates the highest probable level of functioning (World Health Organisation 2001). Performance can be
thought of as what a person “does do” in the usual circumstances of everyday life and capability as what a person “can do” in a defined situation apart from real life (Young et al 1996). Service providers usually want to know what a child’s capability is, that is their capacity in the clinical setting, however in natural environments there are different obstacles and barriers and performance may be quite different (Rosenbaum and Stewart 2004).

Children with CP can show differences between capability and performance. This relationship was examined in 28 physically disabled children using the Activities Scale for Kids (ASK) (Young et al 1996). The ASK is a self-report measure of activity that has both a performance and a capability version, the difference being on how the questions are worded. Capability exceeded performance in the total summary score and capability consistently exceeded performance on all items. In another study, gross motor capability and performance were compared in 307 children with CP aged 6-12 years across environments using the Gross Motor Function measure (GMFM) (Tieman et al 2004). Children were grouped based on capability from their GMFM scores. Those with similar capability demonstrated differences in performance across settings as determined by a parent-completed questionnaire on usual mobility methods.

These studies both suggest that what a child “can do” is often not what they “do do”. This has implications for both the measuring function and planning interventions. Treatments may improve capability in the clinical setting, however if the improvements do not translate into improved performance the child has not received benefit. This is because performance tells more about the child’s usual function in their everyday settings and is more relevant to the child and family (Young et al 1996). Information on capability cannot therefore be automatically extrapolated to performance. Performance in the settings that are important to the children and their usual daily life should be assessed and the environmental factors that hinder or facilitate mobility should be considered (Tieman et al 2004). These studies were important for ensuring that the FMS measured performance during its development.
2.3. Classification of cerebral palsy

Within this thesis, classification of severity of the motor impairment of the children involved was used to describe the samples and to examine various aspects of validity. One of the striking characteristics of CP is its variability of presentation (Liptak and Accardo 2004). It is a heterogeneous group of clinical syndromes with a variety of manifestations (Stanley et al 2000, Graham and Selber 2003). Because of this variability it is important that reliable classifications systems exist for children with CP. Classification is required to further categorise individuals with CP into groups with the purposes of; describing the nature of the problem and its severity, predicting potential future status, comparing different series of cases and evaluating change in individuals at different points in time (Rosenbaum et al 2007). Classification can serve to track incidence and features, educate families with respect to prognosis and assist service planning for service providers (Gorter et al 2004).

2.3.1. Traditional methods of classification

Traditional methods of classification have focused on topographical distribution (Balf and Ingram 1955, Minear 1956), severity and type of movement disorder (Fay 1950, Perlstein 1952). Topographical distribution classifies children based on the distribution of involvement of the limbs of the body. The most common distribution terms used are hemiplegia, diplegia and quadriplegia, however terms such as monoplegia and triplegia are also used (Delgado and Albright 2003).

Spastic diplegia describes a child with gross motor problems, particularly marked in the lower limbs, with usually partially retained fine motor function in the upper limbs (Stanley et al 2000, Bax et al 2007). Spastic quadriplegia refers to the involvement of all four limbs and the trunk (Gorter et al 2004) with severe motor involvement with virtually no hand movements and many have very little speech and language (Bax et al 2007). The child with hemiplegia typically has problems restricted to one side of the body (Bax et al 2007) with involvement of both the upper and lower limb (Stanley et al 2000).
Chapter 2 – Outcome Measurement in CP

There is very little evidence of the reliability of using topographical distribution to classify children with CP and there is also disagreement amongst clinicians regarding the topographical patterns found. Poor reliability of topographical classification can be due to the inconsistency of distinctions between ‘severe diplegia’ and quadriplegia, and between asymmetrical hemi-syndromes and bilateral CP (Howard et al 2005). The Surveillance of Cerebral Palsy in Europe (SCPE) decided to use a simplified classification of spastic children of unilateral or bilateral CP due to the difficulty in agreement between different topographical categories (Surveillance of Cerebral Palsy in Europe (SCPE) 2002). Bilateral spastic CP is the most common syndrome among the different types of CP (Krageloh-Mann et al 1995).

Classification of CP by movement disorder includes spastic CP (85% of the CP population), dyskinetic (7%), ataxic (5%), hypotonic (0.5%) and mixed (2.5%) (Stanley et al 2000). Spastic CP is the most common type of movement disorder and is characterised by abnormal voluntary control, resistance to passive stretch and exaggerated reflexes (Stanley et al 2000). Dyskinetic CP is characterised by involuntary movements and fluctuating muscle tone (Delgado and Albright 2003). Mixed movement disorders often involve spastic with dyskinetic disorders (Gorter et al 2004). Ataxic and hypotonic movement disorders are relatively rare (Reddihough and Collins 2003).

Classification based on the severity of symptoms has used terms such as mild, moderate and severe to help describe the degree of motor impairment (Balf and Ingram 1955, Minear 1956, Blair and Stanley 1985). This method of classification often requires individual judgement and can lack standardisation (Palisano et al 1997, Oeffinger et al 2004). There is little evidence about the reliability of classification systems based on motor impairment, severity and topographical distribution (Gorter et al 2004). The few studies that have been done have shown poor reliability for severity and motor impairment (Blair and Stanley 1985) and classifications by motor type and topography are known to be unreliable (Stanley et al 2000). Because of these issues with traditional classifications, it has become apparent that additional characteristics should be
taken into account for a classification scheme to contribute to the understanding and management of CP (Rosenbaum et al 2007).

For comprehensive classification of CP, the use of four dimensions is recommended (Rosenbaum et al 2007). These are; 1) motor abnormalities, including nature and typology of the motor disorder and functional motor abilities, 2) accompanying impairments, 3) anatomical and neuro-imaging findings, and 3) causation and timing (Rosenbaum et al 2007). Functional motor abilities should be classified using objective scales, such as the Gross Motor Function Classification System (GMFCS) and the Manual Ability Classification System (MACS). Accompanying impairments include such things as presence of epilepsy, IQ, hearing and visual disturbances (Rosenbaum et al 2007).

The focus of this thesis is activity and mobility as measured by the FMS. For this reason, only the GMFCS will be discussed further because it is the relevant classification system for activity limitation with an emphasis on walking. The GMFCS provides information about the child’s gross motor function. This information is lacking when using classifications of movement disorders or topography of CP. It provides supplemental information not provided by traditional classification methods and, as will be discussed below, is a more reliable and valid classification system.

2.3.2. The Gross Motor Function Classification System (GMFCS)

One advance in the classification of children with CP was the development of the GMFCS based on the concepts of disability and functional limitation to address the need for a standardised system to classify gross motor function (Palisano et al 1997). It was developed through nominal group processes and Delphi consensus methods using the Gross Motor Function Measure (GMFM) data from 275 children with CP. The GMFM is a standardized criterion-referenced observational evaluative measure that assesses change in gross motor function (Russell et al 1989). It will be explained in more detail in Chapter Three. The resulting GMFCS is a 5 level ordinal grading system based on the assessment of self-initiated movement with emphasis on function in sitting and walking. Distinctions between levels are based on functional abilities and
limitations, and the need for assistive devices or wheeled mobility. There are 4 age bands each with separate descriptions: 1-2 years, 2-4 years, 4-6 years and 6-12 years. Full descriptions for all age bands are provided in Appendix B. Figure 2.2 illustrates and summarises the descriptors for the levels of the 6-12 age band based on the full version provided in Appendix B. This age band is the most relevant for this thesis and was the most frequently utilised.

The advantage of using the GMFCS for classification is that unlike classification by motor type and topography, it has some evidence of reliability and validity (Palisano et al 1997, Wood and Rosenbaum 2000, Bodkin et al 2003, Morris et al 2004, McDowell et al 2007) and stability over time (Wood and Rosenbaum 2000, Palisano et al 2006). The disadvantage of this system is that descriptors have only been developed and validated for children up to 12 years of age. The reliability and stability in adolescents and adults has not been established. New descriptors for a 13-18 year age band are currently being developed (personal communication with the authors).

The inter-rater reliability of the GMFCS was determined using paired therapists during the original development of the scale with unweighted kappa statistics of 0.55 for children under 2 years of age and 0.75 for children 2 years and older (Palisano et al 1997). Other studies have found good inter-rater reliability of the GMFCS with a generalisability coefficient of 0.93 in 85 children (Wood and Rosenbaum 2000), and good agreement between parent and physiotherapist and between therapists with linear weighted kappas of 0.75 and 0.64 respectively in 184 children (McDowell et al 2007). The reliability between parent report and GMFCS recorded by health professionals was high with intraclass correlation coefficients (ICC’s) of 0.92-0.97 in 97 children with CP aged 6-12 years (Morris et al 2004). The use of ICC’s in this study may have overestimated the agreement because the GMFCS is a categorical system and kappa statistics are more appropriate. Another study examined inter-rater reliability between two experienced physiotherapists using video assessments of 50 children with CP and Down syndrome with a resulting kappa of 0.84 (Bodkin et al 2003).
Figure 2.2 The Gross Motor Function Classification System (GMFCS) for children aged 6-12 years
(figure reproduced with permission from authors based on the descriptives of Palisano et al 1997)
The stability of the GMFCS has also been examined. This involves the extent to which children remain in the same classification level over time. A study examining stability over four different time periods in 85 children with CP showed test-retest reliability to be 0.79 overall with ranges of 0.68-0.87 for different time gaps (Wood and Rosenbaum 2000). A more recent prospective study assessed the stability of the GMFCS in a large sample of 610 children with CP aged 1-13 years (Palisano et al 2006). Children were assessed 2-7 times with 73% of them remaining in the same level for all ratings. Weighted kappas for agreement between first and last rating were 0.84 for children younger than 6 years and 0.89 for children 6-13 years. It is unclear in this study as to whether any of the children had interventions between ratings, which might influence whether children remain in the same level or change levels.

The GMFCS is now used internationally as a common language among clinicians to describe and classify children with CP based on activity limitation (Morris and Bartlett 2004). A review of the impact and utility of the GMFCS found that it is now used extensively in developmental research to describe relationships between functional limitations and comorbidities, for sampling purposes and to select and describe samples, and in clinical practice to set goals and devise interventions (Morris and Bartlett 2004).

Distribution of GMFCS levels among populations of children with CP from Sweden, Canada, Australia and Europe vary slightly (Beckung and Hagberg 2000, Palisano et al 2000, Nordmark et al 2001, Howard et al 2005, Colver and Group 2006, Himmelmann et al 2006). This is shown in Table 2.1. There is a common trend of the highest percentage of children represented in level I and varying spreads among the remaining levels.
Table 2.1 Percentage of children in GMFCS levels in different countries

<table>
<thead>
<tr>
<th>Authors</th>
<th>Sample size</th>
<th>Country</th>
<th>GMFCS level</th>
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<tbody>
<tr>
<td></td>
<td></td>
<td></td>
<td>I</td>
</tr>
<tr>
<td>Nordmark (2001)</td>
<td>167</td>
<td>Sweden</td>
<td>41</td>
</tr>
<tr>
<td>Howard (2005)</td>
<td>323</td>
<td>Australia</td>
<td>35</td>
</tr>
<tr>
<td>Beckung (2000)</td>
<td>116</td>
<td>Sweden</td>
<td>38</td>
</tr>
<tr>
<td>Himmelmann (2006)</td>
<td>367</td>
<td>Sweden</td>
<td>32</td>
</tr>
<tr>
<td>Colver (2006)</td>
<td>818</td>
<td>9 European centres</td>
<td>31</td>
</tr>
</tbody>
</table>

The GMFCS has been used to examine its relationship with limb distribution (topographical distribution) and type of motor impairment in 657 children with CP aged 1-13 years (Gorter et al. 2004). Eighty eight percent of children with hemiplegia were classified as GMFCS I. Children with bilateral CP were represented in all GMFCS levels, with most in III, IV and V. This shows that children with hemiplegia tended to have better activity levels as described by GMFCS with the majority being able to walk independently. Children with bilateral CP showed more diverse activity limitations with most requiring assistance to ambulate and more limited mobility. The overall association between classification at the impairment level (motor impairment and topographical distribution) with that according to function (GMFCS), was statistically significant but low. Grouping by impairment did not provide additional prognostic information in terms of gross motor abilities (Gorter et al. 2004).

A study comparing children with hemiplegia and diplegia within the same GMFCS levels showed that children with hemiplegia performed consistently better on gait and lower extremity function and poorer on upper extremity and school function than those with diplegia (Damiano et al. 2006). The authors suggested that both means of describing and classifying children are required to give full information because GMFCS level does not consider upper extremity
function. However, an alternative method of classification could include using the GMFCS with the more recent MACS (Eliasson et al 2006). This could potentially provide a more meaningful description of activity involving both gross motor function focusing on walking with description and classification of upper limb function.

Motor development curves for each GMFCS level have been created with longitudinal data using the Gross Motor Function Measure (GMFM-66) in 657 children with CP aged 1-13 years (Rosenbaum et al 2002). The children were observed serially for up to four years and the five curves created describe differences in the rates and limits of gross motor development by severity of CP. They show the estimated limit of development decreased as severity of impairment increased with a trend for faster progression to the limit as severity of impairment increased. For each curve an “age-90” was determined, which is the age in years by which children were expected to reach 90% of their motor development potential. Children with more severe impairment tended to reach their limit more quickly, that is, they had a lower age-90. For children in level I the age-90 was 4.8 years, in level II it was 4.4 years, in level III it was 3.7 years, in level IV it was 3.5 years and for level V it was 2.7 years.

This longitudinal data makes it possible to consider prognosis of gross motor development of children with CP based on severity of the initial motor impairment. The information provides parents and clinicians with a way to plan and implement treatments and judge progress. A potential limitation of these curves is that children were classified on the GMFCS level once only, at initial classification. It is possible that children may have changed levels over time; however the authors believe that previous work determining the stability of the GMFCS ensures reclassification would not be an issue (Wood and Rosenbaum 2000). Another potential limitation is that the children were assessed with the GMFM-66 without the use of aids. Consequently, the curves potentially represent the lower limit of the children’s gross motor function because those who use aids would have scored higher on the GMFM if assessed using those aids. Despite these limitations, the curves are an important development in the prognostication of gross motor function and activity in children with CP.
2.4. Surgical and spasticity management for ambulatory children with CP

It is not within the scope of this thesis to discuss all aspects of management of children with CP because the focus is on outcome measurement. However, because Chapter Six addresses changes in mobility after orthopaedic surgery and spasticity management, a brief overview of these interventions will be described. The focus is on surgery and spasticity management for the ambulant child because the FMS is a mobility scale that primarily measures walking.

The secondary abnormalities seen in children with CP are amenable to treatment, however the primary abnormalities are difficult to alter, with the exception of spasticity (Gage and Novacheck 2001). One aim in the management of spasticity is to prevent the development of fixed contractures (Graham and Selber 2003). Options in spasticity management include physiotherapy, orthotics, oral medications, intramuscular injections of botulinum toxin, injections of Phenol to the motor nerves, selective dorsal rhizotomy and intrathecal Baclofen (Gage and Novacheck 2001, Bache et al 2003). Botulinum toxin injections and/or casting are more effective for younger children as are physiotherapy and orthoses, whereas older children often require surgical intervention (Gage and Novacheck 2001).

Most children who have spasticity management will still require orthopaedic surgery for the correction of fixed deformities due to the progressive nature of the musculoskeletal deformities (Graham and Selber 2003). It is now generally accepted that the surgical correction of musculoskeletal deformities for the correction of gait deviations should be performed in one session (Browne and McManus 1987, Nene et al 1993, Abel et al 1999, Fabry et al 1999, Saraph et al 2002, Bache et al 2003, Graham and Selber 2003). This ensures only one admission to hospital and one rehabilitation period, and can be denoted as single event multilevel surgery (SEMLS) (Bache et al 2003). The frequently used procedures are muscle-tendon lengthenings, tendon transfers, rotational osteotomies and bony stabilisation procedures (Bache et al 2003). The surgery
corrects anatomical deformities based on clinical and radiological examination and a biomechanical analysis of gait deviations (Bache et al 2003).

2.5. Outcome measurement in children with cerebral palsy

Outcome research is necessary to quantify the effects of interventions and justify the need for services (Majnemer and Limperopoulos 2002). Information from outcome measurement is used to improve services and provide evidence of the effectiveness of interventions (Law et al 1999). Reliable and valid outcome measures need to be available to ensure this is done accurately (Majnemer and Limperopoulos 2002). Outcome determination in the field of paediatric rehabilitation is challenged by a lack of availability of sound instruments that are appropriate across age groups from infancy to adolescence because the evaluation of outcomes should occur in the context of ongoing growth and development (Majnemer and Limperopoulos 2002).

Recent advances that have influenced outcome measurement include the introduction of the ICF, recognition of the role of child and environmental factors and the appreciation of the importance of key transitions in child development in modifying outcome (Majnemer and Mazer 2004). The ICF can guide the selection of measurement tools to inform goal setting and decision making processes and to determine meaningful outcomes (Rosenbaum and Stewart 2004). It helps promote a broader application of outcome measures to ensure outcomes are being evaluated not only at the impairment level (body functions and structures) but also at the individual and societal levels (activities and participation) (Majnemer and Mazer 2004, Rosenbaum and Stewart 2004). With a recent considerable increase in the number of published studies on outcome assessment for children with CP there has been a trend towards a broader perspective of well-being and level of community integration and an increase in examining performance rather than capability (Majnemer and Mazer 2004).

In the past many outcome studies focused on the impairment rather than functional limitations that result (Majnemer and Mazer 2004, Rosenbaum and Stewart 2004). The focus has now shifted to comprehensively measure all areas
of the ICF and particularly at the activities and participation level which are relevant and more meaningful to the children and their families. The ICF encourages the use of a set of measures rather than one single one to assess all dimensions.

### 2.5.1. Classification of measurement tools

Health status measures can be divided into three broad categories for application of assessment (Kirschner and Guyatt 1985);

- i. discriminative measures,
- ii. predictive measures,
- iii. evaluative measures.

The measurement of functioning and disability can be argued to have one of those three aims (Dekker et al 2005). Discriminative measures are used to distinguish between individuals or groups on an underlying dimension when no external criterion is available (Kirschner and Guyatt 1985). Discriminative tools can lead to the identification of children who are not functioning within age-appropriate or performance-based expectations (Haley et al 1991). Predictive measures are used to classify individuals into a set of predefined measurement categories and generally used as a screening or diagnostic instrument (Kirschner and Guyatt 1985). Evaluative measures are used to measure the magnitude of longitudinal change in an individual or group (Kirschner and Guyatt 1985). This classification of the different types of measures as they relate to children with CP will be discussed further in the introduction of Chapter Three.

Evaluative tools are utilised more widely by clinicians working with children with CP because they are interested in evaluating the effects of interventions and treatments (Young and Wright 1995). The focus of this thesis is on the FMS, which is an evaluative measure that assesses one aspect of activity limitation. The remainder of this chapter and the systematic review in Chapter Three will therefore concentrate on evaluative measures.
2.5.2. Required psychometric properties of evaluative measures

To ensure effective management of children, health service providers need to evaluate the effects of their programs using outcome measures that have demonstrated clinical utility, reliability, validity and clinical responsiveness (Law et al 1999).

Reliability concerns the extent to which measurements are repeatable or consistent (Nunnally 1978, Tooth and Ottenbacher 2004). Validity refers to the degree to which an instrument measures what it is supposed to measure (Dekker et al 2005). The validity of a test is one of its most important attributes and the different aspects that need to be addressed are content, construct and criterion validity (Baxter 2005). Some of these domains overlap which questions the validity of separating them (Baxter 2005). Content validity covers the extent to which the domains of interest are comprehensively sampled by the instrument (Dekker et al 2005). Criterion-related validity entails both concurrent and predictive validity. Concurrent validity concerns how a measure relates to a gold standard while predictive correlates the scale with a future assessment for predicting outcome (Baxter 2005). Construct validity refers to the extent to which a measure scores on theoretically derived hypotheses concerning constructs that are being measured (Dekker et al 2005). Discriminative validity involves assessing the ability of a scale to distinguish between two groups, one group that has the trait or behaviour in question and the other that does not (Streiner and Norman 2003). Responsiveness refers to an instrument's ability to detect important change over time in the concept being measured and is very important criteria for evaluative measures (Dekker et al 2005). Each of these aspects of psychometric properties of measurement tools will be explored fully in the relevant chapters of this thesis.

2.5.3. Outcome measures used for children with CP

In selecting an instrument to measure functioning and disability, the instrument chosen should be best suited for the particular purpose in the specific context of a clinical or research setting, not simply the instrument with the highest rating (Dekker et al 2005). The assessment perspective is important when considering what outcome measures to use and it is increasingly thought that the patient
perspective is considered to be one of the most important (Furlong et al 2005). The age of the child impacts on the selection of assessment tools used (Majnemer and Mazer 2004). Each instrument is validated for a specific age range (Furlong et al 2005). With all of this in mind, each outcome measure available for use in children with CP has been developed for a specific purpose, a specific population and a specific age group. These factors need to be considered when selecting instruments. The following section briefly covers the measurement tools available as they relate to the different domains of the ICF, and will focus largely on activity.

2.5.3.1 Body structures and functions
Tools used to assess body structures and function include such things as goniometers, rulers and radiographs (Goldberg 1991). Diagnostic imaging can evaluate skeletal alignment (Davids et al 2004). Clinical examination assesses the range of motion of all major joints from the pelvis to the foot measured using standardized techniques and goniometry and examination of selective motor control and muscle strength (Davids et al 2004). Gait pathology can be quantified using technical measures. Some clinicians believe that gait analysis is mandatory for optimal treatment of problems relating to ambulation in CP by accurately assessing dynamic gait problems and allowing more precise decision-making (Gage and Novacheck 2001). A contrary view is that the variability seen in gait analyses questions the routine use of this expensive and time-consuming procedure in the management of children with CP (Wright 2003). Gait analysis data provides a biomechanical basis for understanding pathological gait and guides selection of various orthopaedic, neurosurgical and orthotic interventions (Davids et al 2004). Some believe this has added greatly to the understanding of pathological gait patterns and provides an objective outcome measure after surgery for correction of gait (Graham and Selber 2003). Dynamic electromyography data can provide valuable information concerning the timing of muscle activation (Davids et al 2004).

2.5.3.2 Activities and participation
In the past, the focus of management of childhood disability has been to treat the body structures and function with the assumption that improvements there
will translate to improvement in activity and hopefully to the child’s improved participation (Rosenbaum 2007). Assessment of body structures and function does not provide information about the impact on the day to day lives of children. Functional health assessment concentrates on activities and participation and these measures have been the focus in outcome measurement in children with CP in more recent years. There are a number of tools available that assess activity (often in conjunction with participation) and these will be covered in the comprehensive systematic literature review in Chapter Three. Many tools possibly assess both activity and participation and it is difficult to separate the two. For example, the FMS (Graham et al 2004) and Gillette Functional Assessment Questionnaire (FAQ) (Novacheck et al 2000) both assess mobility primarily on an activity level but also include elements of participation. Tools that focus on specifically on participation are summarised in a systematic review of the clinimetric properties of measures of participation (Sakzewski et al 2007) and a structured review of child or family assessed measures of activity performance and participation (Morris et al 2005).

2.5.3.3 Role of environment and personal factors

The influence of factors on the child’s performance and functioning is important to document as they change and grow (Simeonsson et al 2003). Intrinsic variables such as the biological characteristics of CP and child’s personality traits and external variables such as the school environment, family dynamics and peer relationships, can also be determinants of outcome (Majnemer and Mazer 2004). At any given capability level specific environmental factors may have a profound impact on the actual functional performance and a focus on environmental and contextual factors in assessment provides valuable information (Haley et al 1991). Performance of walking, for instance, will depend on the surface, the environmental setting and the social context in which the child has chosen to move (Haley et al 1994). For this reason assessment should include a strong contextual framework that includes all relevant environments and activities that encompass the child’s essential life roles (Haley et al 1994). The extent to which the environmental setting is a determining factor in the lives of the child and family needs to be considered in outcome measurement (Rosenbaum 2007).
2.5.3.4 Capability and performance

The environmental conditions are important when measuring because they help define whether capability or performance are being measured (Young and Wright 1995). The difference between capability and performance is related strongly to environmental factors with capability suggesting it is free from environmental constraints (Rosenbaum 2007). The primary advantages of performance measures are they provide a direct assessment of community function and measure limitations of direct relevance to patients, however in some situations it may be that both capability and performance are measured to identify areas of discrepancy (Young et al 1996). Many surgical and spasticity management interventions might improve capability with a change seen in measures used in the clinical setting. These changes may not necessarily translate into improvements in performance, thus necessitating the need to measure both capability and performance.

Chapter Three explores the available evaluative activity limitation measurement tools that consider environmental setting and also explores whether these tools measure performance or capability. The FMS is one tool that was developed as a performance measure to incorporate the effect of the environmental setting on one aspect of activity, mobility, in children with CP (Graham et al 2004). It is particularly useful for assessing changes in assistance required following complex orthopaedic surgery (Harvey et al 2007). The development of the FMS will be described further in Chapter Four.

2.6. Conclusion

This chapter introduced CP by considering it with reference to the ICF. It also argued the requirements for effective outcome measurement in CP with a focus on evaluative tools that measure activity. Mobility lies within the activity domain of the ICF. The effect of environmental settings on mobility in children with CP was examined. It highlighted the need for the environmental setting to be considered when quantifying mobility with evaluative measurement tools. These background studies formed the basis of the development of the FMS reported in Chapter Four. This chapter provides a foundation for the systematic review reported in Chapter Three.
CHAPTER THREE: SYSTEMATIC REVIEW OF ACTIVITY LIMITATION MEASURES FOR CHILDREN WITH CEREBRAL PALSY

3.1. Introduction

This chapter reports a systematic review and critical evaluation of the literature on activity limitation outcome measures used for children with cerebral palsy (CP). This review has been accepted for publication in Developmental Medicine and Child Neurology (see Appendix I). The review concentrates on evaluative measures used to measure the magnitude of longitudinal change in individuals or groups over time (Kirschner and Guyatt 1985). This review assesses the psychometric properties of evaluative activity limitation measures to determine evidence of reliability, validity and responsiveness. The feasibility and clinical utility of the measures will be examined to ascertain their value and benefit in the clinical setting as well as their practicability. The review will also determine if the measures adequately encompass key areas associated with activity limitation.

In conceptualizing activity and activity limitation, the International Classification of Functioning, Disability and Health (World Health Organisation 2001) defines activity as “the execution of a task or action by an individual” and activity limitations as “difficulties an individual may experience in executing activities”. Participation is “the involvement in life situations” and participation restrictions are “problems an individual may experience in the involvement in life situations” (page 18). For the child with CP, activity includes tasks such as walking, running and climbing stairs while participation includes being involved in recreational activities and taking part in school activities. Numerous measurement tools are available for use in children with CP. A proportion of these assess activity or activity limitation, sometimes in conjunction with measuring participation. The psychometric properties of the tools have not always been tested extensively. The clinical feasibility and practicability varies among the different measures with respect to administration requirements and
purpose, which can affect whether the measures are utilized as routine clinical tools or research tools.

Three broad categories for assessing health indices have been defined (Kirschner and Guyatt 1985). This includes:

i. discriminative indices which distinguish between individuals or groups on an underlying dimension

ii. predictive indices that classify individuals into a set of predefined measurement categories and

iii. evaluative indices that measure change.

In CP, discriminative measures need the capacity to distinguish between individuals with and without a particular characteristic or function (Rosenbaum et al 1990). Included in this category are tools such as the Peabody Developmental Motor Scale (PDMS) (Folio and Fewell 1983) and the Alberta Infant Motor Scale (AIMS) (Piper and Darrah 1994). Predictive measures in children with CP predict possible future status from observations or measures seen in earlier childhood. There are very few of these in the field of CP as predictions for this group are difficult. In a systematic review of assessment measure for functional motor abilities of children with CP no measures were found that were developed for predictive purposes (Ketelaar et al 1998). Evaluative measures will measure change over time or following treatments, for example the Gross Motor Function Measure (GMFM) (Russell et al 1993). Some tools have both a discriminative and evaluative purpose. The Pediatric Evaluation of Disability Inventory (PEDI) (Haley et al 1992) is an example. This review will concentrate on evaluative measures.

Outcome measures available for use in children have been developed for distinct purposes and populations and may not be appropriate for alternative purposes or populations. For example, the Bayley Scales of Infant Development (Bayley 1969) was developed as a discriminative tool for children “at risk” aged 0-18 months. It should not be employed as an evaluative measure for school-aged children because it has not been validated for that purpose in that population. A number of tools which were developed for a specific purpose may
be validated for an alternative purpose. For example, the PEDI was initially developed and validated as a discriminative measure for chronically ill and disabled children from 6 months to 7.5 years (Feldman et al 1990, Haley et al 1991, Haley et al 1993) and has also been validated as an evaluative measure for children with CP (Wright and Boschen 1993, Nordmark et al 2000).

Given that tools for children with CP were developed and validated for varying purposes, no single tool can comprehensively cover all domains required to be assessed. For this reason tools are often used in conjunction to provide different but complementary information.

To ensure a measurement tool is robust it needs to meet requirements of reliability and validity. Reliability is the extent to which a measurement is consistent (Streiner and Norman 2003). Validity concerns the extent to which an instrument measures what it is intended to measure (Streiner and Norman 2003). Evaluative tools are not only required to be reliable and valid, they are also required to be responsive to change. Responsiveness to change is the ability of a measure to detect minimal clinically important differences (Guyatt et al 1987). The most essential feature of an evaluative measure is its responsiveness to clinically important change over time (Rosenbaum et al 1990). These topics will be investigated more thoroughly in the relevant chapters of this thesis.

Recent systematic and structured reviews on outcome measures have focused on quality of life measures (Davis et al 2006), child or family assessed activity and participation measures (Morris et al 2005) or participation measures alone (Sakzewski et al 2007). Earlier reviews on activity based physical function scales for children similar to those in this review covered all tools including discriminative, predictive and evaluative tools (Young and Wright 1995, Ketelaar et al 1998). The one review that has provided an overview of functional assessment measures for children with CP that is most similar to this review is a systematic literature review of measures (Ketelaar et al 1998). The authors identified 17 instruments from the literature developed for evaluative, discriminative and predictive purposes. They found there was a need for measures that can evaluate change in functional abilities, however most
measures are developed and validated for discriminative purposes. Five measures were developed specifically as evaluative measures. Only two measures, the GMFM and PEDI, fulfilled the criteria of reliability and validity with respect to responsiveness to change. Of these two only the GMFM was developed specifically for cerebral palsy. The authors stated that the GMFM and PEDI are complementary as they focus on different aspects of function.

The current systematic review is necessary for a number of reasons. It is almost 10 years since the Ketelaar review was carried out and more recent studies examining outcome measures in CP need to be considered. This review updates the information but also differs from the previous review on a number of points. The current review includes evaluative measures only. This is because the clinical area of interest is measures that assess activity limitation either at one point in time or change in status over time or following interventions. This will be further explained in the inclusion criteria in the methods section. The scientific approach to systematic reviews that is now routinely employed with specific and precise methodology was not used in the Ketelaar review because it pre-dated the more modern approach to systematic reviews. While the authors performed literature searches they did not use a scientific approach to data extraction, quality assessment or data synthesis.

This review will not only have a focused literature search strategy but also a precise review protocol for study selection criteria and procedures, data extraction and synthesis and quality assessment. This review has tighter inclusion and exclusion criteria for both the tools and the studies involved as will be reported in the methods section. The Ketelaar review listed the psychometric testing that had been performed for the instruments included in their review; however the results obtained were not questioned. In addition, the methodology of the studies was not critically analysed. The current review will critically analyse the methodology of the studies assessing psychometric properties of the measures and evaluate whether each tool is robust. The clinical utility and feasibility of the tools will be examined. Also explored will be concepts of environmental setting, participation, assistive devices required and
performance versus capability. These areas were not aims of the previous review.

Of particular interest for this review was determining whether the available tools accounted for different environmental settings, given that studies have shown environmental setting can affect activity and activity limitation in children with cerebral palsy (Palisano et al 2003, Tieman et al 2004). Also of interest was whether the tools incorporated the amount of assistance required and the range of assistive devices used in those different environments. This has not been addressed adequately with the majority of the available measures and will be elaborated further in the discussion.

Given these considerations, the research question for this systematic review was “What are the psychometric properties and clinical utility of the available evaluative measures of activity limitation for children with CP?”

The aims of this systematic review were to:

i. Identify the existing evaluative outcome measures used for children with CP that assess activity limitation

ii. Identify which dimensions of activity limitation they measure;

iii. Identify the studies assessing the psychometric properties of the measures

iv. Critically evaluate the design of these studies;

v. Critically evaluate the psychometric properties of these measures including reliability, validity and responsiveness to change;

vi. Determine if the measures incorporate:

   a. different environmental settings
   b. different assistive devices
   c. performance and/or capability; and

vii. Evaluate the feasibility (including time taken, training and equipment required to administer) and clinical utility of these measures.
3.2. Method

Systematic reviews are designed to help the clinician base their clinical decisions on the best available evidence (Verhagen et al 2001). A systematic review involves the application of scientific strategies, in ways that limit bias, to critically appraise and synthesize all relevant studies that address a specific clinical question using explicit search strategies of comprehensive sources. The scientific review method is the key feature that distinguishes them from traditional narrative reviews (Cook et al 1997). Criteria are used to select studies for inclusion in the review and precise methods used to assess the quality of those studies, extract data from the studies and synthesize findings of the results (Herbert et al 2005). For a systematic review of measurement instruments the aim is to identify all measurements available for a specific purpose using systematic searches and explicit criteria to include or exclude instruments which are then described and evaluated (Dekker et al 2005).

3.2.1. Search Strategy

Electronic searching was performed through the Royal Children’s Hospital library website (http://www.rch.org.au/library) and the University of Melbourne School of Physiotherapy library website (http://www.physioth.unimelb.edu.au). The databases of these collections were examined to locate the most relevant to the review question. The databases that were subsequently searched were selected from these collections because they indexed journals most relevant to health, medicine, allied health and health outcome assessment and would therefore include outcome measures used for children with cerebral palsy. These databases were:

- Medline (1966 to July Week 3 2006)
- Embase (1980 to 2006 Week 30)
- CINAHL (1982 to July Week 4 2006)
- PsychINFO (1806 to July 2006)
- PubMed
- The Cochrane Library
The keywords included in the search strategy were: “cerebral palsy”, “psychometrics”, “reliability”, “validity” and “responsiveness”. These keywords were used because the search was aimed specifically at the psychometric properties of the tools. The search strategy is outlined below. The search did not include keywords such as “evaluative” or “activity limitation” as this would narrow the search excessively and produce a limited output, thus risking exclusion of important tools and references. Several tools comprise a number of domains of which activity limitation is one, therefore using this as a keyword could potentially exclude relevant tools. The term “evaluative” was not used because it is not always clear from first inspection what function a tool performs. Initially terms such as “outcome assessment” and “health assessment” were explored for searching. Matching of these to the other keywords narrowed the search excessively and when matched to “cerebral palsy” the search was too broad. These terms were therefore excluded from the main search.

Key terms were matched to MeSH subject headings and exploded (exp) where relevant to include all subheadings and related terms to each key term used. Terms were searched as keywords (mp) where there were no other thesaurus terms. Terms were then linked to narrow the search. The following strategy was employed to search each database:

1. exp Cerebral Palsy/
2. “Reproducibility of Results”/
3. exp Psychometrics/
4. responsiveness.mp.
5. reliability.mp.
6. validity.mp.
7. 2 or 3 or 4 or 5 or 6
8. 1 and 7

Following the initial main search when it was clear which measurement tools would be included, each of these tools was searched individually as a keyword e.g. WeeFIM.mp., Gross Motor Function Measure.mp. Targeted hand searches were employed in addition to the electronic searching. One method of this involved accessing the reference lists from key articles obtained from the
Chapter 3 – Systematic Review

electronic search. The other method involved searching key journals relevant to outcome measurement in children with CP. The journals searched included Developmental Medicine and Child Neurology and the Journal of Pediatric Orthopaedics because they frequently publish studies using measurement tools to either describe populations of children with CP or assess the effects of treatments. The two methods of targeted hand searching and searching each tool as a keyword electronically were employed to minimize the possibility of missing key studies with the main electronic search.

3.2.2. Inclusion criteria

Initially the title and abstract were used to determine if the article matched the inclusion criteria for the review. These are stated and justified below. Where this was unclear the article was read fully to determine eligibility of inclusion. The studies were reviewed by two people (AH and MM) and assessed for inclusion in the study.

3.2.2.1 Inclusion criteria for the measurement tools

Included in this review were tools that:

i. were developed for children aged 0-18 years including tools that have an upper age limit within this age bracket

ii. were developed for use in children with cerebral palsy or developmental disabilities or neurological conditions including CP

iii. are evaluative measures

iv. quantitatively measure activity or activity limitation or have a subsection measuring this, with particular emphasis on mobility and gross motor function

v. are ordinal scales and are not timed tests or devices measuring metric dimensions

Figure 3.1 shows the process for inclusion of measurement tools included in this systematic review.
This review focuses on children. The adult population with CP presents different problems, functional limitations and environmental factors which require different measurement tools to assess. For this reason the tools included in this review were those developed for children up to 18 years of age. Some evaluative tools developed for children have been validated for a specific age range in childhood and these are included. For example, the PEDI (Haley et al 1992) was developed for children aged 6 months to 7.5 years and the Functional Independence Measure for Children (WeeFIM) (Msall et al 1994) was developed to assess children without disabilities ages 6 months to eight years and children with disabilities aged 6 months to 12 years. These tools provide useful information for their respective age ranges but obviously have limitations for use in older childhood and adolescence. This will be clarified further in the discussion.
Measures were included if they had been developed for children with CP or developmental disorders incorporating children with CP. The previous review of outcome measures (Ketelaar et al 1998) included tools that were developed or used in CP. Measurement tools should be used for the population for which they were developed. If a tool has not been found to be reliable and valid for children with cerebral palsy, then caution is required in using it in that population. The Movement Assessment Battery for Children (ABC) (Henderson and Sugden 1992), for example, was specifically developed for children with motor impairment without any known physical disorder such as cerebral palsy (Leemrijse et al 1999). It can be used to determine presence of Developmental Coordination Disorder (Johnston 2006). Although it may be possible to use this tool in children with mild forms of CP, it has not been tested for reliability and validity in this population.

This review was confined to evaluative measures. Discriminative and predictive measures were not included. The clinical question of interest with this review is the availability of robust measurement tools than can evaluate change over time and following interventions, not the diagnosis of the motor problem or the ability to predict future status. Often the question is not if the child has the condition (which may be determined by discriminative measures) but what their current functional status is or how this is changing over time or determining whether interventions have been effective. “Once the child is diagnosed it is more important to have measures that assess changes in functional capacities of a child over time or after intervention than measures that are developed to answer the question whether or not the child has a motor disorder” (Ketelaar et al 1998).

Tools that focus on participation only or body structures and functions as described in the ICF (World Health Organisation 2001) were not included. The focus of the review is activity or activity limitation measures, particularly those concerned with gross motor skills and mobility. There are some tools which include both activity limitation and participation, for example the Child Health Questionnaire (CHQ) (Landgraf et al 1996) and the Pediatric Outcomes Data
Collection Instrument (PODCI) (Daltroy et al 1998). These tools have been included. Some of the tools included have a subsection of activity as part of a larger and broader inventory. There are not many tools available that purely measure activity or activity limitation.

The tools included in the review are ordinal scales or clinically based judgements about activity. A scale is “a set of numerical values assigned to responses that represent the degree to which the respondent possesses a particular attitude, value or characteristic” (Portney and Watkins 1993) (page 262). They are constructed using a number of different items and often an overall total score is obtained from the sum of the items and are used to quantify performance, in this case activity. They can be self or parent reported measures or measures of direct observation and have different administration methods depending on the purpose of the scale. Other forms of measurement, such as timed tests, assess different elements of activity. Although these are useful they are not a focus of this review. These tests use metric measurement to quantify activity and include tools such as the Uptimer (Pirpiris and Graham 2004) and Timed Up and Go Test (Williams et al 2005). Reasons for excluding these are explained in the exclusion criteria.

3.2.2.2 Inclusion criteria for the studies

Once the tools for inclusion were determined, the studies were deemed eligible to be included in this review based on the following criteria which will be justified below. The studies were eligible if they:

i. evaluated a measure that fulfilled the above criteria
ii. were full papers included in peer reviewed journals
iii. were published between 1966 and July 2006
iv. specifically tested the psychometric properties of the measurement tool
v. evaluated a sample of children with CP alone or the sample clearly included children with CP.

All studies included evaluated one or more of the tools that fulfilled the inclusion criteria for the tools stated above. Only full papers from peer reviewed journals were eligible for inclusion to allow a reproducible search.
strategy. The studies included were published between 1966 and July 2006. The review was performed in July 2006 and electronic databases record back to 1966, therefore literature published before that was excluded. It was essential that the studies were assessing psychometric properties of the selected tools including the various forms of reliability, validity or responsiveness to change. Because the research question for the review concerned tools developed for children with CP, it was essential that the sample within the studies included children with CP alone or children with CP were clearly part of the sample.

3.2.3. Exclusion criteria

3.2.3.1 Exclusion criteria for the tools
Measurement tools not used in this review were those:

i. designed for discriminative (e.g. screening) or predictive purposes
ii. not quantitatively assessing activity or activity limitation
iii. assessing only fine motor, upper limb, communication, activities of daily living or quality of life
iv. that are classifications rather than outcome measures
v. that are individualized for each child using semi-structured interviews
vi. not developed for children with CP or developmental disabilities including CP
vii. designed for infants to assess developmental status
viii. using metric measurement of distance, speed or time parameters

Discriminative and predictive tools were excluded because the focus of the review was evaluative tools. This excluded tools such as the Bruininks-Oseretsky Test of Motor Proficiency (Bruininks 1978), the Bayley Scales (Bayley 1969) and the Functional Motor Assessment Scale (FMAS) (Vermeer 1989). The Bruininks is a discriminative measure for children with motor problems aged 4.5 to 14.5 years. The Bayley Scales is a discriminative measure for “children at risk” aged 2-30 months. The FMAS is a discriminative measure for children with CP aged 4-16 years.
This review focused on physical function and mobility. Although activity can include upper limb function, activities of daily living and communication, these aspects were not the focus of this review. Therefore, tools that assess these dimensions exclusively were not included. Some tools contain numerous domains which include upper limb, communication and activities of daily living, but provided there was a physical function or mobility domain they were included. Pure quality of life tools without domains of activity were also excluded. Furthermore, tools that measure quality and not quantity of activity were excluded.

Classification systems such as the Gross Motor Function Classification System (Palisano et al 1997) are not outcome measures. They perform different functions and are not designed to evaluate change over time. They serve to classify children with cerebral palsy on certain characteristics and allocate them into groups that can be differentiated from one another based on a set of defined variables. The differentiation of the children into clinically significant categories may assist with clinical decision-making (Dobson et al 2007). These classification systems include the classification of gait patterns (Rodda and Graham 2001, Rodda et al 2004, Dobson et al 2006). This review aimed to examine measurement tools that assess change over time and following interventions. For this reason classification systems were excluded from the review.

There are some tools available such as the Goal Attainment Scale (GAS) (Ottenbacher and Cusick 1990) and Canadian Occupational Performance Measure (COPM) (Law et al 1994) that use semi-structured interviews with the child or parent. These tools are individualized for each child and do not have a standard set of items that are applied to each individual (Law et al 1999). It is therefore very difficult to compare results across children and they were not included in this review. The COPM uses a family-centred approach to identify problems, concerns and issues that are relevant to them and has three sections of self-care, productivity and leisure while the GAS uses goal setting and prediction for possible level of attainment with a 5 point scale (Cusick et al 2006). While these tools have the potential to provide very useful,
individualized goals which could reflect important outcomes, this review only includes standardized outcome measures which have a specific set of items that are administered to each child in the same way.

Tools that have not been developed for children with CP or children with developmental or neurological disabilities that include CP were excluded. This includes tools such as the Movement ABC (Henderson and Sugden 1992) which as stated earlier was not developed for children with CP but children with motor impairments without any known physical disorder (Leemrijse et al 1999). The Hoffer scale is sometimes used in the CP population but was developed for children with myelomeningocele (Hoffer et al 1973) and patients with spinal cord injury (Stauffer et al 1978). It is a scale that divides ambulation into four functional levels; community ambulators, household ambulators, non-functional ambulators and non-ambulators. While this scale may provide a meaningful description of ambulatory abilities of children, it has not been validated or shown to be reliable in CP, therefore it was not included in this review.

Tools that were developed to assess developmental status were also excluded on the basis that they focus on developing infants which is a select group. This includes the Peabody Developmental Motor Scale (PDMS) (Folio and Fewell 1983) and AIMS (Piper and Darrah 1994). The PDMS is a discriminative measure for children with delayed motor development aged 0-7 years but has also been tested as an evaluative measure. It tends to be used in infants for developmental status assessment and therefore was not included in this review. The main purpose of the AIMS is to be a discriminative measure for the early detection of motor problems in infants aged 0-18 months. Although the authors report it can be used as an evaluative measure it has not been tested extensively for this. Because it is used for infants as developmental screening it was not included.

There are devices and tests that monitor activity levels using metric measurements of time, speed and distance. These measure a different dimension of activity limitation. They includes tests such as the Timed Up and Go test (TUG) (Williams et al 2005) and devices such as the Uptimer (Pirpiris and
Graham 2004) as well as other activity monitors. The Timed Up and Go test assesses functional ambulatory mobility or dynamic balance by timing the task of standing up from a seat, walking three metres, returning to the seat and sitting down again. The Uptimer is a remote activity monitor that measures the time children spend standing and walking. Other physical activity monitors include pedometers which measure distance walked and/or steps taken over a period of time and accelerometers which measure accelerations produced as a body segment or limb part moves through space (Bjornson 2005). There are many such devices available. These devices are very useful for providing information regarding physical activity in children, and can complement the information derived from scales that are clinically based judgements using self-report or direct observation. As stated earlier this review examines measures that are clinically-based judgements or scales. For this reason timed tests and activity monitors were not included.

3.2.3.2 Exclusion criteria for the studies

Once it was established which tools were included and excluded, the studies were assessed as to whether they were included or excluded. Studies were excluded from the review if they were:

i. descriptive longitudinal health studies
ii. exploring relationships between measures without evaluating psychometric properties
iii. investigations reporting normative data
iv. not inclusive of any children with cerebral palsy in their sample
v. evaluating children younger than 4 years old exclusively, for example the developmental assessment of infants
vi. validating the tool in another language
vii. other adaptations of the tool e.g. computer adaptation
viii. systematic reviews
ix. manuals provided with the tools
x. establishing clinically significant differences

Studies that were descriptive cross-sectional or longitudinal health studies were excluded on the basis that they were not investigating psychometric properties.
of the tools. These studies were using the tools to describe populations of children at one point in time or over time or to compare groups of children with the same tool (Liptak et al 2001, Kennes et al 2002, Vargus-Adams 2005, Vargus-Adams 2006).

A group of studies was excluded because they examined relationships among measures. These studies were not critically evaluating the psychometric properties of the tools, they were looking at associations between different tools without focusing on a particular aspect of psychometric testing (Damiano and Abel 1996, Drouin et al 1996, Ottenbacher et al 1999, Azuala et al 2000, Schneider et al 2001, Tervo et al 2002, Abel et al 2003, Oeffinger et al 2004, Ostensjo et al 2004).

The focus population of this review was children with CP. Studies examining the psychometric properties of the tools in normal populations were excluded. Normative studies have been performed using the ASK (Plint et al 2003), the WeeFIM (Msall et al 1994), the PEDI (Berg et al 2004), the PODCI (Haynes and Sullivan 2001) and the CHQ (Waters et al 2000, Waters et al 2000, Raat et al 2005). Although these studies provide additional information about the tools, this review is concerned with the results of psychometric testing in children with CP. Those studies were therefore excluded. For this same reason, studies that examined the tools using populations that did not include any children with CP in their sample were also excluded. These studies will be referred to in the discussion section of this chapter.

The focus age group for this review included all children with CP up to 18 years of age. Studies that included infants and children less than 4 years old only were excluded. The studies investigating this young age group are primarily evaluating developmental stage of the children and use the tools for developmental screening and assessment purposes. The aim of this review was to examine evaluative tools that measure change in activity over time or following interventions. Consequently, the full spectrum of ages of children and adolescents with CP requires consideration.
Studies that were validating the tools in another language or using an adaptation of the tool were excluded. It was felt these studies would not provide additional relevant information to the review. This involved studies using a Dutch adaptation of the PEDI (Custers et al 2002, Custers et al 2002, Wassenberg-Severijnen et al 2003), the use of the PEDI in Sweden (Nordmark et al 1999) and computer adaptive versions of the PEDI (Haley et al 2005).

This review concerned critically analyzing original studies that were assessing the psychometric properties of the included tools. Other structured or systematic reviews relevant to this topic (Young and Wright 1995, Ketelaar et al 1998, Morris et al 2005, Davis et al 2006, Livingston et al 2007, Sakzewski et al 2007) were excluded because they were not original studies. Also excluded were manuals that are developed for the tools by the authors. These were excluded because they were not published in peer-reviewed journals as stated in the inclusion criteria. This involves the more comprehensive tools such as the GMFM and PEDI that require manuals to administer the tool. The studies reported in these original manuals have largely been published subsequently in peer reviewed journals and hence included in this review. For this reason, it is most unlikely that any crucial results have been overlooked by excluding these manuals.

Studies establishing minimally clinically significant differences were excluded. This concept is very closely related to responsiveness to change and can be considered as an important precursor to testing responsiveness. However, for the purposes of this review studies examining this were excluded because reliability, validity or responsiveness was not assessed. One article on the PEDI was excluded on this basis (Iyer et al 2003).

The inclusion and exclusion criteria for this review were extensive and tight in order to provide a contained and reproducible topic as well as a high quality and thorough critical analysis. The advantages and disadvantages of such extensive and tight criteria will be argued in the discussion.
3.2.4. Data extraction

Data extraction is the process by which the information is obtained from what is reported in each article (Khan and Kleijnen 2007). Data extraction forms are designed and tailored for each specific review to obtain the information relevant to the purpose of that review.

The data extraction form that was developed for this review was individualized to cover areas of general information (including reviewer name, the paper title, authors, journal and publication details), study characteristics and study outcomes. The specific headings on the form were:

i. Study aims
ii. Subject selection
iii. Subject characteristics
iv. Rater characteristics
v. Description of measurement tools
vi. Psychometric qualities including reliability, validity and responsiveness to change
vii. Clinical utility
viii. Statement of findings
ix. Limitations
x. Main conclusions

These headings were deemed to be critical elements to analyse for this review. The study aims included whether they were stated and justified. Subject selection included sample size, population, recruitment procedures, sampling procedures and inclusion and exclusion criteria. In order to analyse outcomes of studies, a clear description of subject selection is required. Subject characteristics included details of age, gender and description of severity of CP. Rater characteristics were relevant for those studies assessing reliability and the data extraction form included information on who the raters were and where they were recruited from.
Description of the measurement tools included which tools were used in each study, whether they were described and what the tools were measuring. Also included was the target population for the tools and information on tool feasibility, such as time taken and training and equipment required for administration of the tool. This information is necessary to have an understanding of each tool’s purpose and from this clinical utility of the tools can be appreciated.

The psychometric testing performed in each study was described including what design of reliability or validity and what statistical analysis was performed. This section provided the principal analysis of the results of the studies. This combined with the quality assessment allowed objective statements to be made regarding the psychometric properties of each tool.

Data were also extracted with respect to a statement of findings, authors identification of limitations and main conclusions of each study. For each thematic area of data extraction, quality assessment was performed. The data extraction and corresponding quality assessment themes and items were recorded on the same form and developed by one person (AH). A copy of this form is included in Appendix C.

Pilot testing of the content of the combined form was carried out by two people (MM and AH) with three articles to ensure all the relevant information was being retrieved and all questions were understood. Revisions were made following this initial testing on the basis of feedback from the two reviewers testing the form.

### 3.2.5. Quality assessment

Quality can be described as “the extent to which all aspects of a study’s design and conduct can be shown to protect against systematic bias, non-systematic bias, and inferential error” (Lohr and Carey 1999) (p472). Quality assessment involves appraising a study’s internal validity. External validity is usually appraised and involves the extent to which the results are applicable outside of the study (Khan et al 1996).
Checklists or systems to assess the quality of included studies have been developed and used in preceding systematic reviews. For the current review an individualised quality assessment checklist was specifically developed by the author (AH). No standardized checklists existed that matched the aims of the review to comprehensively evaluate the type of studies included. There is a lack of empirical evidence in the field of measurement properties to support explicit quality criteria (Terwee et al 2007). The checklist developed included the data extraction and quality assessment utilizing guidelines from other systematic reviews on similar topics (Bialocerkowski et al 2000, Bot et al 2004, Jolles et al 2005, Dobson et al 2007, Eechaute et al 2007) as well as from literature on measuring outcomes and developing quality criteria (Lohr et al 1996, Law et al 1998, Law 2004, Dekker et al 2005, Terwee et al 2007)

There are guidelines available that have been developed with the aim of improving the quality of reporting various study designs. This includes the QUOROM (Quality of Reporting of Meta-analyses) statement for systematic reviews (Moher et al 2000), the CONSORT (Consolidated Standards of Reporting Trials) statement for randomised controlled trials (Moher et al 2005), the MOOSE (Meta-analysis Of Observational Studies in Epidemiology) group for observational studies (Stroup et al 2000) and the STARD (Standards for Reporting of Diagnostic Accuracy) initiative for diagnostic test accuracy (Bossuyt et al 2004). However, these are not direct tools for evaluating the quality of studies and they do not grade the quality to draw conclusions about the strength of the aggregated evidence (Lohr 2004).

For randomized controlled clinical trials criteria lists have been developed, including the Delphi List (Verhagen et al 1998) and the Overview Quality Assessment Questionnaire (OQAQ) (Oxman and Guyatt 1991). However there are no strict guidelines for use of quality assessment in such trials (van Tulder et al 2003). Quality can be used as criterion for inclusion of studies in the review, analysis of the results can be stratified, sensitivity analysis can be performed, quality of studies can be weighted and cumulative meta-analyses of the results can be performed (van Tulder et al 2003).
A review of systematic reviews of prognosis studies found that quality assessment in the primary studies is often incomplete and most of the reviews developed their own set of quality items (Hayden et al 2006). Reviews have been conducted to assess the content of quality assessment tools for non-randomised studies and their use (Deeks et al 2003). The authors identified 194 tools. Many of these omitted key quality domains and six were deemed suitable for systematic reviews, yet each of these required revision to cover all relevant quality domains. A systematic review of 121 critical appraisal tools for allied health research found that most tools were developed for experimental studies and few of them had documented evidence of validity of their items or reliability of use (Katrak et al 2004). There was also a lack of information on tool development processes in most cases.

The validity of quality assessment can be difficult to examine as there is no gold standard or external criterion of quality assessment and there is no widely accepted generic tool that can be applied equally well across study types (Verhagen et al 2001, Katrak et al 2004). “A detailed description of the quality assessment procedure and other choices made in the conduct of the review should enable clinicians and other readers to judge the validity of the procedure” (Verhagen et al 2001) (pg654). The systematic review in this thesis has very explicit methodology enabling it to be easily reproduced, thus supporting its validity.

A grading scale was not used to quantify responses for the quality assessment. Instead a descriptive appraisal of the methodological quality was employed customized to this particular review to ensure relevance to the topic. Because of the heterogeneity of the studies included, combined quantitative analysis is difficult and grading scales are not always appropriate to use. Grading can be used to provide a quality score as an estimate of the methodological quality, as a threshold score for inclusion of the article in the review, as a weighting factor in the statistical analysis or as the input sequence in a cumulative meta-analysis (Verhagen et al 1998). There are many checklists and grading scales for assessing individual reports available, however the reliability, validity,
feasibility and utility of these are often not measured or are variable (Lohr and Carey 1999). Most of the systems available for grading are aimed at randomised controlled trials (Greer et al 2000). Hayden et al (2006) advise against using “quality score” approaches that assign points on the basis of the number of “positive” quality items because it reduces scientific judgement (Hayden et al 2006). Summarizing quality criteria into one overall score can be done in systematic reviews of randomized trials, however it assumes all measurement properties are equally important and is not appropriate for measurement studies (Terwee et al 2007). Grading systems using levels such as (+), (-) and (0) as grades for evidence are available (Greer et al 2000, Bot et al 2004), however there is little evidence of the reliability of this method.

Lohr performed an extensive review of 121 systems for grading the quality of research articles and rating the strength of evidence for four types of research including systematic reviews, randomized controlled trials, observational studies and investigations of diagnostic tests (Lohr 2004). She found that there is no one best approach and that acceptable methods for grading the quality of studies must match the topic and types of studies under review. For example, approaches suitable for observational studies will not be applicable for diagnostics tests.

Particular research questions will require specific approaches for quality assessment. Many of the existing systems for assessing and grading quality are designed for randomized controlled trials and more recently diagnostic studies. The review in question for this thesis focuses on measurement studies and the existing systems are not applicable. For this reason existing criteria lists were not utilized and a quality assessment tool was developed tailored to the specific needs of this review based on similar studies. A grading scale was not used due to a lack of evidence of reliability, validity, feasibility and utility of such systems. A very explicit methodology with quality assessment matched to the aims of the review was employed.
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The quality assessment items were matched to the corresponding themes and items obtained from the data extraction process. The complete form is provided in Appendix C. Examples from this form are included below.

<table>
<thead>
<tr>
<th>Data Extraction</th>
<th>Quality Assessment Questions</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Subject selection</strong></td>
<td></td>
</tr>
<tr>
<td>Is the sample size stated?</td>
<td>Is sample size adequate for aims of study?</td>
</tr>
<tr>
<td>Yes/no</td>
<td>Yes/no/unclear</td>
</tr>
<tr>
<td>Is the sampling procedure described?</td>
<td>Is sampling procedure well-matched to aims of study Yes/no/unclear</td>
</tr>
<tr>
<td>Y/N</td>
<td></td>
</tr>
<tr>
<td><strong>Tool feasibility</strong></td>
<td></td>
</tr>
<tr>
<td>Is the time taken stated?</td>
<td>Is time taken feasible in line with goals of the tool? Yes/no/insufficient info</td>
</tr>
<tr>
<td>Stated/not stated/unclear</td>
<td></td>
</tr>
<tr>
<td>Is the training required</td>
<td>Is the training required feasible? yes/no/insufficient info</td>
</tr>
<tr>
<td>Stated? Yes/no/unclear</td>
<td></td>
</tr>
<tr>
<td><strong>Reliability</strong></td>
<td></td>
</tr>
<tr>
<td>Which type of reliability tested?</td>
<td>Is the result adequate to state the tool is reliable? Yes/no/insufficient info</td>
</tr>
<tr>
<td>Which statistical test was used</td>
<td></td>
</tr>
<tr>
<td>Results of the tests</td>
<td></td>
</tr>
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</table>

The included articles were independently rated for quality by two reviewers (AH and JR) using the combined data extraction and quality assessment form. Guidelines were developed for the reviewers which provided information on some of the questions asked and provided definitions and explanations of the types of validity and reliability. A copy of the reviewer guidelines is provided in Appendix D.
On completion of review of the articles, the reviewers held a consensus meeting with a third person (MM). At this meeting any discrepancies between the reviewers with the articles were discussed and a consensus reached with assistance from the third person. Where consensus was not reached easily all three people involved in this process read the article again until agreement was obtained. This method of consensus for differences between reviewers is a method of improving the agreement as the reliability of most criteria lists is unknown (Verhagen et al 2001, Jackson et al 2005). Following consensus, data synthesis of the extracted evidence was collated and tabulated and the results are shown below.

3.3. Results and Data Synthesis

3.3.1. Search strategy yield

The search strategy included three different stages; the main search strategy, searching each measurement tool as a keyword and hand searching. The initial electronic search using the search strategy and the selected data bases stated earlier yielded 386 articles. From this 56 articles were potential inclusions. On reading the full abstracts or articles from this group and applying the inclusion and exclusion criteria, 18 articles were included. From this initial search it was clear which measurement tools would be included and these could then be searched individually.

When each of the tools was searched as a keyword, 73 articles were retrieved as potentially relevant. From this yield 8 extra articles not retrieved from the initial search were included. Hand searching located another three articles for inclusion. This made a total of 29 articles to be included in this review once the two reviewers applied the inclusion and exclusion criteria. The details of these studies are summarized in Table 3.1.
Table 3.1 Data extraction for the studies included in the review

<table>
<thead>
<tr>
<th>Study</th>
<th>Outcome measure(s)</th>
<th>Sample</th>
<th>Sample size</th>
<th>Ages (yrs)</th>
<th>Gender</th>
<th>Description of sample</th>
<th>Recruitment</th>
<th>Sampling</th>
<th>Psychometrics</th>
</tr>
</thead>
<tbody>
<tr>
<td>Ottenbacher ‘00</td>
<td>WeeFIM</td>
<td>Disabilities</td>
<td>n=174</td>
<td>2-9 yrs</td>
<td>F:63 M:111</td>
<td>mild (50) mod (94) sev (30)</td>
<td>3 centres</td>
<td>proportional</td>
<td>RTC</td>
</tr>
<tr>
<td>Ottenbacher ‘97</td>
<td>WeeFIM</td>
<td>Disabilities</td>
<td>n=205</td>
<td>1-7.5 yrs</td>
<td>F:72 M:133</td>
<td>mild (68) mod (104) sev (33)</td>
<td>RC, schools, homes</td>
<td>proportional</td>
<td>IR reliability TR reliability</td>
</tr>
<tr>
<td>McAuliffe ’98</td>
<td>WeeFIM</td>
<td>CP</td>
<td>n=20 (all CP)</td>
<td>4-12 yrs</td>
<td>D (6) Q (13) H (1)</td>
<td>1 RC</td>
<td>convenience</td>
<td>RTC</td>
<td></td>
</tr>
<tr>
<td>Chen ‘05</td>
<td>WeeFIM</td>
<td>Inpatient rehab.</td>
<td>n=814 CP=90 (11%)</td>
<td>NS</td>
<td>DB</td>
<td>cont validity const validity discrim validity</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Msall ‘94</td>
<td>WeeFIM</td>
<td>Disabilities</td>
<td>n=111 CP=47</td>
<td>2-12 yrs</td>
<td>D (42%), Q (40%), H (17%)</td>
<td>EI/EC</td>
<td>convenience</td>
<td>RTC</td>
<td></td>
</tr>
<tr>
<td>Ottenbacher ’96</td>
<td>WeeFIM</td>
<td>Disabilities</td>
<td>n=67 CP &amp; SB =19</td>
<td>1-6.5 yrs</td>
<td>NS</td>
<td>NS</td>
<td>3 schools</td>
<td>convenience</td>
<td>equiv reliability</td>
</tr>
<tr>
<td>Sperle ’97</td>
<td>WeeFIM</td>
<td>Disabilities</td>
<td>n=30 CP=12</td>
<td>1.5-6 yrs</td>
<td>NS</td>
<td>NS</td>
<td>1 OPC</td>
<td>not stated</td>
<td>TR reliability</td>
</tr>
<tr>
<td>Bjornson ’98</td>
<td>GMFM 88</td>
<td>CP</td>
<td>n=21</td>
<td>4.4-17.7 yrs</td>
<td>(13 M:8</td>
<td>D (13), Q (8)</td>
<td>1 OPC</td>
<td>not stated</td>
<td>TR reliability</td>
</tr>
<tr>
<td>Bjornson ’98</td>
<td>GMFM 88</td>
<td>CP</td>
<td>n=37</td>
<td>3.2-18.1 yrs</td>
<td>(15 M:22</td>
<td>D (100%)</td>
<td>previous RCT</td>
<td>convenience</td>
<td>const validity (responsiveness)</td>
</tr>
<tr>
<td>Nordmark ’97</td>
<td>GMFM 88</td>
<td>CP</td>
<td>n=3 subjects</td>
<td>4-9 yrs</td>
<td>D (100%)</td>
<td>raters from 3 centres</td>
<td>not stated</td>
<td>IR reliability TR reliability</td>
<td></td>
</tr>
<tr>
<td>Russell ‘89</td>
<td>GMFM 88</td>
<td>Disability</td>
<td>n=170 CP=111 13 raters</td>
<td>mild (29) mod (46) sev (36)</td>
<td>2 medical centres</td>
<td>convenience</td>
<td>RTC reliability IR/TRdiscrim validity, RTC</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Russell ‘00</td>
<td>GMFM 66</td>
<td>CP</td>
<td>n=537 (CP)</td>
<td>I(155) II(70) III(104) IV(105) V(103)</td>
<td>DB</td>
<td>stratified</td>
<td>TR reliability</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Russell ‘05</td>
<td>GMFM 88</td>
<td>CP</td>
<td>n=257 (CP)</td>
<td>I(40) II(34) III(93) IV(76) V(14)</td>
<td>DB</td>
<td>randomized &amp; stratified</td>
<td>RTC</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Wang ‘06</td>
<td>GMFM-66 GPFM-88</td>
<td>CP</td>
<td>n=65 (all CP)</td>
<td>I(16) II(8) III(12) IV(19) V(10) &amp; D(31) Q(23) H(11)</td>
<td>7 RC’s</td>
<td>convenience</td>
<td>RTC</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Nordmark ’00</td>
<td>GMFM 88 PEDI</td>
<td>CP</td>
<td>n=18 (all CP)</td>
<td>D (100%)</td>
<td>II(3) III(7) IV(7) V(1)</td>
<td>1 hospital</td>
<td>consecutive</td>
<td>RTC</td>
<td></td>
</tr>
</tbody>
</table>
Chapter 3 – Systematic Review

<table>
<thead>
<tr>
<th>Study</th>
<th>Outcome measure(s)</th>
<th>Sample</th>
<th>Sample size</th>
<th>Ages (yrs)</th>
<th>Description of sample</th>
<th>Recruitment</th>
<th>Sampling</th>
<th>Psychometrics</th>
</tr>
</thead>
<tbody>
<tr>
<td>Vos-Vromans '05</td>
<td>GMFM 88</td>
<td>CP</td>
<td>n=55 (all CP)</td>
<td>2-7 yrs</td>
<td>Mild/mod, I/II</td>
<td>previous RCT randomized</td>
<td>RTC</td>
<td></td>
</tr>
<tr>
<td>Vos-Vromans '05</td>
<td>PEDI</td>
<td>CP</td>
<td>n=55 (all CP)</td>
<td>Gender NS</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Wright '05</td>
<td>GMFM 88/66</td>
<td>CP</td>
<td>n=9 (all CP)</td>
<td>Yr 1 and 2</td>
<td>III(3) IV(3) V(3)</td>
<td>CE program</td>
<td>convenience</td>
<td>RTC</td>
</tr>
<tr>
<td>Wright '93</td>
<td>PEDI</td>
<td>CP</td>
<td>n=40 (all CP)</td>
<td>3-7 yrs</td>
<td>NS</td>
<td>NS</td>
<td>not stated</td>
<td>conc validity</td>
</tr>
<tr>
<td></td>
<td>PEDI</td>
<td></td>
<td></td>
<td>Gender NS</td>
<td></td>
<td></td>
<td></td>
<td>reliability (IR/TR)</td>
</tr>
<tr>
<td>Nichols '96</td>
<td>PEDI</td>
<td>CP</td>
<td>n=65 (all CP)</td>
<td>1-7 yrs</td>
<td>NS</td>
<td>OPC’s (type NS)</td>
<td>convenience</td>
<td>IR reliability, validity (conc &amp; discrim), RTC</td>
</tr>
<tr>
<td></td>
<td>CP and other</td>
<td>CP</td>
<td>n=30 (all CP)</td>
<td>F:29 M:36</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td>F:161 M:149</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Graham '04</td>
<td>FMS</td>
<td>CP</td>
<td>n=310 (all CP)</td>
<td>11 (±/-3.6) yrs</td>
<td>D(124) Q(72) H (114)</td>
<td>OPC</td>
<td>consecutive</td>
<td>IR reliability, validity (IR/TR)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Gender NS</td>
<td></td>
<td></td>
<td></td>
<td></td>
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<tr>
<td>Novacheck '00</td>
<td>FAQ</td>
<td>NM</td>
<td>n=41 (all CP)</td>
<td>3.6-32 yrs</td>
<td>D(19) Q(10) H(5)</td>
<td>1 gait</td>
<td>consecutive</td>
<td>IR reliability (IR/TR)</td>
</tr>
<tr>
<td></td>
<td>disorders</td>
<td></td>
<td></td>
<td>CP=34(83%)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>MS</td>
<td>MS</td>
<td>n=200 (all CP)</td>
<td>5-15 yrs</td>
<td>mild (71) mod (85) sev (13)</td>
<td>OPC’s and other centres</td>
<td>purposive</td>
<td></td>
</tr>
<tr>
<td></td>
<td>disorders</td>
<td>CP=10</td>
<td></td>
<td>F:100 M:100</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>N=166</td>
<td></td>
<td>5-17 yrs</td>
<td>NS</td>
<td>OPC</td>
<td>consecutive</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>CP=8</td>
<td></td>
<td>Gender NS</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Pencharz '01</td>
<td>ASK CHQ</td>
<td>CP</td>
<td>n=180 (all CP)</td>
<td>5-18 yrs</td>
<td>D(46) Q(87) H(45)</td>
<td>1 institution</td>
<td>consecutive</td>
<td></td>
</tr>
<tr>
<td></td>
<td>PODCI CP</td>
<td></td>
<td></td>
<td>Boys&gt;girls</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Vitale '05</td>
<td>CHQ</td>
<td>CP</td>
<td>n=242 (all CP)</td>
<td>5-18 yrs</td>
<td>NS</td>
<td>1 OPC</td>
<td>convenience</td>
<td></td>
</tr>
<tr>
<td></td>
<td>PODCI MS</td>
<td></td>
<td></td>
<td>CP=23</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>disorders</td>
<td>CP=23</td>
<td></td>
<td>F:133 M:109</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>N=166</td>
<td></td>
<td>5-17 years</td>
<td>mild (38) mod (7) sev (33)</td>
<td>1 OPC</td>
<td>convenience</td>
<td>IC discr valid</td>
</tr>
<tr>
<td></td>
<td></td>
<td>CP=8</td>
<td></td>
<td>F:35 M:45</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Wake '03</td>
<td>CHQ</td>
<td>CP</td>
<td>n=80 (all CP)</td>
<td>2-18 years</td>
<td>D(29) Q(25) H(22)</td>
<td>16 centres</td>
<td>consecutive</td>
<td>IC IR TR, RTC, conc, discr valid</td>
</tr>
<tr>
<td></td>
<td>PODCI MS</td>
<td></td>
<td></td>
<td>CP=76</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>disorders</td>
<td>CP=76</td>
<td></td>
<td>F:221 M:249</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Daltroy '98</td>
<td>PODCI</td>
<td>MS</td>
<td>n=470 (all CP)</td>
<td>2-18 years</td>
<td>D(49) Q(38) H(28)</td>
<td>1 OPC</td>
<td>randomised</td>
<td>IC validity: conc, discr RTC</td>
</tr>
<tr>
<td></td>
<td>disorders</td>
<td>CP=76</td>
<td></td>
<td>F:221 M:249</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>McCarthy '02</td>
<td>GMFM CHQ PODCI</td>
<td>CP</td>
<td>n=115 (all CP)</td>
<td>3-10.4 yrs</td>
<td>D(49) Q(38) H(28)</td>
<td>1 OPC</td>
<td>randomised</td>
<td>IC validity: conc, discr RTC</td>
</tr>
<tr>
<td></td>
<td>PODCI</td>
<td></td>
<td></td>
<td>3.1-10.4 yrs</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>GMFM 88</td>
<td>CP</td>
<td>n=64 (all CP)</td>
<td>3-23.3 yrs</td>
<td>NS</td>
<td>DB</td>
<td>stratified</td>
<td></td>
</tr>
</tbody>
</table>

CP = cerebral palsy; NS = not stated; SB = spina bifida; D = diplegia; Q = quadriplegia; H = hemiplegia; mod = moderate; sev = severe; I II III IV V = GMFCS levels; EI = early intervention centre; EC = early childhood centre; DB = database; RC = rehabilitation centres; OPC = outpatient clinics; RTC = responsiveness to change; IR = inter-rater; TR = test-retest; IC = internal consistency; equiv = equivalence; CE = conductive education; MS = musculoskeletal; NM = neuromuscular
On reading one of the articles it was unclear as to whether there were children with cerebral palsy in the sample of children with “neuromuscular conditions” (Young et al 2000). Correspondence from the primary author (Nancy Young) indicated that of the children with “neuromuscular conditions” in the sample there were 10 children with CP (9 with complete data). Based on this information the article was included in the review.

From these 29 articles eight different outcome measures were reported. The tools included were the Activities Scale for Kids (ASK) (Young et al 1995), the Child Health Questionnaire (CHQ) (Landgraf et al 1996), the Gillette Functional Assessment Questionnaire (FAQ) (Novacheck et al 2000), the Functional Mobility Scale (FMS) (Graham et al 2004), the Gross Motor Function Measure (GMFM) (Russell et al 1989), the Pediatric Evaluation of Disability Inventory (PEDI) (Haley et al 1992), the Pediatric Outcomes Data Collection Instrument (PODCI) (Daltroy et al 1998), which is sometimes referred to as the Pediatric Orthopaedic Society of North America (POSNA) scale and the Functional Independence Measure for Children (WeeFIM) (Msall et al 1994). Table 3.2 summarises the features of the eight tools in the review and will be discussed further in section 3.3.3 and 3.3.4.

3.3.2. Descriptive results of studies from data extraction

Table 3.1 displays the tools evaluated in each study, the subject characteristics of the samples, the recruitment and sampling processes and which aspect of psychometric testing was involved for each study.

3.3.2.1 Measurement tools used

The GMFM was the most commonly used tool in this review with 12 studies assessing its psychometric properties. The WeeFIM and PEDI were reported in seven and six studies respectively. The CHQ was assessed in five studies and the PODCI in six. The ASK, FMS and FAQ were reported less often.
3.3.2.2 Subjects selection

Sample size varied in each study. Twelve studies had sample sizes of 1-50, four had sample sizes of 50-100, five had sample sizes of 100-200 and eight had sample sizes of greater than 200. The largest sample size was 814 with 90 of those being children with CP (Chen et al 2005). The populations in each study varied from children with CP only (17 studies) to children with a range of diagnoses that included CP (12 studies). The children with CP could be extracted out in 25 of the studies but not in four of them.

Table 3.1 illustrates the samples for the 12 studies that included children with diagnoses other than CP. These samples were described as children with disabilities (medical or developmental) in six studies, children with musculoskeletal disorders in four studies, children with neuromuscular conditions in one study and children receiving inpatient rehabilitation in one study. These samples included children with other diagnoses such as spina bifida, head injury, developmental delay, Down’s syndrome, congenital talipes equinovarus, scoliosis and other spinal disorders, arthritic conditions, limb deficiency, limb length discrepancy, multitrauma, fractures, congenital impairments, Perthes disease and other miscellaneous orthopaedic and neuromuscular conditions.

In 13 studies children were recruited from more than one centre, in 14 they were recruited from one centre and it was unclear in two studies. The types of centres children were recruited from are shown in Table 3.1. These included various hospital and medical outpatient clinics (nine studies), rehabilitation centres (10 studies), schools or early childhood centres (five studies), motion analysis laboratory (one study) and existing databases or from previous trials (six studies). In five studies it was unclear as to the centre(s) that the children were recruited from.

Thirteen studies used a sample of convenience, six used consecutive samples, three used proportional sampling, three used random sampling (one from an existing randomized trial) and one used purposive sampling. Sampling was not
stated in three studies. Convenience sampling involves choosing subjects on the basis of availability, consecutive being all participants who present within a set time interval. Proportional sampling involves systematic sampling from a set number of groups that represent the population being studied. Purposive sampling involves hand-picking subjects on the basis of specific criteria. Random sampling is a probability method of subject selection where every subject in the population has an equal chance of being chosen (Portney and Watkins 1993). Convenience, proportional and purposive sampling methods are not probability methods of sampling. These methods increase the chance of the sample not being truly representative of the population of interest.

3.3.2.3 Subject characteristics

Table 3.1 summarises the age and gender of the participants in each study. A variety of age ranges was used in the samples usually according to the tools involved and the age range appropriate for those measurement tools. Eight studies included children up to 8 years and the majority of these were assessing the WeeFIM and PEDI. Nine included children up to 15 years and eight included children up to 18 years. Two studies included people over the age of 18 years while the age of subjects was not stated in two studies. Gender of subjects was not stated in 11 of the investigations. In the remaining 18 the ratio of males to females was relatively even.

Description of the severity or classification of the children in the samples varied, as shown in Table 3.1. Six studies described severity of the children involved as mild, moderate or severe. Six classified the children according to GMFCS levels. Twelve described the samples of children with CP according to topography i.e. diplegia, hemiplegia and quadriplegia. Eight studies provided no descriptors of subjects. The larger number of studies using topography rather than GMFCS levels is reflective of the year in which the study was performed and published. The GMFCS has been used to classify children within the last 10 years.
3.3.2.4 Rater characteristics

For the investigations that used raters as part of the procedures, the characteristics of the raters were generally not well detailed. The number of raters used ranged from 1 to 25. The raters used were therapists (in 11 studies), nurses (in two), parents (in three) and doctors (in one). An important consideration here is the difference between what is classified as a “rater” compared to parents or therapists being “respondents” as usual administration of the questionnaires/scales. For example, the PEDI can be administered by therapists interviewing the parents. Inter-rater reliability of therapists administering the tool examines the agreement between the therapists not between the therapist and the parent responding. For some tools inter-rater reliability is really inter-respondent reliability. For example, the FAQ is completed by parents and reliability assesses agreement between parental ratings and does not involve clinicians.

3.3.2.5 Psychometrics tested

Table 3.1 lists the psychometrics tested in each study while tables 3.4-3.11 separate the tools out and describe the testing implemented. These tables will be discussed further in section 3.3.5.

The GMFM was most extensively tested of the tools. Inter-rater reliability was examined in two studies, test-retest reliability in four and internal consistency in one. Content and discriminative validity were tested in one study. Responsiveness to change was examined in eight of the investigations.

The PEDI was examined in six studies. Inter-rater and test-retest reliability were both tested in two separate studies and internal consistency in one. Concurrent and discriminative validity were investigated in three studies and responsiveness was examined in three.

The WeeFIM was cited in seven studies, three of which examined inter-rater, test-retest and equivalence reliability. Only two examined validity (content, construct and discriminative) and concurrent validity was not tested in this
review. Two examined responsiveness, one of which examined it quite extensively.

The FAQ is only cited in one study and this study examined inter-rater and test-retest reliability and concurrent validity. Responsiveness and discriminative validity were not tested. The FMS was examined for inter-rater reliability, responsiveness and concurrent and discriminative validity in one study. The ASK was examined extensively in one study assessing responsiveness as well as inter-rater reliability and content, concurrent, discriminative and construct validity. Another study also examined its concurrent and discriminative validity.

The CHQ and PODCI were both tested for concurrent and discriminative validity in a number of studies (seven studies) but other forms of validity were not investigated. One study examined the reliability of the PODCI and two studies examined its internal consistency. The CHQ was examined for internal consistency in two studies also but reliability was not investigated. Responsiveness was examined in the PODCI but not the CHQ.

3.3.3. Description of the tools included in the review

3.3.3.1 Activities Scale for Kids (ASK)

The ASK is a self-report measure of childhood physical disability developed from first principles using the perspectives of children. It is applicable for children aged 5-15 years with a range of musculoskeletal disorders. It contains 30 items representing nine domains and scores are reported as a percentage of full function. The domains are; personal care, dressing, eating and drinking, play, locomotion, standing skills, stairs, transfers and miscellaneous. There are two versions: a capability and a performance version each covering the same activities but with different response options (Young et al 2000).
## Table 3.2 Features of the measurement tools

<table>
<thead>
<tr>
<th>Outcome measure</th>
<th>Target population</th>
<th>Activities or participation</th>
<th>Aspect of activity measured</th>
<th>Administration: Who and how</th>
<th>Incorporates different settings?</th>
<th>Differentiate assistive devices?</th>
<th>Performance or capability</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>ASK</strong></td>
<td>Children with musculoskeletal disorders 5-15 yrs</td>
<td>Activity and participation</td>
<td>Self-care, play, mobility</td>
<td>Child report and record</td>
<td>Yes</td>
<td>No</td>
<td>Both a performance and a capability version</td>
</tr>
<tr>
<td><strong>CHQ</strong></td>
<td>Generic – all children 5-18 yrs</td>
<td>Activity and participation</td>
<td>HRQOL with physical function subset</td>
<td>Child/parent report &amp; record</td>
<td>Yes</td>
<td>No</td>
<td>Performance</td>
</tr>
<tr>
<td><strong>FAQ</strong></td>
<td>Children with walking disabilities. Age unclear</td>
<td>Activity and participation</td>
<td>Mobility</td>
<td>Parent report and record</td>
<td>Yes</td>
<td>No</td>
<td>Performance</td>
</tr>
<tr>
<td><strong>FMS</strong></td>
<td>Children with CP 4-18 yrs</td>
<td>Activity and participation</td>
<td>Mobility</td>
<td>Clinician interpretation of child/parent report</td>
<td>Yes</td>
<td>Yes</td>
<td>Performance</td>
</tr>
<tr>
<td><strong>GMFM</strong></td>
<td>Children with CP 0-18 yrs</td>
<td>Activity</td>
<td>Gross motor function</td>
<td>Trained therapist observe &amp; score</td>
<td>No</td>
<td>No</td>
<td>Capability</td>
</tr>
<tr>
<td><strong>PEDI</strong></td>
<td>Children with disabilities 0.5-7.5 yrs</td>
<td>Activity and participation</td>
<td>Self-care, mobility and social function</td>
<td>Trained therapist record by observing or interviewing parent</td>
<td>Yes</td>
<td>No</td>
<td>Capability (functional skills) Performance (assistance)</td>
</tr>
<tr>
<td><strong>PODCI</strong></td>
<td>Children with orthopaedic problems 0-18 yrs</td>
<td>Activity and participation</td>
<td>HRQOL with physical function subset</td>
<td>Child/parent report &amp; record</td>
<td>Yes</td>
<td>No</td>
<td>Performance</td>
</tr>
<tr>
<td><strong>WeeFIM</strong></td>
<td>Children with developmental disabilities 0-7.5yrs</td>
<td>Activity and participation</td>
<td>Self-care, mobility and cognition</td>
<td>Clinician report or observe &amp; record</td>
<td>Yes</td>
<td>No</td>
<td>Performance</td>
</tr>
</tbody>
</table>
3.3.3.2 Child Health Questionnaire (CHQ)

The CHQ is a generic paediatric questionnaire designed to determine health-related quality of life in children (Landgraf et al 1996). The 28-item parent form (PF-28) consists of 12 domains and two summary scores (physical and psychosocial) that are scored on a 0-100 scale (Vitale et al 2005). There is also a 50-item parent form (PF-50) consisting of 50 items covering 14 health domains and the two summary scores that are also scored on a 0-100 scale (Vitale et al 2001). There are other elongated versions: the 98-item parent form (PF-98) and an 87-item child form (CF-87). The PF-50 and PF-28 are more commonly used. The domains include physical function, social-emotional, pain, social-physical, behaviour, general health, mental health, self-esteem, parent impact emotionally, family activities, parent impact on time, family cohesion and change in health.

3.3.3.3 Gillette Functional Assessment Questionnaire (FAQ) walking scale

The FAQ walking scale is a 10-level (1-10), parent-report walking scale encompassing a range of walking abilities from non-ambulatory to ambulatory in a variety of community settings and terrains (Novacheck et al 2000). The child is scored independent of the need for assistive walking devices.

3.3.3.4 Functional Mobility Scale (FMS)

The FMS classifies functional mobility in children with CP by rating walking ability at three specific distances, 5, 50 and 500 metres according to the need for assistive devices. The scale incorporates assistive devices ranging from wheelchairs, walkers, crutches and sticks and also includes walking without the need for assistive devices. It uses a six point ordinal scale (1-6) to rate the assistance required at each distance which reflect home, school and community distances (Graham et al 2004).

3.3.3.5 Gross Motor Function Measure (GMFM)

The GMFM was developed as an evaluative measure to assess change in gross motor function in children over time. It is a standardized criterion-referenced observational measure developed for children with cerebral palsy (Russell et al
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1989). The original form has 88 items each scored on a four point ordinal scale (0-3). The items are grouped into five dimensions: lying and rolling, sitting, crawling and kneeling, standing and walking, running, jumping. Scores for each dimension are expressed as a percentage of the maximum score for that dimension and the total score is obtained by averaging the percentage scores across the five dimensions (Russell et al 2000). Rasch analysis was applied to the GMFM-88 in order to improve its interpretability and clinical usefulness. This resulted in a unidimensional interval-measure hierarchical scale, the GMFM-66, consisting of 66 items from the original 88 (Russell et al 2000).

3.3.3.6 Pediatric Evaluation of Disability Inventory (PEDI)

The PEDI was designed as a “discipline free” judgement based method of assessing functional capabilities and performance in chronically ill and disabled children (Haley et al 1992). It is a standardized instrument using parent report through a structured interview by clinicians (Vos-Vromans et al 2005). It consists of three domains: self-care (73 items), mobility (59 items) and social function (65 items) scored as “present” or “absent”. It is conceptualized with three theoretical measurement dimensions: functional behaviours and skills, assistance by caregivers and modifications/adaptive equipment required. Caregiver assistance is determined for eight self-care items, seven mobility items and five social function items scored on a 0-5 scale. Modification and equipment required is scored on a four point scale (Ziviani et al 2001).

3.3.3.7 Pediatric Orthopaedic Data Collection Instrument (PODCI)

The PODCI was designed to measure quality of life and functional levels in children with orthopaedic problems. It is available in three formats: one for parents of younger children (0-10 years), one for parents of adolescents (11-18 years) and a self-report form for adolescents (11-18 years) (Pencharz et al 2001). It has 108 items. A global function scale encompasses four subscales of upper extremity function, transfers and mobility, comfort and physical function and sports. It also has a happy and satisfied scale as well as expectations for treatment. It is scored on a 0-100 scale.
3.3.3.8 Functional Independence Measure for Children (WeeFIM)

The WeeFIM is based on the format of the Functional Independence Measure (FIM) for adults and measures functional independence in children (Ottenbacher et al 1997). It is concerned with the amount of assistance required for children with disabilities to perform basic life activities (Ottenbacher et al 1997). It aims to evaluate disability using a minimal essential data set that is discipline free, easy to use and designed to track outcomes across settings (Msall et al 1994). It consists of 18 items categorized in six specific functional domains of self-care, sphincter control, mobility, locomotion, communication and social cognition. A seven level ordinal scale (1-7) is used to rate the amount of assistance needed for each item (Chen et al 2005). The minimum total rating is 18 and the maximum is 126 (Ottenbacher et al 1996).

3.3.4. Descriptive features of the measurement tools

Table 3.2 lists each of the eight measurement tools included in this review and their characteristics. For each outcome measure it displays:

i. the target population and age range

ii. what domain of the ICF is covered

iii. what aspect of activity limitation is measured

iv. who administers the tool and how

v. whether the tool incorporates different environmental settings relevant to children

vi. whether the tool differentiates the assistive devices used

vii. whether it measures performance or capability

3.3.4.1 Target population of the tools

For two of the tools, the GMFM and FMS, the target population is CP exclusively (Russell et al 1989, Graham et al 2004). The GMFM has been examined in other populations such as Downs’ Syndrome (Russell et al 1998) but was developed and validated to assess children with CP (Russell et al 1989). The PEDI and WeeFIM were both developed for children with developmental disabilities. The FAQ is for children with walking disabilities associated with neuromuscular disorders. The ASK and PODCI are intended for children with
musculoskeletal or orthopaedic disorders. The CHQ is the only generic measure included in this review with the target population including all children.

Two tools are for the full spectrum of childhood and adolescence (0-18 years), the GMFM and PODCI. The PEDI and WeeFIM were both developed for children aged 6 months to 7.5 years. The ASK is for children aged 5-15 years and the CHQ is for children aged 5-18 years. The original article for the FMS states the tool is for children aged 6 years to skeletal maturity, however further development of the tool states that it is for children aged 4-18 years (personal correspondence with author Kerr Graham). It is unclear from the one article on the FAQ what age group it is targeted for, but the study includes the ages of 3.6 to 32 years, suggesting it can be used in both children and adults.

3.3.4.2 ICF domain covered by each tool

Seven of the tools measure both activity and participation as described by the ICF (World Health Organisation 2001), while the GMFM measures activity alone. It can be difficult to separate activity from participation as they can be very closely linked. Some of the tools have items that incorporate the two concepts, for example the FMS and FAQ. Others have different items that cover activity and participation, for example the ASK and the PEDI. The different tools measure different aspects of activity, as illustrated in Table 3.2. The FAQ and FMS measure mobility and the GMFM measures gross motor function. Three tools measure physical function as a subset of a larger inventory of general function (PEDI, WeeFIM and ASK). The CHQ and PODCI are both health related quality of life measures that have a subset on physical function.

3.3.4.3 Administration method

Methods of administration vary from direct observation to self (or parent) report. The GMFM assesses children by direct observation with a trained therapist observing and scoring the child. The PEDI is administered by a trained therapist observing the child or by interviewing the parent. The WeeFIM is recorded by either direct observation or clinician-report and usually a combination of both. All other tools use self-report, which can be parent report by proxy. The ASK uses child-report primarily while the FAQ tends to be
parent-report. The FAQ, ASK, CHQ and PODCI are all completed by the child or parent, whereas the FMS is clinician administered self-report.

3.3.4.4 Incorporation of different environmental settings

All tools except for the GMFM take into account different environments when they evaluate the function of the children. The FMS, however, is the only one that examines in detail the differences in function between these environments. The FMS, FAQ, PODCI and CHQ incorporate community distances into items. The WeeFIM and PEDI limit walking to 150 feet and the ASK does not specify distances but includes indoor and outdoor activity. This concept will be elaborated on further in the discussion.

3.3.4.5 Consideration of different assistive devices

Most of the tools do not differentiate between different assistive devices when assessing function, in particular mobility and locomotion. The FMS clearly distinguishes between different devices used by the same child and is the only one to do so.

3.3.4.6 Capability versus performance

It is important to distinguish whether tools measure performance or capability. Performance is what the child “does do” in the usual circumstances of everyday life. This differs from capability which quantifies what they “can do” in a defined situation apart from real life (Young et al 1996). Studies have shown that performance can differ from actual capability in the same child (Young et al 1996) and across environmental settings (Tieman et al 2004).

Five of the included tools measure performance only (CHQ, FAQ, FMS, PODCI and WeeFIM). The ASK has both a performance and capability version. The PEDI section on functional skills measures capability and the caregiver assistance section measures performance. The GMFM measures capability despite some earlier studies claiming it is a performance measure. The developers of the GMFM classified the tool as a performance measure because it observes activity in a clinical setting (Russell et al 1989). However more recent work on this concept defines capability and performance differently and
terminology has changed. The ICF (World Health Organisation 2001) defines performance as “what an individual does in his or her current environment and can be understood as involvement in a life situation” (p19). Capacity “describes an individual’s ability to execute a task or action in a standardized environment. It aims to indicate the highest probable level of functioning” (p20). Using the framework of the ICF and other work on the concept (Young et al 1996), the GMFM is designed to measure the child’s “best ever performance” (Nordmark et al 2000) and therefore is a capability measure.

3.3.4.7 Feasibility and Clinical Utility

Table 3.3 describes the feasibility and clinical utility for each of the tools included in the review. Feasibility involves the time taken, equipment required and training required to administer the tools (Law et al 1999, Morris et al 2005). Eight studies stated the time taken for the tools involved yet it was not stated in 20 and in one it was unclear. The time taken to administer each tools varied from 9-60 minutes. The GMFM and PEDI are the most time consuming with administration times of 45-60 minutes each. The CHQ and PODCI take approximately 30 minutes each, depending on which version is used. The WeeFIM can be scored in less than 20 minutes and the ASK takes on average 9 minutes to complete. The articles included in this review did not provide any information on time taken to administer the FMS and FAQ. It is the “clinical impression” of experts that the FAQ can be completed by parents in 10 minutes and the FMS completed by clinicians in 10 minutes (Professor Kerr Graham, personal communication).
Table 3.3 *Measurement tools: feasibility and clinical utility*

<table>
<thead>
<tr>
<th>Tool</th>
<th>Feasibility: Time taken</th>
<th>Equipment required</th>
<th>Training required</th>
<th>Clinical utility</th>
</tr>
</thead>
<tbody>
<tr>
<td>ASK</td>
<td>9 minutes</td>
<td>Not stated. Self-report, assume none required.</td>
<td>Not required, child is the respondent.</td>
<td>Measures functional ability, change and outcomes important to children. Can be used by mail. Practical. Minimal floor and ceiling effects.</td>
</tr>
<tr>
<td>CHQ</td>
<td>30 minutes</td>
<td>Self-report, assume none required.</td>
<td>Not required, child or parents are respondents</td>
<td>Generic tool. Broad picture of health status and well-being in wide range of paediatric populations. Includes psychosocial as well as physical with some loss of specificity. Not for whole range CP due to floor &amp; ceiling effects. Discriminates healthy from sick children. Provides parental perspective.</td>
</tr>
<tr>
<td>FAQ</td>
<td>Not stated</td>
<td>Not stated. Self-report, assume none required.</td>
<td>Not required, parent completes</td>
<td>Assessment of community function specific to task of walking for range of walking abilities. Authors state it can evaluate treatment, yet responsiveness has not been tested.</td>
</tr>
<tr>
<td>FMS</td>
<td>Not stated</td>
<td>Not stated. Self-report, assume none required.</td>
<td>Not stated</td>
<td>Assesses mobility over 3 distances to represent home, school and community. Emphasis on range of assistive devices required. Monitors change &amp; discriminate level of disability.</td>
</tr>
<tr>
<td>PEDI</td>
<td>45-60 minutes</td>
<td>Testing manual and software program to score</td>
<td>Training and trial use required but extent unclear.</td>
<td>Describes function in community. Focus on self-care and mobility in everyday life. Considers degree of caregiver assistance and independence. Clinical and research tool. Best as parental report from interview. Can measure change after treatments.</td>
</tr>
<tr>
<td>WeeFIM</td>
<td>&lt;20 minutes</td>
<td>Scoring manual to calculate overall score</td>
<td>Training video and practice ratings.</td>
<td>All disciplines. Tracks function. Used to set goals, treatment plans &amp; evaluate change in ADL over time and post treatment. Includes impact on caregivers. Practical. By observation, interview or over phone.</td>
</tr>
</tbody>
</table>
The amount of equipment and training required for most of the tools was not described well in the studies. Equipment needs were stated in four studies only and it was unclear or not stated in 25. Training requirements were stated in seven studies and it was unclear or not stated in 22. It would appear that for the ASK, CHQ, FAQ, FMS and PODCI little training or equipment is required. These tools rely on self-report and not direct observation, thereby eliminating the need for equipment to administer them. Questionnaire forms are required for the ASK, CHQ, FAQ and PODCI. Formal training is not required for these tools as the parents or child record the information (except for the FMS). The FMS is administered by clinicians based on self-report. There is a brochure available explaining the tool and rating procedure which is recommended to be read prior to administration of the scale. Score sheets or scoring programs are available for the ASK, CHQ and PODCI.

The GMFM requires training through a self-instructional CD ROM before assessing children and a manual and scoring program is necessary. Some “usual physiotherapy equipment” is required to be able to administer it. The PEDI requires training sessions and trial use but the extent of this training is unclear. A manual and score sheets are required as well as a software program to transform raw scores into normative and scaled scores (Nordmark et al 2000). Before using the WeeFIM training is recommended using the training video and practice rating of children. A scoring manual is also available.

The clinical utility of each of the tools as described by the articles is described in Table 3.3. This was addressed in 26 of the 29 studies and the table collates the overall clinical utility of each tool from all studies. Because all measurement tools are evaluative measures, they were all stated to measure change over time or after interventions and assess the effectiveness of treatments. Some of the tools can be administered over the telephone (WeeFIM) or via the mail (ASK). The GMFM requires direct observation, the WeeFIM is either by observation or interview and the PEDI is best obtained by parental report in interview. The FMS requires interviewing the child and parents and the FAQ, CHQ, PODCI and ASK are self report.
The GMFM, FMS and FAQ are quite specific to one aspect of activity. The ASK, PEDI and WeeFIM give a broader description of function by including self-care (all three), social function (PEDI) cognition (WeeFIM) and play (ASK) along with mobility and therefore provide more information. The CHQ and PODCI are health-related quality of life measures that incorporate physical function. They have the advantage of giving a broad picture of functional health status but lose some specificity with that. Both also show floor and ceiling effects in various domains (Pencharz et al 2001, Vitale et al 2001, McCarthy et al 2002, Damiano et al 2005, Vitale et al 2005) and therefore are not appropriate for use in the range of severity of CP seen in the population. The ASK has only minimal ceiling effects and very low floor effects (Young et al 2000, Pencharz et al 2001) suggesting it is appropriate for all severity of CP. Some ceiling effects were seen for the GMFM (Damiano et al 2005, Vos-Vromans et al 2005) indicating it is not able to distinguish higher functioning children while a number of children showed floor effects on the WeeFIM (Ottenbacher et al 2000). The FMS and FAQ were not assessed for floor or ceiling effects.

Parental perspectives are provided on the CHQ and PODCI through parental versions and the PEDI and FAQ because they are parental report scales. The amount of caregiver assistance or impact on caregivers is an important factor when assessing children with CP and this is included in the PEDI, WeeFIM and CHQ. The PODCI also contains information on expectations.

3.3.4.8 Authors identification of limitations of the studies

set and lack of a standardized protocol for testing (Russell and Gorter 2005). For responsiveness of the WeeFIM, a small sample size, subjective rating of meaningful change and using parents as reporters was stated as limitations of the study design (McAuliffe et al 1998). A study assessing reliability and validity of the GMFM, PODCI, CHQ and PEDI stated small sample size and not covering all aspects of psychometric testing as limitations of the study (McCarthy et al 2002). Small sample sizes were a common theme in limitations, reducing the generalisability of the results. For many of the other studies limitations were lacking in detail or limitations of the tool rather than the study were addressed.

3.3.5. Quality assessment results

3.3.5.1 Study aims

For the 29 studies, the aims were stated adequately and were justified except for one (Vitale et al 2001). The aim of this investigation was vague. The authors stated “the study seeks to evaluate the CHQ in orthopaedics and to compare it with the PODCI”, without clarifying what aspects of the CHQ was being evaluated.

3.3.5.2 Subject selection

The sample size was large enough to ensure adequate statistical power in 25 of the studies. In four the sample sizes were small (Nordmark et al 1997, McAuliffe et al 1998, Nordmark et al 2000, Wright et al 2005). Three of these studies assessed responsiveness with sample sizes of 20 (McAuliffe et al 1998), 18 (Nordmark et al 2000) and 9 (Wright et al 2005). One had a small sample size of three (Nordmark et al 1997). This study was examining reliability and involved 15 raters. For all studies the population of participants was described adequately. This includes the samples involving children with diagnoses other than CP that were described as children with disabilities, neuromuscular or musculoskeletal disorders.

The recruitment procedures were described and well-matched to the study aims in 24 studies yet five were inadequately described. For these five (Wright and Boschen 1993, Msall et al 1994, Ottenbacher et al 1996, Nordmark et al 1997,
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Vos-Vromans et al 2005) the recruitment procedures were unclear and it was difficult to assess if they were well-matched to the study aims. The sampling procedure was described well in 23 studies and well-matched to the study aims. Sampling was unclear or not described at all in six (Wright and Boschen 1993, Msall et al 1994, Nordmark et al 1997, Bjornson et al 1998, Bjornson et al 1998, McAuliffe et al 1998, Vos-Vromans et al 2005). Whether the sampling procedures were well-matched to the aims of the study was unable to be determined.

3.3.5.3 Subject characteristics


Description of the subjects using GMFCS levels, topography of CP (hemiplegia, diplegia and quadriplegia) or severity of the condition (mild, moderate, severe) was generally adequately described in 20 studies. This included those studies with mixed samples where topography or GMFCS level is not applicable. It also includes a large number where GMFCS level is not applicable due to the date of the article being before the GMFCS was routinely used in the field of CP. There were nine studies where the subjects were not described well in relation to CP severity, topography or GMFCS level (Wright and Boschen 1993, Ottenbacher et al 1996, Nordmark et al 1997, Sperle et al 1997, Pencharz et al 2001, Vitale et al 2001, Chen et al 2005, Damiano et al 2005, Vos-Vromans et al 2005). One of these was partially described but lacked some information (Vos-Vromans et al
There were five studies that described the sample of children with CP very thoroughly by using more than one descriptor. Four of these described them by GMFCS level and topography (Russell et al 2000, Wake et al 2003, Wright et al 2005, Wang and Yang 2006). One described them by GMFCS level and type of movement disorder (Russell and Gorter 2005).

### 3.3.5.4 Inclusion and exclusion criteria

The inclusion criteria were described adequately to satisfy the aims in 21 studies. There was insufficient information or the criteria were not stated at all in eight (Msall et al 1994, Nordmark et al 1997, Sperle et al 1997, Bjornson et al 1998, Nordmark et al 2000, Novacheck et al 2000, Graham et al 2004, Vitale et al 2005). Exclusion criteria performed poorly overall with only four studies describing them well in order to reduce the effects of confounding factors such as maturation, treatment interfering with results or diverse diagnostic groups within the sample (Russell et al 2000, Vitale et al 2001, Chen et al 2005, Wang and Yang 2006). The remaining 25 studies provided insufficient information or often no information at all regarding exclusion criteria.

### 3.3.5.5 Tool feasibility

Tool feasibility was not explored particularly well in the studies. Many articles did not state the time taken, the equipment required or training required for administration. Although some tools require up to 60 minutes to complete (PEDI, GMFM), these are aimed to be comprehensive. The time taken therefore is in line with the aims of the tool. The tools that require less time to administer aim to be less comprehensive. For the purpose of this review all tools administration times are according to the aims.

It was difficult to extract information regarding equipment needs from the included studies. Self-report measures do not require equipment (ASK, CHQ, FAQ, FMS, PODCI). The GMFM requires “usual physiotherapy equipment” and scoring manuals although the exact equipment required is not known without sourcing the manual. The PEDI and WeeFIM require manuals or software yet descriptions of these are vague.
Training required for administration of the tools varies. This was also not described well. The five tools that are self-report do not require training. The WeeFIM, PEDI and GMFM do require some training and trial use and the requirements appear to be in line with the aim of the tools. Similarly with administration time, the tools that aim to be more comprehensive require more training, such as the GMFM and PEDI.

3.3.5.6 Psychometric testing

The psychometric testing results are summarised in tables 3.4-3.11. The results for each tool are presented separately with reliability, validity and responsiveness listed. The reliability testing procedures and results were comprehensible and easily interpreted in most studies. The validity questions posed were clear yet interpretation of results was a little more difficult to ascertain. Responsiveness was inconsistent among the studies with some thoroughly examining it with sound statistical analysis while others appeared to be investigating change after treatments rather than responsiveness of the tool. These results were often difficult to interpret. Psychometric testing results will concentrate on the total scores of the tools and mobility and physical function domains. Social and cognitive domains will not be interpreted with any depth because the focus of this review is tools that measure activity or activity limitations.

Judgements on reliability are based on guidelines that 0.70 is recommended as a minimum standard for reliability (Nunnally and Bernstein 1994). Criteria for correlation coefficients describe good to excellent correlation if the coefficient is over 0.75, moderate to good if it is 0.50-0.75, fair degree of correlation for 0.25-0.50 and little or no relationship if it is 0.00-0.25 (Portney and Watkins 1993). However, these are not strict cut-off points because they are affected by sample size, measurement error, and the types of variables being studied (Portney and Watkins 1993). Some of the studies used effect sizes to analyse responsiveness. Effect size of 0.2 is considered small, 0.5 moderate and 0.8 large (Cohen 1977).
Table 3.4 *Psychometric properties of the ASK*

<table>
<thead>
<tr>
<th>Authors</th>
<th>Design</th>
<th>Result</th>
<th>Overall Quality assessment</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Reliability</strong></td>
<td>Young (2000)</td>
<td>Inter-rater</td>
<td>ICC 0.99 (between 2 raters only)</td>
</tr>
<tr>
<td><strong>Validity</strong></td>
<td>Young (2000)</td>
<td>Content</td>
<td>Item hierarchy shown</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Concurrent</td>
<td>r=0.92 (child vs clinician report)</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Construct</td>
<td>Divergence (-0.03) &amp; convergence (0.43) shown</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Discriminative</td>
<td>Significant differences between mild, moderate and severe</td>
</tr>
<tr>
<td>Pencharz (2001)</td>
<td>Concurrent</td>
<td></td>
<td>r=0.8 (PODCI) and 0.5 (CHQ)</td>
</tr>
<tr>
<td></td>
<td>Discriminative</td>
<td></td>
<td>Discriminates ambulation level (Hoffer)</td>
</tr>
<tr>
<td><strong>Responsiveness to change</strong></td>
<td>Young (2000)</td>
<td>Mean change, effect size, SRM and t tests</td>
<td>Change seen with performance version more change than capability version</td>
</tr>
</tbody>
</table>

SRM = standardized response means; ICC = intraclass correlation coefficients
The ASK

Table 3.4 shows the psychometric properties of the ASK. One study evaluated reliability of the ASK in a large sample of children (Young et al 2000) as part of a larger study examining validity and responsiveness. Excellent agreement was found between two clinicians, however more extensive reliability testing is required.

The content, concurrent, construct and discriminative validity of the ASK was examined in two studies (Young et al 2000, Pencharz et al 2001). Rasch analysis confirmed all items measured a single construct (physical disability) and showed a hierarchical structure among the items (Young et al 2000). Concurrent validity using Pearson’s r showed high correlation of the child-reported ASK scores with clinician–reported observation ASK score (Young et al 2000). The second showed fair correlations between the ASK and the CHQ physical function subscale and higher between the ASK and the PODCI global function scale (Pencharz et al 2001). Construct validity was supported by low correlations with dissimilar constructs on the Health Utilities Index (HUI) proving divergence and higher for similar constructs proving convergence (Young et al 2000). Discriminative validity testing showed significant differences between the mild, moderate and severely disabled groups of children (Young et al 2000). The ASK was also found to significantly discriminate ambulation level using the Hoffer scale (Pencharz et al 2001). Overall the studies provided evidence of the validity of the ASK.

Responsiveness of the ASK was examined thoroughly by comparing mean change observed on the ASK compared with the CHQ, as well as calculating effect size and t tests in a subset of 34 children (Young et al 2000). The ASK-performance showed similar change to the CHQ and the ASK-capability showed slightly less change. As the authors state, that it is difficult to ascertain the clinical importance of the results and further work is required.
Table 3.5 *Psychometric properties of the CHQ*

<table>
<thead>
<tr>
<th>Authors</th>
<th>Design</th>
<th>Results</th>
<th>Overall Quality Assessment</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Reliability</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Wake (2003)</td>
<td>Internal consistency</td>
<td>Cronbach â 0.75-0.97</td>
<td>Good internal consistency. Requires reliability testing in CP.</td>
</tr>
<tr>
<td>McCarthy (2002)</td>
<td>Internal consistency</td>
<td>Cronbach â 0.80-0.94</td>
<td></td>
</tr>
<tr>
<td><strong>Validity</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Vitale (2005)</td>
<td>Concurrent Discriminative</td>
<td>0.17-0.41 (vs subjective physician score) Detect difference between quadriplegia and hemi/diplegia but not between hemiplegia and diplegia</td>
<td>Only concurrent and discriminative validity tested in CP. Concurrent validity shows moderate to good correlation in 2 well designed studies (Pencharz &amp; McCarthy). Other studies use subjective clinician scores. Discriminative validity indicates that CHQ is unable to discriminate severity or topography adequately.</td>
</tr>
<tr>
<td>Vitale (2001)</td>
<td>Concurrent Discriminative</td>
<td>0.31-0.71 (vs subjective clinician score) CP lower than “normal”</td>
<td></td>
</tr>
<tr>
<td>Pencharz (2001)</td>
<td>Concurrent Discriminative</td>
<td>0.5 (ASK) 0.6 (PODCI)</td>
<td></td>
</tr>
<tr>
<td>McCarthy (2002)</td>
<td>Concurrent Discriminative</td>
<td>0.81 (PODCI) 0.75 (PEDI) 0.74 (GMFM)</td>
<td></td>
</tr>
<tr>
<td>Wake (2003)</td>
<td>Discriminative</td>
<td>Physical function scale able to discriminate between mild and severe</td>
<td></td>
</tr>
<tr>
<td><strong>Responsiveness to change</strong></td>
<td>Not examined</td>
<td></td>
<td>Responsiveness to change needs to be examined in the CP population</td>
</tr>
</tbody>
</table>
The CHQ

Table 3.5 summarises the psychometric properties of the CHQ. Good internal consistency of the items on the CHQ was demonstrated (McCarthy et al. 2002, Wake et al. 2003). No other form of reliability testing was conducted. The concurrent and discriminative validity of the CHQ was examined in a number of studies yet other forms of validity were not addressed. Concurrent validity using Spearman’s rho was supported overall by the studies. High correlations were found between the physical function domain of the CHQ with the transfers and mobility domain of the PODCI, and moderate correlations with the mobility domain of the PEDI and the GMFM (McCarthy et al. 2002). Low correlations were found for dissimilar domains. The CHQ parent form physical subscale correlated moderately with the ASK and PODCI (Pencharz et al. 2001). A weak correlation was found between a subjective physicians physical score with the CHQ (Vitale et al. 2005). One study compared the CHQ to subjective clinicians physical scores and found low to moderate correlations (Vitale et al. 2001).

Discriminative validity of the CHQ was examined in several studies (Pencharz et al. 2001, Vitale et al. 2001, McCarthy et al. 2002, Wake et al. 2003, Vitale et al. 2005). It showed a weak ability to discriminate ambulation level using the Hoffer Scale (Pencharz et al. 2001). It was not able to detect differences between hemiplegia, diplegia and quadriplegia (McCarthy et al. 2002). The CHQ was only able to detect differences between quadriplegia and diplegia/hemiplegia and not between hemiplegia and diplegia (Vitale et al. 2005). In one study children with CP were said to score lower on the CHQ than “normals” and those with more comorbidities lower but the statistical evidence is not available to support this comment (Vitale et al. 2001). The physical function scale was able to discriminate severity between mild and severe (Wake et al. 2003) but those children with moderate disability were not considered. These results suggest the CHQ is not able to discriminate well. Testing of other forms of validity and responsiveness is required for the CHQ in this population.
<table>
<thead>
<tr>
<th>Authors</th>
<th>Design</th>
<th>Result</th>
<th>Overall Quality Assessment</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Reliability</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Novacheck (2000)</td>
<td>Inter-rater</td>
<td>ICC 0.81 &amp; 0.92</td>
<td>Good to excellent reliability for walking scale levels 6-10 only</td>
</tr>
<tr>
<td></td>
<td>Retest</td>
<td>ICC 0.92</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Validity</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Novacheck (2000)</td>
<td>Concurrent</td>
<td>r= 0.42-0.76 with a range of measures</td>
<td>Good correlations with like measures.</td>
</tr>
<tr>
<td></td>
<td>Construct</td>
<td>Lowest with O2 cost, highest with PODCI</td>
<td>Further validity testing required.</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Responsiveness requires examination</td>
</tr>
<tr>
<td><strong>Responsiveness to change</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Not examined</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

ICC = intraclass correlation coefficients
The FAQ

The psychometric properties of the FAQ are summarized in Table 3.6. Reliability of the FAQ was assessed in one study only with excellent inter-rater reliability between parent and the school (Novacheck et al 2000). The higher value was with a shorter time period between ratings. Intra-rater reliability (using parents) was also excellent despite a long mean time between ratings of 13.91 weeks.

The same study aimed to assess content, construct and concurrent validity (Novacheck et al 2000). Pearson correlation coefficients were calculated between the FAQ and the WeeFIM motor subscale, the normalcy index, energy data and PODCI (transfers and mobility and global function scales). The results were all reported together with no distinct separation of what was content, construct or concurrent validity. Correlations ranged between 0.42-0.76 with the highest between the FAQ and PODCI transfers and mobility and lowest with energy data. This study only included children who could walk (level 6-10 on the FAQ) so the results can be applied to ambulant children only. Responsiveness of the FAQ was not tested.
Table 3.7 *Psychometric properties of the FMS*

<table>
<thead>
<tr>
<th>Authors</th>
<th>Design</th>
<th>Result</th>
<th>Overall Quality Assessment</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Reliability</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Graham (2004)</td>
<td>Inter-rater</td>
<td>ICC 0.94-0.95</td>
<td>Excellent inter-rater reliability between surgeon and fellow. Further reliability testing required with broader range of raters.</td>
</tr>
<tr>
<td><strong>Validity</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Graham (2004)</td>
<td>Concurrent</td>
<td>r=0.51-0.89 lowest with oxygen cost, highest with uptime, PODCI, CHQ</td>
<td>Good concurrent validity with strong correlations with like measures. Weak evidence for discriminative validity. Other validity testing is required.</td>
</tr>
<tr>
<td></td>
<td>Discriminative</td>
<td>Differentiation of walking abilities, no formal analysis used</td>
<td></td>
</tr>
<tr>
<td><strong>Responsiveness to change</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Graham (2004)</td>
<td>Mean change after multilevel surgery</td>
<td>Detected change over the post-operative period, however used change in mean scores which is not appropriate for ordinal scales</td>
<td>Further testing of responsiveness is required.</td>
</tr>
</tbody>
</table>

ICC = intraclass correlation coefficients
The FMS

Table 3.7 summarises the psychometric properties of the FMS. The one study examining the FMS showed the scale has excellent inter-rater reliability between two surgeons and one orthopaedic research fellow (Graham et al 2004). Further reliability testing with a broader range of raters is required before reliability of the tool is fully established.

The validity of the FMS was also examined. The study aimed to investigate the content, concurrent and construct validity. Concurrent validity with other measures showed strong correlations using Spearman’s rho with Uptime, the PODCI and the CHQ. Moderate correlations existed with the Rancho scale (also called the Hoffer Scale) and weak correlations with energy expenditure. No formal statistical analysis was performed for the ability of the FMS to differentiate varying degrees of walking ability (discriminative validity); therefore the associations found are not strong evidence of this. Construct validity was examined by examining the scales ability to detect change after multilevel surgery which can be considered as responsiveness. Change over the post-operative period was detected by the scale, however the authors appear to use change in mean scores only which is not the most appropriate test for an ordinal scale.
### Table 3.8 Psychometric properties of the GMFM

<table>
<thead>
<tr>
<th>Authors</th>
<th>Design</th>
<th>Result</th>
<th>Overall Quality Assessment</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Reliability</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Bjornson (1998)</td>
<td>Retest</td>
<td>ICC 0.76-1.0</td>
<td>Overall reliability good to excellent.</td>
</tr>
<tr>
<td>Nordmark (1997)</td>
<td>Inter-rater</td>
<td>Kendall coeff. 0.77-0.88</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Retest</td>
<td>Kendall coeff 0.68</td>
<td></td>
</tr>
<tr>
<td>Russell (1989)</td>
<td>Inter-rater</td>
<td>ICC 0.92-0.99</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Retest</td>
<td>ICC 0.87-0.99</td>
<td></td>
</tr>
<tr>
<td>Russell (2000)</td>
<td>Retest</td>
<td>ICC 0.99</td>
<td></td>
</tr>
<tr>
<td>McCarthy (2002)</td>
<td>Internal consistency</td>
<td>Cronbach $\alpha = 0.99$</td>
<td></td>
</tr>
<tr>
<td><strong>Validity</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Russell (2000)</td>
<td>Content</td>
<td>Item hierarchy shown</td>
<td>Content, concurrent and discriminative validity supported.</td>
</tr>
<tr>
<td>Russell (1989)</td>
<td>Discriminative</td>
<td>Severity hierarchy shown</td>
<td></td>
</tr>
<tr>
<td>Bjornson (1998)</td>
<td>Concurrent</td>
<td>0.82, 0.64 and 0.54</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Construct</td>
<td>Significant correlation of change scores by video with GMFM (not dimension A)</td>
<td></td>
</tr>
<tr>
<td><strong>Responsiveness to Change</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Nordmark (2000)</td>
<td>Friedman test, Wilcoxon matched pairs</td>
<td>Change detected in first 6 months after SDR for total group and GMFCS II/III but not IV/V (small sample = 18)</td>
<td>Appropriate statistics used in 3 studies yet one had a small sample and another had a potential for bias in ratings. Dimensions D (standing) and E (walking, running, jumping) show responsiveness.</td>
</tr>
<tr>
<td>Russell (2005)</td>
<td>Paired t tests</td>
<td>Signif. improvement with aids and AFO vs barefoot in GMFCS III/IV for dimension D and I-IV for E</td>
<td>Other studies tended to assess change after treatment or over time rather than responsiveness of the tool.</td>
</tr>
<tr>
<td>Russell (1989)</td>
<td>ANOVA</td>
<td>More change in mild than moderate and severe</td>
<td></td>
</tr>
<tr>
<td>Russell (2000)</td>
<td>ANOVA</td>
<td>Mean change on ANOVA significant</td>
<td></td>
</tr>
<tr>
<td>Vos-vromans (2005)</td>
<td>Effect size and SRM</td>
<td>High for total score and D and E (less effect for A, B and C)</td>
<td></td>
</tr>
<tr>
<td>Wang (2006)</td>
<td>t tests and ROC</td>
<td>Both versions showed change</td>
<td></td>
</tr>
<tr>
<td>Wright (2005)</td>
<td>t tests, ES and SRM</td>
<td>Version 66 higher ES than 88 (small sample = 9)</td>
<td></td>
</tr>
</tbody>
</table>

SDR = selective dorsal rhizotomy; ES = effect size; SRM = standardized response means; ROC = receiver operating curves; ANOVA = analysis of variance; ICC = intraclass correlation coefficient
Chapter 3 – Systematic Review

The GMFM

Table 3.8 summarises the psychometric properties of the GMFM. The results showed that the GMFM is a reliable tool for children with CP. Inter-rater reliability and test-retest were high (Russell et al. 1989, Nordmark et al. 1997, Bjornson et al. 1998, Russell et al. 2000). Internal consistency was very high (McCarthy et al. 2002). Overall reliability of the GMFM was good to excellent.

The GMFM was tested for concurrent, content, construct and discriminative validity in three studies and these were all supported by the evidence (Russell et al. 1989, Bjornson et al. 1998, Russell et al. 2000). Change in GMFM-88 scores were correlated with video-based evaluations, therapists judgement and parent’s judgements, the higher correlation with the more objective measure (Russell et al. 1989). Content and discriminative validity were examined with the GMFM-66 by investigating the hierarchy of items and correlating GMFM scores with GMFCS levels (Russell et al. 2000). A hierarchical structure and interval scaling was determined through Rasch analysis and a clear relationship with milder children scoring higher and more involved children scoring lower on the GMFM. One study examined the construct validity by looking at change in GMFM-88 score over time correlated with change judged by masked video analysis (Bjornson et al. 1998). Spearman rank coefficients showed significant correlations at 12 months and 24 months post rhizotomy in all dimensions except for lying/rolling. No statistics were used however to quantify the amount of change in GMFM scores that occurred over the time period.

Responsiveness of the GMFM was examined in several studies (Russell et al. 1989, Nordmark et al. 2000, Russell et al. 2000, Russell and Gorter 2005, Vos-Vromans et al. 2005, Wright et al. 2005, Wang and Yang 2006). Initial testing of the GMFM-88 showed a significant difference existed between two time points in a “responsive” group as identified by both parent and clinician and milder children changed more than the moderate and severe group (Russell et al. 1989). Another study examined the amount of change within 12 months based on expected change using GMFCS level and age. Results of another study showed a significant difference in mean GMFM-66 score (Russell et al. 2000) with
children younger than 5 years showing more change, particularly those in
GMFCS levels I and II.

Different studies used different statistical analysis to explore responsiveness.
Effect size and standardised response mean was used to examine the
responsiveness of the GMFM-88 (Vos-Vromans et al 2005). Effect size was
high for total GMFM score and the standing and walking dimensions (D and E)
but low for dimensions A, B and C. Change scores, effect size and standardized
response means were used to assess changes following a conductive education
program resulting in a medium effect size for the 88 version but higher for the
66 version (Wright et al 2005). The sample size in this study was small (9) so
cautions is required with interpreting the results regarding the magnitude of
change.

One study examined the responsiveness of both GMFM versions (the 66 and 88)
using a therapist judgement on change as the external standard over a 3 month
period (Wang and Yang 2006). The change scores and the receiver operating
characteristic curve showed that significant differences existed in GMFM scores
for those judged as having large improvement. No significant improvement was
seen for those judged not to have changed. The sensitivity of the two versions
was similar but the 66 appeared to have higher association with therapist
judgement of change. A limitation of this study was that the same therapist
performed the judgement of change as well as the administering the GMFM.
The chance of bias from the therapists having prior knowledge of performance
on the GMFM before judging change must be considered.

Responsiveness of the GMFM-88 to detect changes in function with aids or
orthoses showed significant improvements when using aids compared to none
and when using orthoses versus barefoot in children GMFCS levels III and IV
for standing and levels I-IV for walking (Russell and Gorter 2005). However,
there is no comparable measure in this study to determine how much change
actually did occur so the study assesses change not responsiveness. Sensitivity
of the GMFM-88 and PEDI was examined for a small sample of children at 6
and 12 months following rhizotomy using to see at what stage change occurred
(Nordmark et al 2000). The GMFM detected significant change in the group over time in the second 6 months after rhizotomy, these changes seen in moderate (GMFCS II and III) but not severely involved children (IV and V).

Overall responsiveness was tested well in two studies and showed that dimensions D and E of the GMFM are responsive (Vos-Vromans et al 2005, Wright et al 2005), despite one of these studies using a small sample size. The other studies do show changes after treatments or interventions; however they appear to be examining change after interventions rather than responsiveness of the tool. This issue will be elaborated on in the discussion and in Chapter Six.
Table 3.9 Psychometric properties of the PEDI

<table>
<thead>
<tr>
<th>Authors</th>
<th>Design</th>
<th>Result</th>
<th>Overall Quality assessment</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Reliability</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Nichols (1996)</td>
<td>Inter-rater</td>
<td>ICC 0.15-0.95</td>
<td>Very high internal consistency. Reliability of mobility domain</td>
</tr>
<tr>
<td></td>
<td>Retest</td>
<td>ICC 0.67-1.0</td>
<td>good to excellent yet lower for self-care and social function</td>
</tr>
<tr>
<td>Wright (1993)</td>
<td>Inter-rater</td>
<td>ICC 0.72-0.85</td>
<td>domains.</td>
</tr>
<tr>
<td></td>
<td>Retest</td>
<td>ICC &gt;0.95</td>
<td></td>
</tr>
<tr>
<td>McCarthy (2002)</td>
<td>Internal</td>
<td>Cronbach α 0.98-0.99</td>
<td></td>
</tr>
<tr>
<td></td>
<td>consistency</td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Validity</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Nichols (1996)</td>
<td>Concurrent</td>
<td>r=0.24-0.95 (PDMS)</td>
<td>Concurrent validity generally very good with high correlations</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>for related domains and lower for unrelated domains.</td>
</tr>
<tr>
<td>Wright (1993)</td>
<td>Concurrent</td>
<td>r=0.59-0.85 (GMFM) – inadequate details in method and results</td>
<td>Good discriminative ability of tool for topography of CP</td>
</tr>
<tr>
<td>McCarthy (2002)</td>
<td>Discriminative</td>
<td>r=0.91 (GMFM), 0.88 (PODCI), 0.74 (CHQ) multivariate ANOVA - significant differences for hemiplegia, diplegia and quadriplegia</td>
<td>Requires further testing in this population for other forms of validity e.g. content, construct</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Authors</th>
<th>Statistical Analysis</th>
<th>Result</th>
<th>Overall Quality Assessment</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Responsiveness to Change</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Nordmark (2000)</td>
<td>Friedman test, Wilcoxon matched pairs</td>
<td>Detected change in 1st 6 months after SDR for both moderately and severely involved (small sample = 18)</td>
<td>Good statistics used with tool showing responsiveness as detected by parents in one study, inconclusive in one study and third study assessed change after SDR rather than responsiveness of the tool.</td>
</tr>
<tr>
<td>Vos-vromans (2005)</td>
<td>Effect size and SRM</td>
<td>Every dimension high vs parent change score</td>
<td></td>
</tr>
<tr>
<td>Wright (2005)</td>
<td>Effect size and SRM</td>
<td>Medium ES for PEDI part II and small for part I after CE. Small sample size (9)</td>
<td></td>
</tr>
</tbody>
</table>

SRM = standardized response mean; ICC = intraclass correlation coefficient; SDR = selective dorsal rhizotomy; ANOVA = analysis of variance; CE = conductive education.
Chapter 3 – Systematic Review

The PEDI

The psychometric properties of the PEDI are summarized in Table 3.9. The inter-rater and retest reliability was found to be high in the mobility domain and total score (Wright and Boschen 1993, Nichols and Case-Smith 1996). Inter-rater reliability was low in the self-care domain which may be due to differences in familiarity of the child’s self-care function between the responding parents and therapists (Nichols and Case-Smith 1996). Internal consistency was excellent (McCarthy et al 2002).

Validity testing for the PEDI focused on concurrent validity. Strong correlations, using Spearman’s rho, of the mobility and self-care domains were shown with the GMFM and like domains of the PODCI and CHQ (McCarthy et al 2002). Correlations were low in unrelated domains. The PEDI total score and mobility score had slightly weaker correlations with the GMFM (Wright and Boschen 1993). This study did not detail well the methodology or results; therefore it is difficult to make a clear judgement. Moderate to high correlations using Pearson’s r were shown of the PEDI with the related PDMS domains (Nichols and Case-Smith 1996). Examination of discriminative validity of the PEDI mobility domain showed differences in physical health using topographical distribution of CP (McCarthy et al 2002).

Responsiveness of the PEDI was examined in three studies (Nordmark et al 2000, Vos-Vromans et al 2005, Wright et al 2005). Correlation of change on the PEDI versus parent change score was high on every dimension (Vos-Vromans et al 2005). The sensitivity of the PEDI for a small sample of children following rhizotomy showed it was able to detect change within the first 6 months (earlier than the GMFM) for both moderately and severely involved children (Nordmark et al 2000). The responsiveness of the PEDI following a conductive education program showed small and medium effects sizes (Wright et al 2005), however the sample size in this study limits the ability to assess the magnitude of change and hence responsiveness. Generally responsiveness of the PEDI was supported by the evidence although one study had a small sample and one appears to be assessing change after treatment rather than responsiveness of the tool.
Table 3.10 Psychometric properties of the PODCI

<table>
<thead>
<tr>
<th>Authors</th>
<th>Design</th>
<th>Result</th>
<th>Overall Quality assessment</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Reliability</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Daltroy (1998)</td>
<td>Inter-respondent</td>
<td>r=0.45-0.87 (parent vs child), lowest for happy &amp; satisfied expectations, highest for physical domain</td>
<td>High internal consistency and moderate to good retest reliability. Variable inter-respondent (child vs parent) reliability for subjective domains.</td>
</tr>
<tr>
<td>McCarthy (2002)</td>
<td>Retest</td>
<td>r=0.71-0.97</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Internal consistency</td>
<td>Cronbach α&gt;0.80 (parent) &gt;0.76 (child)</td>
<td></td>
</tr>
<tr>
<td><strong>Validity</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Vitale (2005)</td>
<td>Concurrent</td>
<td>0.28-0.61 (clinicians subjective ratings)</td>
<td>Only concurrent and discriminative validity tested.</td>
</tr>
<tr>
<td></td>
<td>Discriminative</td>
<td>Physical function scales detect differences between topography of CP</td>
<td>Concurrent validity is good for related physical domains in 3 studies (McCarthy, Pencharz, Damiano,) but other studies use subjective clinician ratings that are not validated.</td>
</tr>
<tr>
<td>Vitale (2001)</td>
<td>Concurrent</td>
<td>0.29-0.78 (clinicians subjective ratings)</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Discriminative</td>
<td>CP scored lower than “normals”</td>
<td></td>
</tr>
<tr>
<td>Daltroy (1998)</td>
<td>Concurrent</td>
<td>0.12-0.76 (clinician ratings )</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Discriminative</td>
<td>Can detect aspects of extremity involvement</td>
<td></td>
</tr>
<tr>
<td>Pencharz (2001)</td>
<td>Concurrent</td>
<td>0.60 (CHQ) 0.80 (ASK)</td>
<td>One shows tool has discriminative ability for ambulation. Other studies provide inconclusive evidence of discriminative ability indicating further testing is required. Other forms of validity testing are required in CP.</td>
</tr>
<tr>
<td></td>
<td>Discriminative</td>
<td>Significantly discriminates Hoffer ambulation level</td>
<td></td>
</tr>
<tr>
<td>McCarthy (2002)</td>
<td>Concurrent</td>
<td>0.88 (GMFM) 0.50-0.81 (PEDI)</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Discriminative</td>
<td>0.68-0.81 (CHQ)</td>
<td></td>
</tr>
<tr>
<td>Damiano (2005)</td>
<td>Concurrent</td>
<td>0.56-0.94 (GMFM)</td>
<td></td>
</tr>
<tr>
<td><strong>Responsiveness to change</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Daltroy (1998)</td>
<td>Change score vs subjective transition score</td>
<td>Low correlations between parents and physicians of change - subjective method and missing statistics in study.</td>
<td>Insufficient evidence to support responsiveness to change in CP. Further work is required.</td>
</tr>
<tr>
<td>Damiano (2005)</td>
<td>ANOVA pre and post treatment</td>
<td>Change seen in 1 group only – appears to test change after treatment, not responsiveness</td>
<td></td>
</tr>
</tbody>
</table>

ANOVA = analysis of variance
Table 3.10 displays the psychometric properties of the PODCI. The reliability of the PODCI has not been tested extensively. Internal consistency was found to be high in two studies for the parent and adolescent version (Daltroy et al 1998, McCarthy et al 2002). Correlations for test-retest reliability were good and variable for inter-rater reliability between the parents and their child (Daltroy et al 1998). The lowest correlations were for “happy and satisfied” and “expectations”. The highest were for “physical function and sports” and “transfers and mobility”. These areas of disagreement may reflect the different perspectives of the child and parent on the effect of the disability in subjective domains.

Concurrent and discriminative validity were tested in six studies yet other forms of validity were not examined. Concurrent validity testing resulted in variable results. One study investigating the “uniqueness” of the PODCI appears to be testing concurrent validity by correlating it to the GMFM-88 using Pearson’s r (Damiano et al 2005). Correlations were variable with the strongest for PODCI transfers and mobility with total GMFM score and the standing dimension D and the lowest for dimensions A, B and C with sports and physical function. Two studies correlated PODCI scores to subjective ratings from the clinicians/physicians treating the children (Vitale et al 2001, Vitale et al 2005). Correlations ranged from low in the pain/comfort domain to high in the sports and physical function domain (Vitale et al 2001, Vitale et al 2005). Using clinician ratings that are subjective and not validated weakens the evidence of validity of the PODCI. One of these studies correlated the PODCI to the CHQ with the expectation that similar domains would correlate highly, however the data was not presented in the article (Vitale et al 2005).

High correlations using Spearman’s rho were shown between the transfers and mobility domain of the PODCI with the GMFM, the PEDI and the CHQ like domains (McCarthy et al 2002). Lower correlations were found for unlike domains as predicted. Correlations between the PODCI and CHQ were found to be moderate to good and high with the ASK (Pencharz et al 2001). Correlations between physician ratings of global function and parent scales with the PODCI
were variable using Pearson’s r (Daltroy et al 1998). The lowest were in the domains of “happy and satisfied”, “expectations” and “comfort” and the highest in the physical function areas. Similar results were obtained with the adolescent version. Overall the concurrent validity of the PODCI is supported from the studies.

Discriminative validity was also examined in several studies (Daltroy et al 1998, Pencharz et al 2001, Vitale et al 2001, McCarthy et al 2002, Vitale et al 2005). One study examined whether the PODCI could differentiate differences between subjects with and without lower extremity involvement and with and without upper extremity involvement (Daltroy et al 1998). As could be expected, the PODCI upper extremity function scale appeared to discriminate with and without upper extremity problems and the physical function and sports detected with and without lower extremity involvement the best. Other studies of discriminative validity show a strong ability for it to discriminate ambulation level using the Hoffer scale (Pencharz et al 2001). The transfer and mobility scale can significantly detect differences between hemiplegia, diplegia and quadriplegia (McCarthy et al 2002). One study described the mean scores in relation to topographical distribution of CP and the physical function scales detected differences between hemiplegia, diplegia and quadriplegia (Vitale et al 2005). Another study stated that children with CP scored lower on the PODCI than “normals” and those with more comorbidities scored lower, however there is no statistical evidence to support this (Vitale et al 2001). Other forms of validity testing need to be examined before the tool is fully validated in this population.

Responsiveness of the PODCI was examined in two studies (Daltroy et al 1998, Damiano et al 2005). One compared change pre and post treatment in PODCI and GMFM (Damiano et al 2005). Compared with a non-matched group of children who had no intervention, changes were seen in GMFM for the groups who had muscle/tendon surgery and rhizotomy. In the PODCI global function scale only the muscle/tendon surgery group showed change. Neither showed changes in the intrathecal Baclofen group. It is difficult to draw strong conclusions from this study as the control group was not matched to the surgical
groups. Also the study appears to be examining change after treatment rather than responsiveness of the tool. In another study the correlation of change scores with transition scores as rated by parents and physicians were low (Daltroy et al 1998). t tests were used to examine the responsiveness of the PODCI with the authors stating it is sensitive to change, particularly the global scale, however there were no p values given and it is difficult to make a clear statement on the results.
Table 3.11 *Psychometric properties of the WeeFIM*

<table>
<thead>
<tr>
<th>Authors</th>
<th>Design</th>
<th>Result</th>
<th>Overall Quality Assessment</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Reliability</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Ottenbacher (1997)</td>
<td>Inter-rater</td>
<td>ICC 0.73-0.97</td>
<td>Good to excellent reliability. Equivalence reliability for different methods of administration shows can be administered by direct observation or over the telephone</td>
</tr>
<tr>
<td>Ottenbacher (1996)</td>
<td>Retest</td>
<td>ICC 0.85-0.98</td>
<td></td>
</tr>
<tr>
<td>Sperle (1996)</td>
<td>Equivalence</td>
<td>t tests – no difference in method of administration</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Retest (stability)</td>
<td>ICC &gt;0.90</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Equivalence</td>
<td>t tests – no difference in method of administration</td>
<td></td>
</tr>
<tr>
<td><strong>Validity</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Chen (2005)</td>
<td>Construct</td>
<td>Differences in item difficulty across ages 2 distinct dimensions (motor &amp; cognitive)</td>
<td>Insufficient evidence to support all forms of validity from these studies. Further work is required in CP.</td>
</tr>
<tr>
<td>Msall (1994)</td>
<td>Content</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Discriminative</td>
<td>Higher scores hemiplegia &amp; diplegia than quadriplegia (weak statistical analysis)</td>
<td></td>
</tr>
<tr>
<td><strong>Responsiveness to change</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>McAuliffe (1998)</td>
<td>Mean change after surgery</td>
<td>Variability in amount of change seen.</td>
<td>Extensive and thorough statistics used in one study showing good responsiveness (Ottenbacher). Other study assessed change after surgery, not responsiveness of the tool.</td>
</tr>
<tr>
<td>Ottenbacher (2000)</td>
<td>Reliability change index, proportional change, ES, SRM, t tests</td>
<td>All show significant change over time (3 time points in 1 year)</td>
<td></td>
</tr>
</tbody>
</table>

ICC = intraclass correlation coefficient; ES = effect size; SRM = standardized response means
The WeeFIM

Table 3.11 summarises the psychometric properties of the WeeFIM. It was found to have very good inter-rater and retest reliability (Ottenbacher et al 1997). This study also found that ICC values for a shorter interval between ratings (3-7 days) were generally higher than for the long interval (20-30 days). Equivalence reliability was tested in two studies where the authors compared administration of the WeeFIM by direct observation with parental interview (Sperle et al 1997) and direct observation and interview with over the phone interview (Ottenbacher et al 1996). There were no significant differences found between methods of administration in either study. Test-retest (stability) of repeated administration over 7-14 days was excellent (Ottenbacher et al 1996).

Validity of the WeeFIM in the CP population has not been thoroughly assessed. One study vaguely explored WeeFIM scores against topography of CP (discriminative validity) and found that mean WeeFIM was higher for hemiplegia and diplegia as compared to children with quadriplegia (Msall et al 1994). The analysis of this was not extensive and the study provides insufficient information to make any strong statements about the discriminative ability of the tool. The WeeFIM was investigated for item fit, dimensionality and motor item difficulty in three different age groups (Chen et al 2005). Rasch analysis found two distinct dimensions (motor and cognitive). Order of motor item difficulty varied across age groups some of which can essentially be explained by the developmental age and stage of the child.

Responsiveness of the WeeFIM was examined in two studies (McAuliffe et al 1998, Ottenbacher et al 2000). To assess change after orthopaedic surgery and physiotherapy, mean change was used to measure difference from admission to phone follow-up in 20 children with CP (McAuliffe et al 1998). For the mobility items there was a lot of variability in the amount of change in the sample with the greatest change in children with diplegia (compared with quadriplegia). The study did not use a comparable measure to detect if change actually occurred; hence it appears to be examining change after surgery not responsiveness of the tool. A well controlled study used extensive statistical measures to assess the
ability of the WeeFIM to detect change in functional status in a large sample of children assessed three times over a 1 year period (Ottenbacher et al 2000). All statistical methods showed statistically significant or reliable changes over time. Effect size was moderate for the motor subscale and total score and small for transfers and locomotion. The results of this study indicate the WeeFIM is a responsive measure.

3.4. Discussion and main review findings

3.4.1. Psychometrics
The main finding from this systematic review was that some tools have been extensively tested and performed well while others still require further testing in this population before they can be classified as robust measurement tools. The GMFM and the ASK performed the best overall with sound psychometric properties in most areas. In general, the reliability of the eight tools included was found to be adequate with most tools showing good to excellent reliability. Validity testing results of all tools except the GMFM and ASK were variable with more work in specific areas required. Concurrent validity of the tools was tested extensively with good results, an area that has been covered amply in the literature with studies excluded from this review exploring relationships among outcome measures (Drouin et al 1996, Ottenbacher et al 1999, Azualia et al 2000, Schneider et al 2001, Tervo et al 2002, Abel et al 2003, Oeffinger et al 2004). Further research is required examining the construct validity of the tools. Discriminative validity also requires more investigation, particularly the ability of the tools to discriminate GMFCS levels (Palisano et al 1997) as this is now the preferred classification system of severity of motor impairment for children with CP. This has been addressed more recently for the GMFM, PODCI, WeeFIM and FAQ (Bagley et al 2007).

This review examined evaluative measures. These need to be responsive to change in order to be valid (Rosenbaum et al 1990). This has been examined more extensively since the Ketelaar review which found that only the GMFM and PEDI showed responsiveness (Ketelaar et al 1998). Confusion in the literature regarding responsiveness arises from a lack of distinction between
responsiveness defined as the effect of treatment and responsiveness defined as the correlation of changes in the instrument with changes in other measures (Terwee et al 2003). The studies included in this review examining responsiveness to change used variable methods of analysis. As a result some of the studies examined treatment effects without providing information about the ability of the instrument to detect clinically important change. Often it is not known the change in score that constitutes a clinically important difference hence the use of indices estimating responsiveness (Guyatt et al 1987). Some authors used several different measures due to lack of consensus on the single best procedure for determining responsiveness (Ottenbacher et al 2000, Young et al 2000, Vos-Vromans et al 2005). Responsiveness is perhaps not a separate measurement property with current measures assessing longitudinal validity or the magnitude of treatment effect (Terwee et al 2003). These concepts will be explored further in Chapter Six. For this review responsiveness was examined well for the ASK, GMFM, PEDI and WeeFIM. It was not examined at all for the CHQ or the FAQ despite the authors of the FAQ stating it can be used to measure change.

One of the difficulties experienced with conducting this review was examining and comparing measures when some are multidimensional, such as the CHQ and PODCI. This review focused on activity limitation measures. In many of the studies the domains were not separated out clearly and it was difficult to compare or examine the activity related domains in isolation.

### 3.4.2. Clinical Use

The main theme to emerge from examining the clinical utility of the tools was that each has strengths and weaknesses. Some tools take longer to administer and are more comprehensive (e.g. PEDI and GMFM) while others are quick to administer and more practical (e.g. ASK, FAQ and FMS). The WeeFIM and PEDI include similar domains, have sound psychometric properties and correlate well on similar constructs (Ziviani et al 2001). The WeeFIM has a minimum data set and therefore is more time efficient. It can be utilized when the detail obtained from the PEDI is not required. Some of the tools are specific to CP (e.g. GMFM, FMS) and some are specific to walking or mobility (e.g.
FMS, FAQ). The two health-related quality of life measures (e.g. CHQ, PODCI) give a broader perspective but lose some specificity and have floor and ceiling effects. Different tools utilize reporting from the parent, the child or the clinician therefore giving varied perspectives on the child’s function. These perspectives can be vastly different depending on who reports. This issue will be elaborated on further in the grand discussion. In general, no one tool can cover all domains of the ICF effectively and a range of tools is required, some of which are better suited for clinical use and others for research.

3.4.3. Environmental context and other factors

One aim of this systematic review was to determine if the tools incorporated different environmental settings and different assistive devices. All apart from the GMFM incorporate different environmental settings, however only the FMS differentiates the difference in function between the home, the school and the community. These are all important settings to children with CP. The FMS and CHQ are the only tools to measure activity specific to the school environment. The FMS is the only tool that differentiates the various assistive devices a child with CP might use for activity, particularly mobility. Also of interest was whether the tools measure performance or capability. Performance measures are stated to be more reflective of community function and therefore more relevant to children (Young et al 1996). All of the tools except the GMFM measure performance with the ASK and PEDI incorporating both.

3.4.4. Limitations

It is important to discuss the limitations of this review. The inclusion and exclusion criteria were extensive and focused. By restricting it to evaluative tools a number of tools that are used for discriminative purposes were excluded. In addition, by restricting it to tools used for children with CP and to studies that must have children with CP in their sample, a number of studies were excluded. Excluded were studies examining the psychometric properties of the ASK in children with musculoskeletal disability without CP (Young et al 1995) and children without musculoskeletal disability (Plint et al 2003) and the WeeFIM in children without developmental delays (Msall et al 1994). Studies on unimpaired children using the PODCI (Haynes and Sullivan 2001) and the CHQ
(Waters et al 2000, Waters et al 2000, Raat et al 2005) were also excluded on this basis. Several studies were excluded on the basis that they did not obviously include children with CP in their sample. This essentially involved studies on the validity and reliability of the PEDI (Feldman et al 1990, Ziviani et al 2001, Berg et al 2004). These studies add to evidence of psychometric properties of the tools. Although studies were excluded, it was necessary to have tight methodology because the area of measurement tools in children with CP is extremely broad. In order to examine it properly a number of well-designed and thorough reviews on different aspects are required.

3.5. Conclusion and recommendations

This systematic review identified eight evaluative outcome measures that assess activity limitations in children with CP. Each examines different dimensions of activity limitation including mobility, gross motor function, self-care and cognitive and social function. The designs of the 29 studies selected were critically analysed in order to evaluate psychometric properties. Overall, the ASK and GMFM showed the most robust psychometric properties. The reliability of all tools was good, although further examination of validity and responsiveness to change is required for the CHQ, FAQ, FMS and PODCI in this population. The PEDI and WeeFIM require further examination of some aspects of validity.

All tools measure performance except for the GMFM, which measures capability. The PEDI and ASK consider both. Different environmental settings are incorporated within all tools except for the GMFM. The FMS, which is specific to mobility, is the only tool that addresses different environmental contexts and different assistance required within those. However, the FMS also requires further psychometric testing, such as inter-rater reliability for a range of raters, better designed discriminative, concurrent and construct validity testing and further investigation of responsiveness.

The feasibility and clinical utility of the measures were evaluated showing that each has a different purpose for application in CP. Because no single measure adequately addresses all aspects of the ICF, it is advisable in the clinical
decision making process for the selection of the tool to be based on established psychometric properties of the tool together with the goals of the measurement.
CHAPTER FOUR: DEVELOPMENT OF THE FUNCTIONAL MOBILITY SCALE AND PILOT STUDY

4.1. Introduction

This chapter provides the background on the original development of the FMS and reports on an early pilot study that investigates the clinical applicability of the FMS. The systematic review in Chapter Two showed that, excluding the FMS, no activity limitation measures considered the range of assistive devices that a child with CP might use during mobility tasks. Although many other measures incorporated different environmental settings, the FMS was the only one to differentiate the difference in function between the settings. Prior to development of the FMS, existing measures did not address mobility in children with CP in different environments, particularly with respect to the different levels of assistance required.

It is observed in the clinical setting that children with CP use different levels of assistance for their mobility and this can impact on their long term function and participation. In settings such as the gait laboratory it is important to be able to quantify the precise level of support children require. This is particularly important before and after interventions where clinical experience reveals that assistive device requirements change over the post-operative rehabilitation period. This was illustrated in a study before the development of the FMS that examined the effects of proximal femoral varus rotation osteotomies on the gait of children with CP (Murray-Weir et al 2003). The study recorded functional walking status of the children; however the authors stated that not all changes in ambulation status were reflected in their method of reporting, particularly changes of assistive devices to less cumbersome ones. It was also known clinically that some interventions increased dependency initially but the question of whether the children were permanently adversely affected needed to be addressed. The FMS was devised to fulfil this role by providing a new and unique outcome measure specific to mobility.
Development of the FMS aimed to produce a simple and clinically feasible evaluative measure for children with CP specific to mobility. Evaluative measures are used to “measure the magnitude of longitudinal change in an individual or group on the dimension of interest” (Kirschner and Guyatt 1985). Using the ICF, mobility is conceptualized as part of the activities and participation component and can be described as “moving by changing body position or location or by transferring from one place to another, by carrying, moving or manipulating objects, by walking, running or climbing, and by using various forms of transportation” (page 135) (World Health Organisation 2001).

The aim of the FMS was to primarily address walking (d450 ICF), moving around in different locations (d460) and moving around using equipment (d465). This includes walking independently in various places as well as using equipment such as walkers and wheelchairs yet excludes forms of transportation and driving.

Mobility, thus defined, is important because it enables children to move around within their homes, schools and other community settings. This enables them to participate as fully as possible with their family and peers and gain satisfaction from such participation. It is important that there are clinically feasible and psychometrically sound tools available to measure mobility.

4.2. Initial development

The FMS was developed by the group working in the Hugh Williamson Gait Laboratory of the Royal Children’s Hospital, Melbourne Australia. This group consisted of two orthopaedic surgeons, one orthopaedic fellow, and two senior physiotherapists with extensive clinical experience in children with CP. The author of this thesis was one of those senior physiotherapists. The development and scaling of items was based on clinical observation and experience. The team developed the FMS prior to the commencement of this thesis. The scale had been introduced in the clinical setting and a preliminary study performed (Graham et al 2004). The aim of this thesis was to validate it further to ensure it could be considered a reliable and valid outcome measure for use by clinicians with children with CP.
There are two reasons for wanting to develop a new instrument. Firstly, the construct is a new one and no scale exists which measures it. A second reason is if people are dissatisfied with the existing tools and feel some key aspect of the construct is omitted (Streiner and Norman 2003). The results of the systematic review in Chapter Three showed that existing tools measuring physical function of children with CP did not adequately consider different assistance children may need in different environmental settings. There are few scales that concentrate on mobility in isolation. It is more usually reported as a subsection of a larger inventory. The FMS was developed to fulfil this need in outcome measurement for children with CP.

4.2.1. Target population

The target population for the FMS was children with CP, GMFCS levels I to IV, aged 4 to 18 years. It was aimed for children GMFCS levels I to IV because the intent of the scale was to measure mobility. The FMS focuses on walking mobility with one category for the use of wheelchairs. Children in GMFCS levels I-IV have the ability to walk or mobilise in a wheelchair. Children classified as level V have severe physical impairment with no means of independent mobility and are transported by adults (Palisano et al 1997). The FMS would have significant floor effects if children in level V were incorporated.

The lower age limit was set at 4 years based on evidence of gross motor development and the development of walking. Gait studies on 186 normally developing children showed that a mature gait pattern, including temperospatial parameters, is established at the age of 3 years (Sutherland et al 1980). Many children with CP begin walking later than typically developing children. Research has shown that walking can be achieved up to the age of 9 years (Campos da Paz Jr et al 1994, Bottos et al 1995, Fedrizzi et al 2000). These studies also show that children with hemiplegia and some with diplegia will achieve walking at ages as young as 2-5 years. Motor development curves have been created using longitudinal observations of children with CP to describe patterns of gross motor development (Rosenbaum et al 2002). Based on these curves, 90% of the potential GMFM-66 score is reached by age 3.5 for children
Chapter 4 – Development of the FMS

GMFCS level IV, by age 3.7 for children level III, by age 4.4 in level II and by age 4.8 in level I. This suggests that children are still developing motor abilities before these ages and would potentially have inconsistent mobility before the age of 4 years. The upper age limit was set at 18 years because the FMS was developed for children and adolescents with CP. Also, at the centre were the scale was developed children tend to be transitioned to adult services at age 18.

4.2.2. Generation of items

One of the major aims in developing the scale was to represent the different environmental settings that are relevant to children in order to differentiate function in each. These environments include the home, school and wider community. Chapter Two examined the studies that have evaluated the effect of environmental setting on mobility methods of children with CP and showed that children employ different mobility methods in different environments (Palisano et al 2003, Tieman et al 2004).

Three items were developed and assigned distances in order to represent each environmental setting;

i. 5 metres – to represent the home setting
ii. 50 metres – to represent the school setting
iii. 500 metres – to represent the community setting

The numbers were set as a guide for clinicians administering the tool, not as an exact distance.

4.2.3. Scaling of items

The scaling of the three items involved the theoretical construct of incorporating the range of assistive devices available that were most commonly used for ambulation or mobility in Australian children with CP. A six point ordinal scale from 1-6 was developed depending on the amount of assistance required from the most to the least amount of assistance. The six levels were;

1. child uses a wheelchair (or wheeled devices) for mobility
2. child uses a walker such a posterior walker or an anterior walker
3. child uses forearm or Canadian crutches
4. child uses one or two single point sticks
5. child can walk independently on level surfaces without assistive devices
6. child can walk independently on all surfaces without assistive devices

The order of scaling was deliberately chosen to be the reverse order of the GMFCS to minimise confusion between the two and to ensure that clinicians thought carefully when rating children. Because the FMS primarily rates independent mobility, walkers that provide large amounts of support where the child requires adult assistance to be strapped in were excluded. Each item of the scale represents different environments and provides unique information. For this reason, the items are considered separate and are not weighted or summed to produce one score.

4.3. Modifications to the FMS

The primary author of this thesis (AH) took on sole responsibility for clinical implementation of the FMS and developed the scale further. This occurred in the initial stages of this doctoral candidature following initial clinical use of the scale and feedback from clinicians. Further development and modifications were made as described below.

4.3.1. Clarification of purpose

The original description of the FMS (Graham et al 2004) made no reference to whether it was designed to measure performance or capability. As explored in Chapters Two and Three, performance measures are more reflective of everyday function and are more relevant to the children and their families (Young et al 1996). To ensure that the FMS measured performance rather than capability, a brochure was developed which included instructions on how to administer the scale to obtain a performance rating. One of these brochures can be found at the back of this thesis.

4.3.2. Extra scaling categories

On feedback from therapists administering the FMS, two more categories for scoring were added including:

i. C – for crawling
ii. N – for does not do
Chapter 4 – Development of the FMS

Initial use discovered that a number of parents reported that children crawl at home for independent mobility. The “C” was added for this reason. There were also occasional reports that some children with CP never ventured anywhere for distances of 500 metres other than in a car. Some parents reported that they did not take their children with CP to shopping centres and other such places. Therefore an “N” was designated as an option for the 500 metre distance for these instances. Figure 4.1 shows the FMS items and scaling.

Figure 4.1 The Functional Mobility Scale (FMS)
4.3.3. Method of administration

Originally it was stated that parents could complete the FMS themselves (Graham et al 2004). After trial use it was decided that to obtain a self or parent report of mobility that reflected performance and not capability, administration by the clinicians asking questions of the children or parents was preferred. The FMS therefore is a clinician administered child or parent report measure of performance.

4.3.4. Educational material

Early clinical experience showed considerable variability in the administration of the test and interpretation of scale categories. Educational material was developed to delineate these explicitly. A brochure of the FMS was produced for distribution to clinicians. This included information on the reasoning behind the scale’s development and guidelines for administration to ensure the rating obtained reflects performance rather than capability. The brochure included qualifiers to help distinguish a child’s function between levels and sample questions and clinical examples. Also developed was a pocket sized version of the scale for clinicians to carry with them while assessing children in the clinical setting such as clinics, wards and gait laboratories. These are both located inside the back cover of this thesis.

4.5. Administration and feasibility

4.5.1. Administration

The FMS is designed to be administered by physiotherapists, orthopaedic surgeons and fellows, physicians and paediatricians who assess and manage children with CP. Administration of the FMS requires the clinician to ask a few questions of the child or parents regarding their “usual” methods of moving around within the three environments. It is therefore based on clinician interpretation of self report and not direct observation of the child. Depending on the replies given, the clinician then assigns a number from 1-6 depending on the assistance required for each distance that represents each environmental setting. For example, if a child uses crutches indoors at home, a walker to go from class to class at school and a wheelchair for longer distances such as the
shopping centre, they would be rated as 3, 2, 1. If a child is able to walk around home without assistive devices but only on level surfaces and uses single point sticks at school and a walker for the shopping centre, they would be rated as 5, 4, 2.

4.5.2. Clinical feasibility

Feasibility includes the time needed to administer the measurement as well as the equipment and training required (Dekker et al 2005). In developing the FMS the aim was to produce a measure that was not time consuming to administer, nor require expensive equipment and extensive training. It was intended to be an easily accessible clinical tool for utility in everyday clinical situations.

The FMS can be administered in less than 10 minutes. Because it is child or parent report and not direct observation, no equipment is required for administration. For this reason it has the potential to be administered over the telephone. Training requirements are minimal. The brochure provides the information necessary for the administering clinicians and it is recommended that this is read before utilizing the FMS. There are no formal training requirements or testing of clinicians in order to successfully use the tool. The burden on clinicians using the tool is therefore small.

4.6. Potential utility of the FMS

The FMS could potentially be used to document functional mobility status of children with CP. Used in conjunction with the GMFCS a very good overall picture of a child can be obtained. For example, a child with CP who is classified as GMFCS level III and rated 2, 2, 1 on the FMS indicates that the child requires assistive devices for ambulation (GMFCS III) and the devices required for mobility are a walker at home (2) and at school (2) and a wheelchair for longer distances (1).

Because the FMS is quick and easy to administer it can be utilized by different clinicians in many different clinical settings. It therefore has the potential to be used in routine clinical practice as an outcome measure for activity in children with CP. However, because it is specific to mobility, it provides information on
one aspect of activity only. In order to provide a complete assessment of a child with CP a range of measures is required. Clinical utility of the FMS will be further explored in Chapter Nine following the outcomes of the investigations in this thesis.

The FMS has been developed as an evaluative measure. It has the potential to measure change over time in children with CP and therefore track progress. It also can be used to assess the effect of interventions such as orthopaedic surgery, spasticity management and physiotherapy by measuring change before and after the interventions. The FMS was initially developed because there were no tools available that could quantify the changes in assistive devices children with CP used throughout the rehabilitation period following major surgical interventions. To investigate the potential of the FMS to track change following major surgical interventions, an initial pilot study was performed studying a group of children following single event multilevel surgery (SEMLS). The details are outlined below.

4.7. Pilot Study

4.7.1. Introduction

This pilot investigation examined the ability of the FMS to detect change following SEMLS in children with CP. This was an early small investigation to determine how useful the FMS was at quantifying mobility in the children for which the scale was originally developed. From this early work, future research questions for examining the psychometric properties and clinical utility of the FMS would be developed and investigated. The research questions formed as a result of this pilot work are the subject matter for the rest of the thesis.

At the centre where this study was performed, the correction of musculoskeletal deformities producing gait deviations in children with CP is usually performed in one session (Bache et al 2003). SEMLS refers to the correction of all orthopaedic deformities in one session (Graham and Selber 2003) and can be defined as at least two orthopaedic procedures at different anatomical sites in each limb, that is, a minimum of four procedures (Pirpiris et al 2003). In order
to determine the effects of major interventions such as SEMLS, there is a need for tools that can accurately measure change over time in activities such as mobility. The unique feature of the FMS being able to distinguish different assistive devices is very important, particularly for monitoring post-surgical progress when, for instance, progression from a walker to crutches represents a functional improvement.

The major research question for this investigation was whether the FMS could detect changes in mobility in children with CP following SEMLS. It was hypothesized that the FMS would show change, both deterioration and improvement, at set post-operative time points.

4.7.2. Method

A retrospective study was conducted of the gait laboratory records and videos of a cohort of children with CP who had undergone SEMLS. The study was approved by the Ethics in Human Research Committee of the Royal Children’s Hospital (reference number 26086A) and The University of Melbourne Human Ethics Research Committee (0606057). Written consent was obtained from each participating child’s parent or legal guardian.

4.7.2.1 Participants

The participants were a consecutive sample of children with CP recruited from the Hugh Williamson Gait Laboratory of the Royal Children’s Hospital, a tertiary care hospital in Melbourne, Australia. Included were children with CP aged 4-18 years at the time of surgery and who had SEMLS (planned on the basis of a three dimensional gait analysis) between 1997 and 2002. A necessary criterion for inclusion was attendance of the children at the Hugh Williamson Gait Laboratory for their pre-operative and routine post-operative assessments for a complete data set to be collected. A complete data set consisted of FMS scores and GMFCS ratings pre-operatively and at 3, 6, 9 and 12 months post-operatively. Excluded were children with a diagnosis other than CP, children less than four years and older than 18 years of age and children with incomplete follow-up. Four children were excluded because of incomplete data for the first 12 months post-operatively due to non-attendance at the laboratory.
4.7.2.2 Procedure

The gait laboratory records and video recordings of the included children were reviewed by the principal investigator (AH). The FMS rating and Gross Motor Function Classification System (GMFCS) level for each child were recorded from their assessments performed pre-operatively and then at 3, 6, 9, 12 and wherever possible 24 months post-operatively.

4.7.2.3 Data analysis

Odds ratios were calculated using ordinal logistic regression models (Agresti 2002) for FMS ratings at 5, 50 and 500 metres to quantify the change from pre-operative status to each post-operative time. Odds ratio less than 1 indicated lower FMS ratings on the average (children worse) at a post-operative time than pre-operatively; odds ratios greater than 1 indicated higher FMS ratings (children improved) at a post-operative time. Standard errors for significance tests of these odds ratios and for the construction of 95% confidence intervals for true change in FMS were calculated using the information sandwich formula to account for the repeat measurements on the same children over time. All analyses were performed using Stata [StataCorp 2005 Stata statistical software Release 9.0. College Station; TX].

4.7.3. Results

4.7.3.1 Descriptive results

There were 66 participants in the study, 32 females and 34 males. All children had spastic diplegic CP. Pre-operatively there were 18 children at GMFCS levels I (28%), 24 at level II (36%) and 24 at level III (36%). They had a mean age of 10 years (SD 2 years 6 months) at the time of surgery (range 6 to 16 years). There was an average of eight procedures per child (range 4 to 12 procedures). All children were assessed three-monthly to 12 months post-operatively and 54 children (82%) were assessed at 24 months post-operatively. Data was not available for 12 children at 24 months because they had not attended the gait laboratory at this time point.
4.7.3.2 FMS results

As a group there were significant deteriorations in mobility at 3 and 6 months post-operatively for the 5, 50 and 500 metre distances (Table 4.1). Mobility for the group had returned to baseline levels or better by 12 months (odds ratios >1) and had improved at 24 months post-operatively.

Table 4.1 Odds ratios for each FMS distance at post-operative time points

<table>
<thead>
<tr>
<th>Time Post-op</th>
<th>FMS 5m OR (CI)</th>
<th>p value</th>
<th>FMS 50m OR (CI)</th>
<th>p value</th>
<th>FMS 500m OR (CI)</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>3 months</td>
<td>0.13</td>
<td>&lt;0.001*</td>
<td>0.09</td>
<td>&lt;0.001*</td>
<td>0.24</td>
<td>&lt;0.001*</td>
</tr>
<tr>
<td></td>
<td>0.07-0.24</td>
<td></td>
<td>0.04-0.17</td>
<td></td>
<td>0.14-0.43</td>
<td></td>
</tr>
<tr>
<td>6 months</td>
<td>0.36</td>
<td>&lt;0.001*</td>
<td>0.32</td>
<td>&lt;0.001*</td>
<td>0.50</td>
<td>0.004*</td>
</tr>
<tr>
<td></td>
<td>0.23-0.58</td>
<td></td>
<td>0.19-0.55</td>
<td></td>
<td>0.32-0.80</td>
<td></td>
</tr>
<tr>
<td>9 months</td>
<td>0.69</td>
<td>0.08</td>
<td>0.77</td>
<td>0.24</td>
<td>0.90</td>
<td>0.628</td>
</tr>
<tr>
<td></td>
<td>0.45-1.04</td>
<td></td>
<td>0.49-1.19</td>
<td></td>
<td>0.57-1.41</td>
<td></td>
</tr>
<tr>
<td>12 months</td>
<td>1.12</td>
<td>0.55</td>
<td>1.22</td>
<td>0.33</td>
<td>1.47</td>
<td>0.071</td>
</tr>
<tr>
<td></td>
<td>0.77-1.64</td>
<td></td>
<td>0.82-1.81</td>
<td></td>
<td>0.97-2.22</td>
<td></td>
</tr>
<tr>
<td>24 months</td>
<td>2.08</td>
<td>0.002+</td>
<td>2.16</td>
<td>0.001+</td>
<td>2.23</td>
<td>&lt;0.001+</td>
</tr>
<tr>
<td></td>
<td>1.33-3.24</td>
<td></td>
<td>1.37-3.41</td>
<td></td>
<td>1.44-3.45</td>
<td></td>
</tr>
</tbody>
</table>

OR: odds ratio  CI: confidence interval  
*statistically significant result – worse than baseline  
+ statistically significant result – better than baseline

To explore the effect of SEMLS on assistance required for the FMS 500 metre distance, the children classified as GMFCS level III pre-operatively were investigated more closely. They represented those who required assistive devices to walk before their surgery. For 500 metres the deterioration at 3 months was not statistically significant (odds ratio 0.53, p=0.21) but the improvement at 9 and 12 months was significant (odds ratio 2 p=0.01 and odds ratio 3.33 p=0.004 respectively). Pre-operatively 71% of children at this level used a wheelchair for mobility for 500 metres. At 3 months 83% were using a wheelchair, at 6 months 71% but by 9 months only 58% and at 12 months only 50% of this group were using a wheelchair for longer distances.
Chapter 4 – Development of the FMS

4.7.3.3 GMFCS

See Figure 4.2 for stability of GMFCS level. GMFCS level remained stable throughout most of the post-operative period for children classified pre-operatively as level III but not for children classified as I or II. The same 24 children classified as level III remained level III until 12 months post-operatively when 3 children became level II.

![GMFCS level chart]

Figure 4.2 Percentage of children in GMFCS levels post-operatively

Most (86%) of the 42 children who pre-operatively were GMFCS levels I and II (combined to represent all independent walkers without assistive devices) became level III at 3 months post-operatively. The corresponding percentages at later times were much smaller (Table 4.2).

Table 4.2 Children originally able to walk without assistive devices

<table>
<thead>
<tr>
<th>Operative stage</th>
<th>GMFCS I/II</th>
<th>GMFCS III</th>
</tr>
</thead>
<tbody>
<tr>
<td>Pre-operative</td>
<td>42</td>
<td>0</td>
</tr>
<tr>
<td>3 months post</td>
<td>6 (14%)</td>
<td>36 (86%)</td>
</tr>
<tr>
<td>6 months post</td>
<td>20 (48%)</td>
<td>22 (52%)</td>
</tr>
<tr>
<td>9 months post</td>
<td>30 (71%)</td>
<td>12 (29%)</td>
</tr>
<tr>
<td>12 months post</td>
<td>34 (81%)</td>
<td>8 (19%)</td>
</tr>
<tr>
<td>2 years post</td>
<td>34 (92%)</td>
<td>3 (8%)</td>
</tr>
</tbody>
</table>
4.7.4. Discussion

The major finding of this investigation was that changes were shown on the FMS throughout the post-operative period following SEMLS. The FMS discriminated change at three monthly time periods after surgery from pre-operative performance in a group of children with CP. Moreover the FMS demonstrated both deterioration and improvement in mobility over the post-operative course. The data showed large deteriorations in mobility at 3 and 6 months post-operatively. This was followed by improvement with mobility status back to baseline by 12 months post-operatively and then further improvement to better than baseline at 24 months post-operatively.

The odds ratios for the 500 metre distance followed a similar pattern to the 5 and 50 metre distances, although the changes were smaller at 3 and 6 months and slightly larger at 24 months. The smaller deterioration seen at 3 and 6 months is possibly because 39% of children used wheelchairs for the 500 metre distance prior to surgery whereas no children required them for 5 or 50 metres. Following SEMLS 56% of children used wheelchairs for 500 metres at 3 months and 39% of children at 6 months. The changes seen from pre to post-operative therefore, are not as striking for the 500 metre distance.

The FMS can show changes in the assistance required for longer distances (500 metres) following SEMLS. These changes in endurance were demonstrated for children classified as GMFCS III pre-operatively. This group represents those children who use assistive devices to walk and many use wheelchairs for longer distances. The deterioration seen at 3 and 6 months was not statistically significant with the percentage of children using a wheelchair for 500 metres not changing greatly compared with pre-operatively. Improvements were seen at 9 and 12 months with fewer children using wheelchairs for 500 metres. These children were able to progress to walking with assistive devices such as walkers or crutches rather than rely on a wheelchair for community distances. These changes were despite a stable GMFCS level which remained at III. The FMS therefore, is able to provide additional information in mobility changes and
assistance requirements that may not be detected by other scales or classifications.

An interesting finding on GMFCS stability was found. Previous studies have shown that the GMFCS is stable over time with many children remaining in the same GMFCS level (Wood and Rosenbaum 2000, Palisano et al 2006). The current study found the GMFCS level changed for many children after SEMLS, particularly for higher functioning children. This was in the initial post-operative phase where children initially deteriorated in function and then improved over the rehabilitation period. The GMFCS level was more stable when comparing pre-operative level to 12 months post-operatively.

A key function of evaluative outcome measures is to detect longitudinal clinical change in the functional performance of individuals or groups (Haley et al 1991). For children with CP undergoing SEMLS the FMS can be used to monitor post-operative change. The FMS data combined with the changing GMFCS distribution shows initial deterioration in mobility and function after surgery was followed by improvement. Considering the extensive nature of SEMLS, this deterioration in the early post-operative phase is not surprising. This corresponds with what is observed in children in the clinical setting and the information allows education of children and parents of the expected post-operative progress. The data also showed that improvements can occur up until 2 years post-operatively. This is valuable information for the child and family particularly when they may feel that progress is slow. It is important to note that this study did not assess responsiveness of the FMS; it examined changes in mobility after surgery as measured by the FMS. Responsiveness of the FMS will be examined in Chapter Six of this thesis.

There is always the possibility that natural history may contribute to positive changes observed following interventions in younger children who are still developing motor skills. A study creating motor development curves using longitudinal observations of children with cerebral palsy has been developed to describe patterns of gross motor development (Rosenbaum et al 2002). From these curves, 90% of the potential GMFM-66 score is reached by age 3.7 for
children GMFCS level III, by age 4.4 in children level II and by age 4.8 in children level I. Using this information it is unlikely that the positive changes seen after SEMLS in the present study were due to natural history because the sample did not include children younger than 6 years and the majority of children were aged 8-12 years.

The FMS is quick to administer and requires no equipment or formal training. It is a readily available and practical tool that measures mobility. The FMS is clinically useful, particularly when used in conjunction with other measures such as video assessment and clinical examination in situations when it is impractical to use three dimensional gait analysis. It has the unique feature of distinguishing clearly the different assistive devices used in the different environmental settings. Other measures group all assistive devices together and therefore do not have the ability to detect changes that occur as children progress from one assistive device to another. What is important from the results of this study is that the FMS is able to detect both deterioration and improvement in mobility; this is an important ability of the tool.

The main limitation of this study was that it was a retrospective analysis. The FMS was not administered according to revised guidelines and there was less ability to control procedures prior to data collection. One potential problem of retrospective analyses is missing data. This was minimized in this study by excluding children who did not have complete data for the first 12 months. Not all GMFCS levels were represented in this study. The sample included levels I-III only because those children with CP who attend a gait laboratory and consequently have SEMLS tend to be ambulant, thus excluding levels IV and V. The results therefore can only be generalized to this population.

4.8. Conclusion and recommendations

The FMS was developed to provide an activity outcome measure for children with CP specific to mobility. It is unique because it distinguishes different levels of assistance required for a range of environmental settings. It is a clinically feasible measure that has many potential uses. Initial pilot work showed the FMS detects changes in mobility following SEMLS at regular time points.
during rehabilitation. This is particularly for children classified as GMFCS level III, where other scales do not always show changes in the level of assistance required over time. The pilot work is promising; however before it can be described as a psychometrically robust tool it requires further examination. The remainder of this thesis is dedicated to examining requirements including inter-rater reliability, construct, concurrent and discriminative validity as well as responsiveness to change. The grand discussion in Chapter Nine of this thesis will examine possible changes to the FMS following all investigations.
CHAPTER FIVE: RELIABILITY OF THE FUNCTIONAL MOBILITY SCALE

5.1. Introduction

Chapter Three critically evaluated the psychometric properties of activity limitation measures for children with CP, including the FMS. The development and initial pilot testing of the FMS were reported in Chapter Four. The systematic review highlighted the need for further testing of the inter-rater reliability of the FMS for rating functional mobility in children with CP. This was considered to be a necessary prerequisite before confirming its validity for use by a range of clinicians. The current chapter describes the methodology and results from experiments investigating the inter-rater reliability of the FMS.

All measurements contain a proportion of error (Guilford 1954, Nunnally 1978). Reliability concerns the extent to which measurements are repeatable or consistent (Nunnally 1978, Tooth and Ottenbacher 2004). Reliability also concerns the error associated with measurement (Rothstein 2003). It is the proportion of true variance in obtained scores where an obtained score includes both a true component and an error component (Guilford 1954). There are a number of potential sources of error and variability of scores that can affect the reliability of results from a measurement procedure. These include variations from the test or scale (the equipment used), the observers or raters (the people collecting the data) and in the characteristic being measured in the participants (Nunnally 1978, Streiner and Norman 2003).

In clinical practice it is important to have reliable measurement procedures to enable appropriate and effective clinical decision making. Clinicians routinely use measures to evaluate the current status of their patients as well as to measure the effectiveness of treatments and interventions. For children with CP, there are a number of tools to measure aspects of activity, such as the FMS which measures mobility. These tools were appraised in Chapter Three. They can be used to measure the child at one point in time and also to evaluate change over time as the children grow and to assess the effectiveness of interventions such as
spasticity management and orthopaedic surgery. To ensure that the information obtained from these measurements is accurate and consistent, the measurement procedure needs to be reliable. When outcome measurement is used for research purposes, the process for measurement needs to be reliable to ensure that the results obtained can be largely attributable to true change or variance and not due to error in the measurement process.

Reliability testing of a measurement procedure determines how consistent the measurement is (Streiner and Norman 2003). The decision as to whether a particular measurement tool or method is reliable relates to whether the level of measurement error is considered acceptable for practical use and what level of reliability is considered to be clinically acceptable (Bruton et al 2000). A measurement cannot be considered as either reliable or unreliable because it is context dependent and relates to how the measurement will be used in practice (Keating and Matyas 1998, Rothstein 2003). For example, the requirement for reliability of a test to diagnose a life-threatening condition is very different to a test that measures gross motor function of children with CP.

There are a number of other terms that are sometimes used interchangeably with reliability such as ‘objectivity’, ‘reproducibility’, ‘stability’, ‘agreement’, ‘association’, ‘sensitivity’ and ‘precision’ (Streiner and Norman 2003). When the unit of measurement is on a categorical scale, as is the case for the FMS, reliability is typically assessed as a measure of agreement. Agreement refers to the extent to which raters or instruments produce essentially the same score (Tooth and Ottenbacher 2004).

There are several forms of reliability; inter-rater (or inter-observer), intra-rater (or intra-observer) and retest reliability. Inter-rater reliability concerns variations in scores between two or more raters who measure the same group of participants. Intra-rater measures the variation that occurs within an observer when observing the same group across two or more occasions (Streiner and Norman 2003). Retest reliability involves administering the same test to the same set of people after a period of time (Nunnally 1978). Intra-rater and retest reliability are closely related. The term “retest” reliability tends to be used when
Chapter 5 – Reliability

there are no observers involved in measurement (Streiner and Norman 2003). For example, there are measurement tools used for children with CP, such as the FAQ, that require the parents to complete a questionnaire rather than a clinician to administer it. Retest reliability is the most appropriate method of examining this tool because there are no “raters” to examine for consistency and we are interested in the ability of the tool to be consistent. The FMS requires a clinician to administer it by asking a series of questions of the child and/or parent. It is important that a high level of inter-rater reliability is demonstrated so that it can be used by different observers consistently.

Streiner (2003) has stated:

Although many investigators maintain that demonstration of both inter- and intra-observer reliability is a minimum requirement of a good test, this may be unnecessary. If we recognize that inter-observer reliability contains all the sources of error contributing to intra-observer reliability, plus any differences that may arise between observers, then it is evident that a demonstration of high inter-observer reliability is sufficient; the intra-observer is bound to be higher (Streiner and Norman 2003) (page 137).

For the current study the focus was on the examination of inter-rater reliability of the FMS with the stipulation that if low reliability was demonstrated, further examination of intra-rater reliability would be conducted. If low inter-rater reliability was demonstrated, the causes might be the result of differences within or between observers. Further investigation of intra-rater reliability would be required to identify where the source of the differences causing the low reliability were.

CP is usually recognised in early childhood and persists through the lifespan (Bax et al 2005, Rosenbaum et al 2007). Consequently children and adults with CP are managed by various health professionals over extended periods. Because management of patients with CP requires a multidisciplinary setting (Bax et al 2005), they will have many assessments by different clinicians throughout their childhood and adolescent years. This includes clinicians working in both the hospital and community settings. Inter-observer reliability is essential in this
population because different clinicians will be involved in the care of each person with CP. It is particularly important when using tools such as the FMS that have been developed for use by a range of clinicians. Intra-observer reliability is not as clinically relevant because it is unlikely that the same clinician will measure the same person with CP on an ongoing basis. In addition, as explained above, demonstration of high inter-observer reliability implies high intra-observer reliability.

Internal consistency is another factor to consider when examining reliability of a multi-item measurement scale. This concerns the estimates of reliability based on the average correlation among the individual items within a test (Nunnally 1978). The items should be moderately correlated with each other and each should correlate with the total score (Streiner and Norman 2003). Internal consistency is usually calculated using Cronbach alpha (Cronbach 1951) and is dependent on the number of items within a scale. For questionnaires in which the items are merely different aspects of a clinical phenomenon that do not have to be correlated, internal consistency is not relevant (Terwee et al 2007). This principle applies to the FMS because it has only three items within it. Each of those items measures the same construct over different distances. Examination of internal consistency is not a priority in this study due to the low number of items within the FMS and because each of those items would not be expected to correlate. If it was calculated, the coefficient would be very low and would not be clinically meaningful.

A preliminary study examined the inter-rater reliability of the FMS in 310 children with CP (Graham et al 2004). It reported high intraclass correlation coefficients (ICC’s) of 0.95 for 5 metres, 0.94 for 50 metres and 0.95 for 500 metres. The study also used Cronbach alphas as another index to assess inter-rater reliability with very similar results (0.95 for 5 metres, 0.94 for 50 metres and 0.96 for 500 metres). Internal consistency was not examined. A major limitation of the Graham study was the small number of raters used, all from the same clinical profession. Only three raters in total were used; two orthopaedic surgeons and one orthopaedic research fellow. This is an important limitation because the FMS is intended to be used by a range of clinicians from different
professional backgrounds. Reliability is specific to the population and raters it is tested on. Although it is promising that inter-rater reliability was high, it is important that reliability of the scale is examined further using a broader range and number of clinicians. This will enable the results to be generalized further and ensure the FMS can be administered reliably by a range of clinicians. It is also important to determine if there are any systematic biases for raters related to their clinical background. Surgeons might rate children differently to physiotherapists and the recognition of any such biases would provide evidence for the need to modify how the FMS is currently administered. This might include training required for clinicians or developing stricter guidelines for administration of the scale.

Another limitation of the Graham study was the use of ICC’s to analyse the data. It has recently been recommended that the kappa statistic is the most appropriate method of analysis for agreement between raters when the data are categorical (Tooth and Ottenbacher 2004). ICC’s can only be applied to ordinal data when the data fit the underlying assumptions of the ICC statistic, such as the assumption that intervals are equivalent (Tooth and Ottenbacher 2004). For the FMS, where it cannot be assumed that the intervals are equivalent, ICC’S are therefore not appropriate. Given that an ICC yields equivalent results to quadratic weighted kappas for ordinal data (Tooth and Ottenbacher 2004), it is likely that an ICC would overestimate the level of agreement for the FMS because there are only 6 levels within it. Unweighted kappa statistics would provide the most clinically meaningful results of agreement for the FMS.

The FMS was devised to rate the mobility of children with CP presenting with a wide range of severity of motor impairment (GMFCS levels I-IV). Within this spectrum of motor impairment there is the potential for differences in agreement between different observers. Children presenting with differing severity also present with differing stability or consistency of their mobility status. Some children may be very consistent with methods of mobility used and ability. Rating the mobility status of these stable children would be easier for clinicians than for those children whose mobility status is inconsistent or changing. It is important that agreement within this variation be examined to ensure that the
FMS is reliable for all severity of CP. Although the FMS was developed for children aged 4-18 years, there may be differences in the agreement between observers for different age groups within this range. At certain ages children may be more inconsistent with their mobility status due to changes associated with growth and motor development. During these periods it might be more difficult for clinicians to consistently rate the mobility of the children. It is important to examine the agreement for different age groups to ensure the FMS is reliable for the entire age range it is intended for.

A thorough examination of the inter-rater reliability of the FMS is necessary to provide evidence of its reliability across the spectrum of clinicians and children with CP it is relevant for. This will ensure the clinical utility of the scale. The research question of the current study was “what is the inter-rater agreement of the FMS using kappa statistics with a large sample and range of clinicians as raters”.

The specific aims were to determine;
   i. the inter-rater agreement for each distance of the FMS in children with CP using a large number of clinicians as raters from different clinical professions
   ii. the inter-rater agreement between clinicians for different age groups and severities of CP of children
   iii. the presence of any systematic bias of raters according to their clinical professions

The main hypothesis was “the inter-rater agreement for each distance of the FMS is greater than 0.8 as measured by kappa statistics in children with CP using a large group of raters from various clinical backgrounds”. The value of 0.8 was chosen because kappa statistic values of 0.61-0.81 represent substantial agreement and 0.81-1.00 represent almost perfect agreement (Landis and Koch 1977). Values lower than 0.6 would be considered as poor, slight, fair or moderate. Although these divisions are arbitrary, they are useful for providing benchmarks from which the results obtained can be discussed. It was anticipated
that the FMS would show substantial agreement as a minimum to ensure its validity and clinical utility.

It was also hypothesized that “the level of inter-rater agreement for the FMS in children with CP is lower for children aged 2-6 years compared to children aged 6-12 or 12-18 years”. In addition, data analysis explored whether there are differences for level of clinician agreement for the FMS when rating children with CP depending on;

i. the severity of motor impairment, as described by GMFCS level or topographical distribution

ii. clinician rater combinations and rater clinical profession.

5.2. Methods

5.2.1. Participants

Several clinician raters were required as well as a sample of children for testing. Participants therefore included both clinicians as raters and children with CP.

5.2.1.1 Clinician raters

There were three categories of raters involved who were selected based on the range of clinicians working with children with CP within the hospital setting and in the community:

i. hospital physiotherapists involved with the gait laboratory or CP clinics,

ii. orthopaedic surgeons or fellows attending the clinics or gait laboratory and

iii. community physiotherapists involved in the children’s management.

These categories represented a good balance of key users of the FMS. The hospital physiotherapists were selected from the group of physiotherapists who attend the clinics and the gait laboratory. The orthopaedic surgeons were selected from those that consult at the clinics or gait laboratory. Selection was based on which surgeon and physiotherapist was attending the appointments on a particular day. The community physiotherapists were selected if they were the primary physiotherapist involved in the care of a particular child recruited who
had been randomized to have one rater as their community physiotherapist. This spread of professional backgrounds was selected because these are the clinicians who at that point in time were the primary users of the scale for children with CP attending the CP clinics and gait laboratory. The spread ensured there was more than one professional type represented with a large number of raters involved. It also ensured both hospital and community clinicians were represented.

5.2.1.2 Children

Inclusion criteria for the participants who were rated using the FMS were:

i. Children with CP aged 2-18 years
ii. Children classified as GMFCS levels I-IV
iii. Children who attended the CP clinics or gait laboratory of the Royal Children’s Hospital, Melbourne

The age upper age limit was set at 18 years because the FMS has been primarily devised for children and adolescents with CP aged 4-18 years. This study included children from 2 years on because one aim was to examine reliability in different age groups. Particular interest was whether reliability is lower in younger children due to motor skills and gait patterns that are still developing. Research on the development of walking in children with CP has shown that walking can be achieved up to the age of 9 years (Campos da Paz Jr et al 1994, Bottos et al 1995, Fedrizzi et al 2000). These studies also show that children with hemiplegia and some with diplegia will achieve walking at ages as young as 2-5 years. With this range of potential age of acquisition of walking, it was thought that reliability might be lower in the younger children due to walking skills that were still developing causing inconsistent performance. This could cause a change in performance between ratings and also make assigning a particular FMS category to the changing child more difficult due to inconsistency. Children younger than 2 years were excluded also based on this research. Many children would not be walking at all at 2 years of age and the FMS is a scale that essentially describes walking mobility.
Chapter 5 – Reliability

Children classified as GMFCS levels I-IV were included because the FMS was developed for these levels as explained in Chapter 4. Levels I-IV represent the range of severity of motor impairment in children with CP with some form of independent mobility. The FMS focuses on independent mobility. The children were recruited from the two major clinics for children with CP at the Royal Children’s Hospital, Melbourne, Australia.

Excluded were children:

i. with a diagnosis other than CP

ii. younger than 2 years and older than 18 years of age

iii. classified as GMFCS level V

iv. who had undergone orthopaedic surgery within the previous six months or Botulinum toxin injections within the previous 3 months.

v. whose parents were not able to understand the information explaining the study or the instructions due to language, cognitive or other difficulties

vi. whose parents were upset or anxious associated with their clinic or gait laboratory appointment.

If children had a diagnosis other than CP they were excluded because the scale has been devised for CP and the population of interest for this study was children with CP. Children younger than 2 years and older than 18 years were excluded for the same reasons as justified in the inclusion criteria. Children classified as GMFCS level V were excluded because these children have severe motor impairments and usually require adult assistance for all mobility with no means of independent mobility (Palisano et al 1997). The FMS is not an appropriate tool to use for these children because it focuses on independent mobility.

Children who had received recent surgery or botulinum toxin injections were excluded. In the initial post-intervention period the physical function of these children was likely to be changing as a result of the interventions. This could introduce change within the participants between the two ratings as a result of treatment effect and produce a source of variation. In turn this could result in different scores from the two raters not attributable to rater performance. The
aim of the study was to examine inter-rater agreement and rater performance, not the effect of treatment. These sources of variation in the participants were therefore deliberately minimised. An exception was when both ratings were performed on the same day. It would be most unlikely that change as a result of treatment effect of an intervention prior to both ratings would occur within a 4 hour period. It was considered that ratings performed on the same day would not introduce a source of variation within the participants.

Children whose parents were unable to understand due to language or cognitive problems were excluded if they could not fully comprehend what was required. Where interpreters were available for parents the children were included. Before parents were approached to consider participation in the study, the principal investigator consulted with the primary clinician involved in the care of each child on the day of the clinic visit. If it was felt by either the clinician or investigator that the parents were too upset or emotional, these parents were not approached and those children not included.

5.2.1.3 Sample size of children

A large enough sample size to provide sufficient precision to result in reasonably narrow confidence intervals around the observed kappa statistics was required. Agreement of 0.8 or greater was aimed for because this is a very good level of agreement (Landis and Koch 1977). Sample size was obtained using table II in Walter et al (1998) using a method with a design of two raters per participant, power of 80%, confidence interval of 95%, and the expected lower limit for kappa as 0.7 (Walter et al 1998). To do this a sample of 117.1 (or 118) participants was required. It was estimated that around 10% attrition of the ratings would occur where community physiotherapists were involved (8 participants) because this required them to return the information via mail. A sample size of 126 was aimed for to account for this potential attrition.

5.2.2. Procedures

The study was approved by the Ethics in Human Research Committee of the Royal Children’s Hospital (reference number 25118A) and The University of Melbourne Human Ethics Research Committee (reference number 060211).
Written consent was obtained from each participating child’s parent or legal guardian prior to data collection as outlined below.

5.2.2.1 Recruitment

A consecutive sample was utilized for this study in order to obtain the required number of participants. A random sample was not used because it would not be possible to recruit the required sample of 118 within a 4-6 month period based on the number of eligible children who attend the clinics. The clinics where the children were recruited from represent the majority of children that the FMS is used with in clinical practice. By using a consecutive sample from these clinics there was a high chance that the sample would be representative of the population for which the study was aimed. The advantage of using a sample of convenience was the required number of participants could be recruited within the time frame stated above. The limitation was that it could not be assumed that the sample exactly represented the target population.

The primary investigator (AH) approached potential participants and recruited the children. Participants were recruited from the gait laboratory and three CP clinics at the Royal Children’s Hospital, Melbourne, Australia. Potential participants were identified by the primary investigator through prospective gait laboratory and CP clinic appointment lists. Once eligibility from these lists was determined the primary investigator attended the clinics and gait laboratory appointments. The parents or guardians of eligible children were invited to participate after a full explanation of the study was provided. On agreement for their child to participate, full consent was obtained with signed consent forms and a written information sheet given to the parents/guardians.

5.2.2.2 Allocation of raters

Each child was rated on the FMS on one occasion by two different raters with two ratings per child the result. The decision to use only two raters per child reflected the nature of administration of the scale. The FMS is administered within 10 minutes by asking a few questions of the child and/or parents regarding how they choose to “move around”. It was decided that if more than two raters were assigned to each participant, the child and/or parents could
potentially start “rehearsing” the answers by recalling what they had answered previously, thus introducing an element of bias.

Participating children were assigned to two raters using rater combinations. Possible rater combinations and order of raters were:

i. laboratory physiotherapist followed by orthopaedic surgeon
ii. laboratory physiotherapist followed by community physiotherapist
iii. orthopaedic surgeon followed by community physiotherapist
iv. orthopaedic surgeon followed by laboratory physiotherapist
v. community physiotherapist followed by laboratory physiotherapist
vi. community physiotherapist followed by orthopaedic surgeon.

The raters were allocated by computer randomization in a block design by a statistician. Within each block of 6 participants, the order of the above six rater combinations was selected randomly. This was then repeated for each consecutive block of six participants so that within each the sequence of raters occurred randomly.

5.2.2.3 Testing procedure

At the clinic or laboratory appointment the principal investigator (AH) was responsible for recruiting the children, coordinating the ratings and collecting key information including date of birth, age, severity of motor impairment of CP as determined by GMFCS (Palisano et al 1997) and topographical distribution, and the walking scale of the FAQ (Novacheck et al 2000). The principal investigator was not a rater because she was required to be independent of the rating process to minimize chance of bias. The collection of information such as GMFCS level could influence the FMS rating and it was essential that each rater be unaware of previous ratings obtained. Her role was to collect information and coordinate the raters. After the parents or guardians had signed the consent forms and the children were classified according to their GMFCS level, they completed the FAQ. The data obtained from the FAQ were not utilized for this study yet was part of data collection for discriminative and concurrent validity (see Chapter Eight). A copy of the FAQ can be found in Appendix E.
Each child was then rated by two different raters. Where the rater combination allocated to a child was laboratory physiotherapist and orthopaedic surgeon, both ratings were performed on the same day at the hospital. This was because both raters were available to rate the children on the same day and minimized the chance of change occurring between ratings influencing the results. Each rating was performed in the absence of the other rater and raters were blind as to the previous rating obtained. Where a community physiotherapist was involved, the child was rated at the hospital appointment by one rater and the second rating was performed by the child's community physiotherapist within two weeks. This was either before or after the hospital rating, depending on who was allocated to rate the child first. If the child was due to see their community physiotherapist within 2 weeks, the rating was performed in person either at home or school depending on where sessions usually occurred. The rating was performed via telephone if the physiotherapist was not due to see the child in person within two weeks.

A time frame of 2 weeks was selected because this was deemed long enough for the physiotherapist to either see the child in person or contact them over the telephone yet not too long to allow changes in mobility to occur. Obtaining some of the FMS ratings over the telephone could possibly introduce a source of variation between raters with less information provided to those administering it over the telephone. Although the FMS is administered by self-report and not direct observation, the child is often present when scored. Information from having the child present can inadvertently guide the clinician’s rating. Ratings obtained over the telephone are not influenced by this extra information. To determine the effect of this, data analysis involved comparing ratings obtained over the telephone with those performed in person.

The raters were provided with an information package prior to testing children. This included the FMS brochure that explains the scale and how to administer it (Appendix A). Also provided were study information sheets and a standard form to complete the rating obtained, date of rating and additional comments. The community physiotherapists had this package mailed to them with a stamped, self-addressed envelope for return of the rating to the principal investigator.
Where the community physiotherapist was allocated as the first rater, the package was sent ahead of the clinic visit to ensure the rating was performed prior to the child attending their hospital appointment.

5.2.3. Data analysis

Descriptive statistics for participant characteristics were compiled including means and standard deviations for continuous variables and frequency and percentages for categorical variables. Kappa statistics with 95% confidence intervals were used to analyse agreement between ratings due to the ordinal nature of the data. Kappa is an extension of simple percent agreement and is defined as “the proportion of the total amount of agreement not explained by chance for which the observers accounted” (Cohen 1960) (p38). The data were primarily analysed using unweighted Kappa statistics because the basic or unweighted Kappa considers all disagreements in ratings as equal in seriousness (Tooth and Ottenbacher 2004). For a scale such as the FMS with only six levels it was decided that disagreement of one level is a clinically significant difference that should be considered. Weighted Kappa was developed for use with ordinal data (Cohen 1968) and reflects the degree of disagreement by attaching greater emphasis to large differences between ratings to small differences (Sim and Wright 2005). In this situation, the use of weighted Kappas is likely to result in inflated agreement that is not clinically meaningful.

Kappa statistics for agreement were obtained for:

i. each FMS distance (5, 50 and 500 metres)

ii. each FMS distance for different GMFCS levels

iii. each FMS distance for different age groups (2-6 years, 6-12 years and 12-18 years)

iv. each FMS distance comparing ratings performed in person and those obtained over the telephone

v. each FMS distance according to different rater combinations

Kappa does not indicate whether disagreement is due to random differences or systematic differences between clinicians ratings and the data should be examined accordingly (Sim and Wright 2005). The presence of any systematic
Biases between raters was examined using Wilcoxon signed rank tests. This test examines differences within paired scores and examines both the direction of the difference and the relative amount of difference (Wilcoxon 1945). This test was used to examine each FMS distance for different rater combinations independent of the order of raters, resulting in comparisons between:

i. hospital physiotherapists and surgeons
ii. hospital physiotherapists and community physiotherapists
iii. surgeons and community physiotherapists.

The p value for the Wilcoxon signed-rank tests was set at 0.05 for rejecting the null hypothesis. This represents a 5% probability of making a Type I error and incorrectly rejecting the null hypothesis (Portney and Watkins 1993). This was considered an appropriate level of significance for the study in question.

The data were stored and organized using the EpiData for Windows program ([http://www.epidata.dk/](http://www.epidata.dk/)). All analyses were performed using Stata [StataCorp 2005 Stata statistical software Release 9.0. College Station; TX. Stata Press.].

5.3. Results

5.3.1. Descriptive results of participant characteristics

The sample included 123 children who matched the inclusion criteria. Of these 123, five were lost due to attrition. For these five children one rating had been completed by either a hospital physiotherapist or surgeon with the second rating to be completed by a community physiotherapist. This second rating was either not completed by the community physiotherapist or the package was sent to the incorrect physiotherapist. These 5 participants lost to follow-up left a total sample of 118.

The participants had a mean age of 10.3 years (SD 3.6) with a range of 2.5-18 years. There were 16 children aged 2-6 years, 66 aged 6-12 years and 36 aged 12-18 years. There were 74 males and 44 females. The children were classified as GMFCS levels I-IV. There were 13 children GMFCS level I, 49 level II, 44 level III and 12 level IV. There were 21 children with a hemiplegic distribution
of CP, 76 with diplegia and 21 with quadriplegia. Table 5.1 summarises the participant characteristics.

Table 5.1 *Participant characteristics*

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>n</th>
<th>118</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (y,m), mean (SD)</td>
<td>Total</td>
<td>10.3 (3.6)</td>
</tr>
<tr>
<td>Age n (%)</td>
<td>2-6 years</td>
<td>16 (14%)</td>
</tr>
<tr>
<td></td>
<td>6-12 years</td>
<td>66 (56%)</td>
</tr>
<tr>
<td></td>
<td>12-18 years</td>
<td>36 (30%)</td>
</tr>
<tr>
<td>Gender, n (%)</td>
<td>Male</td>
<td>74 (63%)</td>
</tr>
<tr>
<td></td>
<td>Female</td>
<td>44 (37%)</td>
</tr>
<tr>
<td>GMFCS, n (%)</td>
<td>I</td>
<td>13 (11%)</td>
</tr>
<tr>
<td></td>
<td>II</td>
<td>49 (42%)</td>
</tr>
<tr>
<td></td>
<td>III</td>
<td>44 (37%)</td>
</tr>
<tr>
<td></td>
<td>IV</td>
<td>12 (10%)</td>
</tr>
<tr>
<td>Topography</td>
<td>Hemiplegia</td>
<td>21 (18%)</td>
</tr>
<tr>
<td></td>
<td>Diplegia</td>
<td>76 (64%)</td>
</tr>
<tr>
<td></td>
<td>Quadriplegia</td>
<td>21 (18%)</td>
</tr>
</tbody>
</table>

For each FMS distance the mode of the rating was calculated combining both ratings for each child with a total of 236 ratings. For 5 metres it was level 5 with 100 ratings (42%), for 50 metres it was level 5 with 88 ratings (37%) and for 500 metres it was level 1 with 116 ratings (49%). Most children walked independently on level surfaces for 5 and 50 metres and used a wheelchair for 500 metres. For 5 metres 161 of the ratings (68%) were between levels 4-6 i.e. the high level end of the scale. For 50 metres ratings tended to be at 5 and 6 (114 or 48%) with the children walking independently or at the other end 1-2 (93 or 39%) using a wheelchair or walker. For 500 metres the children either tended to walk on level surfaces (69 or 29%) or they used a wheelchair (116 or 49%).

There were five hospital physiotherapists, three orthopaedic surgeons and 36 community physiotherapists involved as raters. Hospital physiotherapists performed 80 ratings, orthopaedic surgeons performed 80 ratings and community physiotherapists performed 76 ratings. The majority of ratings were performed in person (87.3% of first ratings and 88.1% of second ratings) with
only a small number performed over the telephone (12.7% of first ratings and 11.9% of second ratings). The number of occasions where both ratings were performed on the same day was 45 (38%) and another 40 (34%) were done within 1-7 days apart.

5.3.2. Inter-rater reliability results

Guidelines for ranges of kappa values describing the strength of the agreement have been reported (Landis and Koch 1977). Using these guidelines, a kappa of 0.00 indicates poor agreement, 0.00-0.20 indicates slight agreement, 0.21-0.40 fair agreement, 0.41-0.60 moderate agreement. 0.61-0.80 substantial agreement and 0.81-1.00 almost perfect agreement. The authors state that these divisions are clearly arbitrary yet they do provide useful benchmarks for discussing results. Interpreting the magnitude of Kappa needs to take into account other factors influencing magnitude such as prevalence, bias and nonindependence of ratings and the choice of weighting scheme of Kappa (Sim and Wright 2005).

The overall unweighted kappa statistics for inter-rater reliability for each FMS distance showed substantial agreement with:

i. 5 metres: κ = 0.69

ii. 50 metres: κ=0.71

iii. 500 metres: κ = 0.66

Weighted kappas using a quadratic weighting system were calculated for each FMS distance as a comparison with results of 0.87 for 5 metres, 0.92 for 50 metres and 0.86 for 500 metres. Table 5.2 summarises the unweighted kappa, the weighted kappa and percentage agreement for each FMS distance.

Table 5.2 Kappa values for each FMS distance

<table>
<thead>
<tr>
<th>FMS distance</th>
<th>Kappa (CI) (unweighted)</th>
<th>Agreement</th>
<th>Kappa (CI) (weighted)</th>
<th>Agreement</th>
</tr>
</thead>
<tbody>
<tr>
<td>5m</td>
<td>0.69 (0.60, 0.77)</td>
<td>76%</td>
<td>0.87 (0.74, 0.99)</td>
<td>96%</td>
</tr>
<tr>
<td>50m</td>
<td>0.71 (0.62, 0.80)</td>
<td>78%</td>
<td>0.92 (0.74, 1.00*)</td>
<td>98%</td>
</tr>
<tr>
<td>500m</td>
<td>0.66 (0.55, 0.76)</td>
<td>77%</td>
<td>0.86 (0.68, 1.00*)</td>
<td>96%</td>
</tr>
</tbody>
</table>

CI = confidence interval

*CI's truncated to 1.00 at upper end
The data were examined further for differences in agreement within different age groups of children for: 1) children aged 2-6 years, 2) children aged 6–12 years and 3) children aged 12-18 years. There were too few children less than 4 years to analyse between 2-4 years. Table 5.3 summarises the unweighted kappa statistics and percentage agreement for these age bands for each FMS distance. For all three distances the 6-12 year group showed the highest reliability and agreement. For 50 and 500 metres the lowest reliability and agreement was for children aged 2-6 years. These can be considered descriptive patterns only as the large confidence intervals suggest the differences are not significant. There were differences in frequency of children in each group with only 16 children in the 2-6 year group compared to 66 in the 6-12 years and 36 in the 12-18 years bands.

Table 5.3 *Kappa values and agreement for different age groups of children*

<table>
<thead>
<tr>
<th>Age</th>
<th>FMS distance</th>
<th>Kappa (CI)</th>
<th>% Agreement</th>
</tr>
</thead>
<tbody>
<tr>
<td>2-6 years</td>
<td>FMS 5m</td>
<td>0.66 (0.41, 0.91)</td>
<td>75%</td>
</tr>
<tr>
<td></td>
<td>FMS 50m</td>
<td>0.60 (0.37, 0.84)</td>
<td>69%</td>
</tr>
<tr>
<td></td>
<td>FMS 500m</td>
<td>0.38 (0.11, 0.66)</td>
<td>69%</td>
</tr>
<tr>
<td>6-12 years</td>
<td>FMS 5m</td>
<td>0.75 (0.63, 0.87)</td>
<td>82%</td>
</tr>
<tr>
<td></td>
<td>FMS 50m</td>
<td>0.77 (0.64, 0.90)</td>
<td>83%</td>
</tr>
<tr>
<td></td>
<td>FMS 500m</td>
<td>0.73 (0.58, 0.89)</td>
<td>83%</td>
</tr>
<tr>
<td>12-18 years</td>
<td>FMS 5m</td>
<td>0.56 (0.40, 0.72)</td>
<td>67%</td>
</tr>
<tr>
<td></td>
<td>FMS 50m</td>
<td>0.65 (0.49, 0.80)</td>
<td>72%</td>
</tr>
<tr>
<td></td>
<td>FMS 500m</td>
<td>0.69 (0.50, 0.88)</td>
<td>78%</td>
</tr>
</tbody>
</table>

Agreement was examined for differing levels of motor impairment as classified by GMFCS levels. The results varied according to GMFCS level. Table 5.4 summarises the unweighted kappa statistics and percentage agreement for each GMFCS level at each FMS distance. For levels I and II at 5 and 50 metres and level I at 50 metres the reliability and percentage agreement were lower than for levels III and IV. For 500 metres level IV had the highest agreement with level II the lowest. Caution needs to be exercised in interpreting this data. The low kappa values for GMFCS levels I and II at 5 metres and for level I at 50 metres can in part be explained by the spread of scores rated. Examination of the agreement tables for these comparisons show that the majority of scores are up the highest end of the FMS, i.e. 5 and 6. With a reduced number of cells for...
calculation of the kappa statistic, there is the potential of the results to be skewed toward a lower kappa value. For GMFCS level IV at 50 metres there was perfect agreement hence the kappa of 1.0. For level IV at 500 metres all children were rated as using wheelchairs by all raters, resulting in too few categories for a kappa statistic to be calculated. The confidence intervals around the kappa values for different GMFCS levels are wide suggesting the sample size is not large enough to detect differences at any meaningful level. Because of this, no strong statements on differences in agreement between levels can be made.

<table>
<thead>
<tr>
<th>GMFCS level</th>
<th>FMS</th>
<th>Kappa (CI)</th>
<th>% agreement</th>
</tr>
</thead>
<tbody>
<tr>
<td>I</td>
<td>5m</td>
<td>0.32 (-0.08, 0.71)</td>
<td>77%</td>
</tr>
<tr>
<td></td>
<td>50m</td>
<td>0.08 (-0.37, 0.54)</td>
<td>62%</td>
</tr>
<tr>
<td></td>
<td>500m</td>
<td>0.70 (0.18, 1.00*)</td>
<td>85%</td>
</tr>
<tr>
<td>II</td>
<td>5m</td>
<td>0.09 (-0.15, 0.32)</td>
<td>71%</td>
</tr>
<tr>
<td></td>
<td>50m</td>
<td>0.43 (0.25, 0.60)</td>
<td>80%</td>
</tr>
<tr>
<td></td>
<td>500m</td>
<td>0.46 (0.27, 0.64)</td>
<td>67%</td>
</tr>
<tr>
<td>III</td>
<td>5m</td>
<td>0.71 (0.56, 0.86)</td>
<td>77%</td>
</tr>
<tr>
<td></td>
<td>50m</td>
<td>0.57 (0.39, 0.75)</td>
<td>75%</td>
</tr>
<tr>
<td></td>
<td>500m</td>
<td>0.56 (0.37, 0.76)</td>
<td>82%</td>
</tr>
<tr>
<td>IV</td>
<td>5m</td>
<td>0.84 (0.39, 1.00*)</td>
<td>95%</td>
</tr>
<tr>
<td></td>
<td>50m</td>
<td>1.00 (0.43, 1.00*)</td>
<td>100%</td>
</tr>
<tr>
<td></td>
<td>500m</td>
<td>0.00^ 0.00</td>
<td>100%</td>
</tr>
</tbody>
</table>

Topography

<table>
<thead>
<tr>
<th>Topography</th>
<th>FMS</th>
<th>Kappa (CI)</th>
<th>% agreement</th>
</tr>
</thead>
<tbody>
<tr>
<td>Hemiplegia</td>
<td>5m</td>
<td>0.42 (0.00, 0.84)</td>
<td>71%</td>
</tr>
<tr>
<td></td>
<td>50m</td>
<td>0.46 (0.05, 0.86)</td>
<td>76%</td>
</tr>
<tr>
<td></td>
<td>500m</td>
<td>0.50 (0.21, 0.79)</td>
<td>71%</td>
</tr>
<tr>
<td>Diplegia</td>
<td>5m</td>
<td>0.66 (0.54, 0.77)</td>
<td>75%</td>
</tr>
<tr>
<td></td>
<td>50m</td>
<td>0.68 (0.56, 0.80)</td>
<td>76%</td>
</tr>
<tr>
<td></td>
<td>500m</td>
<td>0.64 (0.51, 0.78)</td>
<td>76%</td>
</tr>
<tr>
<td>Quadriplegia</td>
<td>5m</td>
<td>0.82 (0.61, 1.00*)</td>
<td>86%</td>
</tr>
<tr>
<td></td>
<td>50m</td>
<td>0.73 (0.36, 1.00*)</td>
<td>86%</td>
</tr>
<tr>
<td></td>
<td>500m</td>
<td>0.00^ 0.00</td>
<td>100%</td>
</tr>
</tbody>
</table>

* CI’s truncated to 1.00 at upper end  ^ too few categories to calculate kappa

The reliability and agreement for topographical distribution was examined and these results are also summarized in Table 5.4. The reliability and agreement was lowest for children with hemiplegia and highest for quadriplegia. All raters scored children with quadriplegia at 500 metres as using a wheelchair resulting
in too few categories for a kappa statistic to be calculated. Once again the confidence intervals are very wide; therefore differences in levels of agreement are not statistically significant.

Agreement was similar for different methods of administration for 5 and 50 metres. For 5 metres the agreement for both ratings done in person was $\kappa = 0.68$ (CI 0.58-0.78) and for one of the ratings done on the telephone was $\kappa = 0.70$ (CI 0.53-0.87). For 50 metres the agreement was $\kappa = 0.70$ (CI 0.60-0.81) where both ratings were done in person and $\kappa = 0.72$ (CI 0.52-0.91) where one rating was performed over the telephone. For 500 metres the agreement appeared lower when both ratings were performed in person with $\kappa = 0.66$ (CI 0.53-0.79) compared to 0.78 (CI 0.54-1.01) where one was performed over the telephone. Although the second kappa is higher, the large confidence intervals do not support this difference as statistically significant. Also, only 12% of ratings were performed over the telephone.

Agreement was slightly higher when both ratings were performed on the same day compared to when ratings were performed up to 2 weeks apart, also with large confidence intervals. For 5 metres the agreement for same day ratings was $\kappa = 0.74$ (CI 0.60-0.88) and for different days $\kappa = 0.65$ (CI 0.53-0.76). For 50 metres the agreement for same day ratings was $\kappa = 0.77$ (CI 0.61-0.92) compared to different days $\kappa = 0.67$ (CI 0.55-0.79). For 500 metres the agreement for same day ratings was $\kappa = 0.76$ (CI 0.58-0.95) compared to ratings performed on different days $\kappa = 0.59$ (CI 0.45-0.72).

Kappa statistics were calculated for different rater combinations and the results are summarized in Table 5.5. For 5 metres the lowest agreement was between hospital and community physiotherapists. For 50 and 500 metres the lowest agreement was between surgeons and community physiotherapists. The width of the confidence intervals ensures that these are not statistically significant differences. What can be stated is that overall the agreement was moderate to substantial for all combinations of raters.
Table 5.5 *Kappa values for different rater combinations*

<table>
<thead>
<tr>
<th>Raters</th>
<th>FMS distance</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>5 metres</td>
</tr>
<tr>
<td>hospPT &amp; surgeon</td>
<td>0.74 (0.59, 0.89)</td>
</tr>
<tr>
<td>hospPT &amp; commPT</td>
<td>0.57 (0.40, 0.75)</td>
</tr>
<tr>
<td>surgeon &amp; commPT</td>
<td>0.71 (0.57, 0.85)</td>
</tr>
</tbody>
</table>

5.3.3. Systematic biases of raters

The analyses and results above describe the agreement for different rater combinations. The results do not provide information regarding possible systematic biases of certain clinician groups, for example whether surgeons rate children differently to physiotherapists. This was better addressed using the Wilcoxon signed-rank tests. Signed ranks were calculated for the three combinations of raters (hospital physiotherapist and surgeon, hospital physiotherapist and community physiotherapist, and surgeon and community physiotherapist) for each FMS distance. The results are organized for each distance in Tables 5.6-5.8. Within these tables, “Disagreements” represent the number of observations where raters disagree and the direction (+ or -) of the disagreements, “Agreements” represents the number of observations where both raters agree and “Observations” represents the total number of observations (the number of children scored by each rater). Adding up both disagreements gives the total number of disagreements. If there are clearly more in one direction there is a possible bias in ratings. If there are near equal numbers of positive and negative disagreement then no real pattern can be determined.

Table 5.6 *Wilcoxon signed rank tests for FMS 5 metres*

<table>
<thead>
<tr>
<th></th>
<th></th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Disagreements (+)</td>
<td>1</td>
<td>6</td>
<td>6</td>
</tr>
<tr>
<td>Disagreements (-)</td>
<td>7</td>
<td>5</td>
<td>3</td>
</tr>
<tr>
<td>Agreements (n)</td>
<td>34</td>
<td>27</td>
<td>29</td>
</tr>
<tr>
<td>Observations (n)</td>
<td>42</td>
<td>38</td>
<td>38</td>
</tr>
<tr>
<td>p value</td>
<td>0.03</td>
<td>0.66</td>
<td>0.33</td>
</tr>
</tbody>
</table>
Table 5.7 Wilcoxon signed rank tests for FMS 50 metres

<table>
<thead>
<tr>
<th></th>
<th></th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Disagreements (+)</td>
<td>2</td>
<td>4</td>
<td>10</td>
</tr>
<tr>
<td>Disagreements (-)</td>
<td>6</td>
<td>1</td>
<td>3</td>
</tr>
<tr>
<td>Agreements (n)</td>
<td>34</td>
<td>33</td>
<td>25</td>
</tr>
<tr>
<td>Observations (n)</td>
<td>42</td>
<td>38</td>
<td>38</td>
</tr>
<tr>
<td>p value</td>
<td>0.15</td>
<td>0.17</td>
<td>0.06</td>
</tr>
</tbody>
</table>

Further information can be extrapolated from the 2x2 tables for each rater combination about where the differences rest. It is unnecessary to display all tables, however two are shown in the following examples to illustrate this point.

Table 5.8 Wilcoxon signed rank tests for FMS 500 metres

<table>
<thead>
<tr>
<th></th>
<th></th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Disagreements (+)</td>
<td>3</td>
<td>4</td>
<td>4</td>
</tr>
<tr>
<td>Disagreements (-)</td>
<td>4</td>
<td>6</td>
<td>6</td>
</tr>
<tr>
<td>Agreements (n)</td>
<td>35</td>
<td>28</td>
<td>28</td>
</tr>
<tr>
<td>Observations (n)</td>
<td>42</td>
<td>38</td>
<td>38</td>
</tr>
<tr>
<td>p value</td>
<td>0.76</td>
<td>0.50</td>
<td>0.45</td>
</tr>
</tbody>
</table>

Table 5.6 shows the results for the FMS 5 metre data. The only significant result was for ratings between hospital physiotherapists and surgeons with a p value of 0.03. Of 42 observations there were eight disagreements with seven of those eight in the negative direction suggesting a bias. On examining at the 2x2 table for this result (Table 5.9), it is evident that the disagreements are at the higher end of the scale where surgeons tend to rate more children as 6 than 5 compared to hospital physiotherapists.

Table 5.9 Agreement between surgeons and hospital physiotherapists for FMS 5 metres
Chapter 5 – Reliability

Ratings between hospital and community physiotherapists for 5 metres show that of 38 observations there were 11 disagreements. There were near equal numbers of positive and negative scores (six and five respectively) with a p value of 0.66. This suggests that differences in ratings between these groups are not significant and no pattern of bias in ratings is evident, despite a large number of disagreements. This is illustrated on Table 5.10.

Table 5.10 Agreement between community and hospital physiotherapists for FMS 5 metres

<table>
<thead>
<tr>
<th>FMS</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
<th>crawl</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>2</td>
<td>0</td>
<td>1</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>2</td>
</tr>
<tr>
<td>3</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>4</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>5</td>
<td>2</td>
<td>0</td>
<td>0</td>
<td>8</td>
</tr>
<tr>
<td>5</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>2</td>
<td>16</td>
<td>2</td>
<td>0</td>
<td>20</td>
</tr>
<tr>
<td>6</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>crawl</td>
<td>1</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>3</td>
<td>5</td>
</tr>
<tr>
<td>Total</td>
<td>2</td>
<td>3</td>
<td>2</td>
<td>7</td>
<td>19</td>
<td>2</td>
<td>3</td>
<td>38</td>
</tr>
</tbody>
</table>

There were nine disagreements of 38 observations between surgeons and community physiotherapists for FMS 5 metres with more positive than negative scores. The p value of 0.33 suggests this is not significant however the 2x2 table shows that surgeons rated slightly more children as 6 than 5 compared to community physiotherapists.

Table 5.7 summarises the results for the FMS 50 metre data. Between hospital physiotherapists and surgeons there were eight disagreements of a total 42 with more negative than positive scores. This is similar to the same rating combinations for 5 metres although the p value is smaller at 0.15. Once again the disagreements are at the higher end of the scale with surgeons rating more children as 6 than 5 compared to hospital physiotherapists. Between hospital and community physiotherapists there were only five disagreements of a total 38 observations with more positive than negative scores, however the numbers are low. The p value and 2x2 table suggest there is no pattern of bias or significant difference in raters. There were 13 of 38 disagreements for surgeons.
and community physiotherapists with more positive scores. The p value is nearly significant at 0.06 and the 2x2 table shows the disagreement is at the higher end of the scale with surgeons rating more children at 6 than 5 compared to the community physiotherapists.

Table 5.8 summarises the results for the FMS 500 metre distance. For all three rater combinations there were between 7-10 disagreements and the numbers of positive and negative scores were near equal. All p values are substantially larger than 0.05. These results suggest there are no significant differences or patterns between raters at this distance and the only patterns to emerge from the 2x2 tables were that surgeons tended to rate more children at 1 (using wheelchairs) than both community and hospital physiotherapists, although the difference is not striking.

5.4. Discussion

5.4.1. Inter-rater agreement of the FMS

The FMS was shown to have good inter-rater agreement for the 5, 50 and 500 metre distances using a range of clinicians as raters. This supports the tool as a reliable measure of mobility for children with CP. Unweighted kappa statistics were used primarily for data analysis with the understanding that a difference of one level on the FMS was clinically meaningful and should be factored into the analysis. The results obtained using no weights are particularly promising because the use of weighted kappas produces a higher value, as evidenced by the weighted kappas calculated for 5, 50 and 500 metres. If inter-rater reliability is high it can be argued that it is unnecessary to test for retest reliability as it is bound to be higher (Streiner and Norman 2003). Retest reliability was not examined in the current study because the unweighted kappas calculated for inter-rater reliability showed substantial agreement.

The preliminary study examining the reliability of the FMS used ICC’s with results of 0.95 for 5 and 500 metres and 0.94 for 50 metres (Graham et al 2004). Weighted kappa using quadratic weights with ordinal data has been shown to yield equivalent results to that using the ICC (Tooth and Ottenbacher 2004).
Quadratic weighted kappas were calculated in the current study for each FMS distance to enable comparison with the previous study. They resulted in good agreement although the values are slightly lower than the Graham study. The current study examined reliability using a range of clinicians and more than 40 different raters. The previous study examined reliability between two surgeons and one research fellow only. This may account for some of the difference. The Graham study used ICC’s to analyse the data. These are designed primarily for use with interval or ratio data. Although they can be applied to ordinal data with the assumption that intervals are equivalent, the kappa statistic is more appropriate to analyse categorical data such as the FMS (Tooth and Ottenbacher 2004).

The results of the reliability of the FMS found in this study are comparable to the inter-rater reliability of tools that also focus on measuring or classifying activity (Russell et al 1989, Young et al 1995, Nordmark et al 1997, Palisano et al 1997, Novacheck et al 2000, Young et al 2000, McDowell et al 2007). The inter-rater reliability of the GMFCS has been shown as 0.55 for children younger than 2 years and 0.75 for children 2-12 years using unweighted kappas (Palisano et al 1997) and 0.57-0.75 using linear weighted kappas (McDowell et al 2007). The FAQ was shown to have good inter-rater reliability with ICC’s of 0.81-0.92 (Novacheck et al 2000). Inter-rater reliability of the GMFM is high with Kendall coefficients of 0.77-0.88 (Nordmark et al 1997) and ICC’s of 0.92-0.99 (Russell et al 1989). ICC’s of 0.96-0.99 was found for the ASK (Young et al 1995, Young et al 2000). The reliability of other tools can be more variable when there are several domains or subscales within them. For example, the inter-rater reliability of the PEDI contains mobility, self-care and social function domains. ICC’s vary from 0.18-0.94 (Nichols and Case-Smith 1996), 0.72-0.85 (Wright and Boschen 1993) and 0.64-0.74 (Berg et al 2004) with the mobility domain having higher ICC’s. Physical function may be more consistent and easier to score compared to domains such as social and cognition, particularly when parental report is involved.

The sample used in this study comprised children who were predominantly GMFCS level II or III, spastic diplegia and aged 6-12 years. An explanation of
this demographic distribution can be explained by the recruitment process through the CP clinics at the hospital. Many children attending the clinics and gait laboratory are those who are having or will have interventions such as orthopaedic surgery and spasticity management to improve their gait pattern and mobility. This largely involves children classified as GMFCS levels II-III, aged 6-12 years as reflected in the demographic displayed. The higher incidence of males reflects the epidemiological patterns of CP with males being at greater risk of congenital and acquired CP with ratios of 1.37:1 to 2.5:1 reported (Stanley et al 2000).

There were no strong patterns to emerge from the analysis of reliability in relation to different age groups, GMFCS levels or topographical distributions. The ratings from the 6-12 year group appeared to have higher agreement with all three distances having substantial agreement, however there were considerably more children in this group. The ratings from the younger children (2-6 years) had only fair agreement for 500 metres and moderate for 50 metres. The small number in that age group (16 children) may be a potential explanation for the lower values. Another factor is the possibility that these younger children were still developing some motor skills, particularly their walking. Further evidence of developing motor skills is found in the growth motor curves as described by Rosenbaum and colleagues (Rosenbaum et al 2002). From these curves, children with CP reach 90% of their potential GMFM-66 scores by age 3.5 if they are classified as GMFCS level IV, by age 3.7 for level III, by age 4.4 for level II and by age 4.8 for level I. The children aged 2-6 years in this study would still be changing and it is possible that inconsistency in motor skills between ratings may account for some of the disagreement. Examination of inter-observer agreement of the ratings from the GMFCS also showed higher results in older children (12-18 years) compared to younger children (4-12 years) (McDowell et al 2007).

Agreement was moderate or substantial for most GMFCS levels except for level I at 5 and 50 metres and level II at 5 metres where only fair agreement was obtained. The confidence intervals for these kappa statistics are wide. Therefore, caution needs to be exercised in reporting whether there were real differences
between levels. For some levels of the GMFCS, only two or three FMS levels were represented in the calculations, affecting the kappas obtained. This further clouds the ability to make strong statements about the results. However, there are possible explanations for the fair agreement of levels I and II. These GMFCS levels represent the more functionally able children. The difference between these two levels is based on the abilities of the child to perform more high levels activities such as running and incorporates concepts of balance, speed and coordination (Palisano et al 1997). It is possible that difficulties in distinguishing between these two levels would result in lower agreement. This is supported by previous studies where there is less agreement between GMFCS levels I and II (Palisano et al 1997, McDowell et al 2007). In relation to topographical distribution of CP, this study found substantial agreement between raters for children with diplegia and quadriplegia for all three distances and moderate agreement for children with hemiplegia. Like the results for GMFCS levels the confidence intervals are wide and the results need to be interpreted with caution.

The FMS can be administered in person or over the telephone as demonstrated by examination of different methods of administration. The scale is a self report or parent report scale and does not require direct observation to administer it. Results showed that for 5 and 50 metres, agreement was very similar for ratings where both were performed in person and where one of the ratings was performed over the telephone. For 500 metres there was a small difference with a higher kappa where one rating was performed over the telephone however the width of the confidence intervals suggests this is not a meaningful difference. All results for method of administration showed substantial agreement. This supports the reliable use of administering the FMS either in person or over the telephone and this enhances the clinical utility of the scale.

Similar findings were ascertained for different administration methods of the WeeFIM. There were no statistically significant differences on t tests when comparing administering the WeeFIM via interview over the telephone or in person (Ottenbacher et al 1996) or when comparing administration via direct observation and interview with a parent (Sperle et al 1997). These results
support the potential different administration methods of the WeeFIM. The clinical utility of the measures is enhanced by demonstrating that outcome measures can be administered by different methods. If the measure is able to be administered via the telephone, information collection for routine clinical care or for research purposes is more readily obtained.

5.4.2. Systematic bias of raters

The examination of systematic biases of raters proved to be primarily inconclusive. The kappa statistics obtained showed that all rater combinations had substantial or moderate agreement for each FMS distance. Wilcoxon signed-rank tests showed a trend towards the surgeons rating children higher at the higher end of the FMS than either community or hospital physiotherapists, however this was only significant for 5 metres. There were no trends evident between hospital or community physiotherapists. Closer examination of the agreement tables showed that on a number of occasions the surgeons rated more children as “6” on the FMS where the physiotherapists rated them as “5”.

The difference between 5 and 6 on the FMS is similar to the difference between GMFCS levels I and II. For 5 and 6 on the FMS and I and II on the GMFCS, all children are able to walk independently. Children in level II on the GMFCS and 5 on the FMS are independent on level surfaces only and require the use of a rail for stairs. Children in level I on the GMFCS and 6 on the FMS are independent on all surfaces including uneven ground, but have difficulty with balance, coordination and speed. Distinguishing between these two levels requires knowledge and appreciation of high level motor tasks and often involves more intricate probing when questioning the child or parent to distinguish their abilities. As stated earlier, research has shown that agreement and differentiating between GMFCS level I and II is lower (Palisano et al 1997, McDowell et al 2007). In the case of the FMS it is possible that the different background training of surgeons and physiotherapists may play a role in the trend of surgeons rating more children higher. Physiotherapists may have a greater appreciation of motor skills and high level activities and hence be able to question more thoroughly and distinguish the differences.
5.4.3. Factors affecting agreement

There are a number of reasons for potential differences in ratings affecting the reliability and agreement of the FMS. There are child/parent factors, rater/clinician factors and scale factors. The stability of mobility status of the child over the rating period and performance on the day of rating may impact the results. Thirty eight percent of children had both ratings performed on the same day where performance would be considered stable. The remaining 62% had ratings performed up to 2 weeks apart. Although a time interval of 2-14 days between testing is considered usual (Streiner and Norman 2003), there is a chance that mobility may have changed between ratings. This may have produced different scores, resulting in lower agreement between raters. Although not statistically significant due to the wide confidence intervals, the agreement of ratings performed on the same day was shown to be higher than those performed up to 2 weeks apart.

The FMS is administered by the clinician asking a few questions of the child and/or parents to obtain a “performance” rating of the child’s mobility. The manner in which the questions are worded is integral to the score obtained. Different raters/clinicians may word the questions slightly differently, resulting in slightly different answers. Some may ask vague questions in order to get a true “performance” rating while others may be more direct. Some may probe more intricately into differences between different levels, for example between levels 5 and 6. The pamphlet gives direction for administering the scale but does leave some room for clinical interpretation to ensure it is not too prescriptive or rigid. A study examining similar questions from three different tools (the CHQ, PODCI and PEDI) found that differences in wording of questions have a significant effect on parents’ responses (Wren et al 2007). Another factor is the response of the child or parent to the questions asked of them. On different occasions they may respond differently to very similar questions.

The knowledge of parents and community physiotherapists of the child’s mobility in different environments may also affect ratings. Because parents live with their children they would have first-hand knowledge of what their child
does at home and in the community, however they may not be so familiar with their mobility at school. Hospital physiotherapists or surgeons asking questions of the parent about their child’s mobility at school may obtain uninformed responses from the parent. Most of the community physiotherapists involved in this study see the children either at home or at school and therefore it is likely that they are more familiar with the child’s mobility at school or home but less so within the community. The issue of who reports for scales and how that affects accuracy will be discussed further in the grand discussion (Chapter Nine).

5.4.4. Limitations and recommendations

There are a number of limitations of this study. The sample size used was adequate to examine the overall inter-rater reliability of the FMS. However, because one of the aims of the study was to examine reliability for different age groups and GMFCS levels, it would be preferable to have a larger sample to ensure adequate numbers within each of those different groups to enable effective analysis. The sample comprised children aged 2-18 years with CP, GMFCS levels I-IV. The results cannot be extrapolated to children less than 2 years of age, adults, children classified as GMFCS level V or children with diagnoses other than CP.

Although a range of clinicians were used as raters, only two raters per child were allocated due to the possibility of responses being “rehearsed”. If this could be minimized it would be important to examine reliability with a larger number of raters per child. This would strengthen the FMS as a reliable tool amongst different clinicians and improve its’ generalisability. A large number of clinicians with differing clinical interests were represented in the rater group. Further examination of reliability with paediatricians, developmental physicians and rehabilitation physicians would be important to perform to further enhance the scale’s clinical utility. Another potential limitation of the study is the relatively high level of awareness of the scale among many of the raters used. Examination of reliability using raters with less exposure to the scale or with less clinical experience, such as students or newly graduated clinicians, would
determine how much experience is required to be able to administer the scale consistently.

A consecutive sample was used and this has the potential to introduce a biased sample that does not represent the population of interest. A population based sample would have minimised this bias. The clinics from which the children were recruited comprised the children that the FMS was routinely used for. Although a sample of convenience was used, there was a good chance the sample did in fact closely resemble the population the scale is intended for. The children were recruited from one centre only. It would be important to do further reliability testing using children from a number of centres and different types of centres including hospitals, schools and rehabilitation centres to enhance the generalisability of the results.

The recruitment procedure resulted in a sample with a bias towards children classified as GMFCS level II and III with diplegia and aged 6-12 years. The very mildly affected children who do not routinely attend those clinics and the more severely affected children who do not have interventions for gait correction surgery were under-represented. Further examination of the reliability of the FMS using a more even spread of GMFCS levels is warranted in order to be more representative of the CP population.

For some of the children in this study there was a gap up to two weeks between ratings. This gap was deemed to be sufficient to allow enough time for community physiotherapists to rate the children and return the information and not too long that change in the child’s mobility would occur in the mean time. There is the possibility that where the gap between ratings was 1-2 weeks, change did actually occur thus introducing a source of variation not attributable to the rater or the scale itself. A smaller gap, where practical, may minimize this risk.

The FMS has been devised for children with CP aged 4-18 years. It has the potential to be used in other populations such as myelomeningocoele and acquired brain injury. Reliability within those populations should be examined...
before it is widely used to measure their mobility. The scale also has the potential to be used in adults with CP and once again examination of reliability in adults is required beforehand.

5.5. Conclusion

Substantial agreement for each of the three distances of the FMS was found using a range of clinicians as raters. Although there were some differences in agreement for children of different ages, GMFCS levels and topographical categories, these differences were not significant. There was similar agreement when administering the scale in person and by telephone, suggesting that the FMS can be administered by either method. Closer examination of rater bias by clinical background showed a tendency of surgeons to rate more of the higher functioning children at the limit of the scale than physiotherapists, however only one of the comparisons was statistically significant. The FMS can be used as a reliable tool for different clinicians to assess mobility in children with CP.
CHAPTER SIX: RESPONSIVENESS TO CHANGE OF THE FUNCTIONAL MOBILITY SCALE

6.1. Introduction

This chapter reports the responsiveness to change of the FMS in children with CP following orthopaedic surgery and spasticity management using botulinum toxin injections. Evaluative measures such as the FMS are designed to measure the magnitude of longitudinal change for individuals or groups over time (Kirschner and Guyatt 1985). A fundamental component of the validating process of evaluative instruments is to establish responsiveness to change to ensure they can accurately detect change in function (Deyo and Inui 1984, Kirschner and Guyatt 1985, Guyatt et al 1987, Guyatt et al 1989, Rosenbaum et al 1990, Fitzpatrick et al 1992, Guyatt et al 1992, Lohr et al 1996). Evaluative measures are also used to quantify treatment benefits (Kirschner and Guyatt 1985). It is important to establish responsiveness to show that the tool is able to detect the effects of treatment over time (Furlong et al 2005).

There is a lack of clarity in the literature about the definition of responsiveness (Husted et al 2000, Terwee et al 2003). In general terms, responsiveness can be described as the ability of a measure to detect minimally clinical important differences in function when change is believed to have occurred (Guyatt et al 1987). It relates to changes in scores that are due to change in health status and not artefact of measurement (Guyatt et al 1992). A review of the literature on this topic identified 25 definitions of responsiveness which could be grouped into three categories (Terwee et al 2003). One category defined responsiveness as the ability to detect change in general, regardless of whether it was relevant or meaningful (Terwee et al 2003). A second category is the ability to detect clinically important change and requires a judgement on what is considered to be important change (Terwee et al 2003). The third category, an extension of the previous two, is the ability to detect real changes in the construct being measured (Terwee et al 2003). The third category requires a gold standard for what constitutes a real change in the construct being measured. This third
Chapter 6 – Responsiveness

definition is most relevant to evaluative measures because it is important to demonstrate that change has occurred (Vos-Vromans et al 2005).

Some authors have argued that responsiveness and validity are not separate properties (Liang 2000, Terwee et al 2003). They argue that responsiveness is an aspect of validity in a longitudinal setting because a responsive measure has the purpose to measure change over time and validity concerns whether the instrument is measuring what it intends to measure (Vos-Vromans et al 2005). Others argue there is a difference between longitudinal construct validity and responsiveness (Kirschner and Guyatt 1985, Guyatt et al 1989, Husted et al 2000). Longitudinal construct validity is the relationship between changes in an index and external measures over time whereas responsiveness is the power of the test to detect a clinically important difference (Kirschner and Guyatt 1985). The necessity for distinguishing between responsiveness and validity remains based on the fact that an instrument can be valid and still fail to detect clinically important change when it occurs (Guyatt et al 1989). For the purposes of this thesis, responsiveness is considered separate from validity and is operationally defined as “an instrument’s ability to detect important change over time in the concept being measured” (Dekker et al 2005).

Responsiveness can be classified into two major groups; internal and external responsiveness (Husted et al 2000). Internal responsiveness describes the ability of a measure to assess change over a prespecified time frame and is assessed using various methods such as effect size, paired t tests and responsiveness coefficients (Husted et al 2000). These methods are limited because they do not relate changes in the measure to corresponding changes on an external criterion at the individual patient level, and may report change without a true change occurring in the health condition being measured (Husted et al 2000). External responsiveness reflects the extent to which changes in a measure over a specified time frame relate to corresponding changes in a reference measure of health status. It is assessed using correlations, regression and receiver operating characteristic (ROC) curves (Husted et al 2000). External responsiveness has a broader application because it concerns the measure itself, not the treatment under investigation (Husted et al 2000). This is important to understand in the
context of this thesis because measures of external responsiveness were used to examine the responsiveness of the FMS. This was to determine that it could show change when a true change had occurred in the mobility of the children being assessed.

An important aspect of determining responsiveness is determining what constitutes a minimally clinical important difference (MCID) (Guyatt et al 1987). The MCID can be defined as “the smallest difference in score in the domain of interest which patients perceive as beneficial and which would mandate, in the absence of troublesome side effects and excessive cost, a change in patients management” (Jaeschke et al 1989). The MCID is not a fixed property of the instrument. It depends on the study setting in which it is going to be used (Terwee et al 2003). One issue with newly developed instruments and some established ones is that it is not known what change in score constitutes a clinically important difference (Guyatt et al 1987). In many clinical settings no “gold standard” exists for what represents a real change in clinical status (Deyo and Inui 1984). Defining a MCID for a specific study requires a judgement from someone as to what important change is, which is often subjective (Terwee et al 2003). For the purposes of this thesis, the MCID was defined by a small group of clinicians experienced in the use of the FMS.

A clinically important change can be defined and evaluated from the perspective of the patient, his or her proxy, society and the health professional (Liang 2000). In the absence of a gold standard, the independent, concurrent agreement about change by patient and physician can serve as a basis for defining the presence and magnitude of functionally important changes (Deyo and Inui 1984). However, a limitation of this method is that clinician judgements are not completely independent of patient judgements because the patients self-report to the clinician will influence that assessment (Deyo and Inui 1984). Using the patient perspective introduces the possibility of varying criteria for important changes (Ward et al 2000) and also introduces the possibility of response bias and reliability issues (Liang 2000). Therefore, any estimate of MCID will be associated with a degree of uncertainty (Guyatt et al 2002). For many measures used in clinical practice clinicians develop an intuitive sense of the MCID
through repeated use. Therefore, clinical experience with an instrument is a feasible and valid method of determining the significance of changes in instrument score (Jaeschke et al 1989).

There is a lack of consensus in the literature about the best methods to assess responsiveness with many authors using multiple indices of responsiveness (Husted et al 2000, Terwee et al 2003, Vos-Vromans et al 2005). A review of the literature of the different methods used for calculating responsiveness found 31 different methods (Terwee et al 2003) which had two different frames of reference (Vos-Vromans et al 2005). The first included methods based on the changes due to treatment and/or time effect such as effect size, standardised response means, paired t tests and analysis of variance. The second included methods in which an external criterion (patient or professional) was used to define whether a patient has changed or not, such as sensitivity and specificity using receiver operating characteristic (ROC) curves, correlation of change with an external criterion and regression models (Vos-Vromans et al 2005).

Many of the above methods for assessing responsiveness, such as mean change, t tests, effect size, standardised response means, rely on the data being continuous. The FMS is a categorical scale and these methods are therefore not appropriate. Appropriate methods for analysing categorical data include examining the magnitude of correlations of change with other measures (Guyatt et al 1989), by ensuring that scores improve with application of a treatment of known efficacy (Kirschner and Guyatt 1985) and by using ROC curves (Deyo and Centor 1986). If one can clearly demonstrate a group of subjects who are stable and group who have changed, responsiveness is readily measured (Guyatt et al 1989). ROC curves can assess the ability of a measure to reflect both change and no change in an external standard using sensitivity and specificity in detecting improvement or deterioration, however information on the magnitude of change in the external criterion is sacrificed because change must be dichotomised (Deyo and Centor 1986). In order to be sure that the instrument measures important changes, a correlational approach with some other measure is required (Terwee et al 2003).
For the current study, measures of external responsiveness appropriate for use with categorical data were chosen to examine the responsiveness of the FMS. Correlation of change scores on the FMS with change scores on an external criterion, the Functional Assessment Questionnaire (FAQ) was chosen. The FAQ is another measure of mobility and although it cannot be described as a “gold standard”, it also measures mobility in children with CP on a categorical scale.

Like validity, responsiveness is an abstract construct established incrementally over time by the accumulation of evidence about the performance of a measure (Rosenbaum et al 1990). A previous study on the FMS reported to assess sensitivity to detect change after operative intervention in children with CP (Graham et al 2004). Change in FMS scores was correlated with change on other measures such as Hoffer ambulation level, Uptime, CHQ, PODCI and energy expenditure with Spearman rank correlations ranging from 0.52-0.89. These correlations are encouraging; however the tools used as external criterions are not similar to the FMS. The FAQ, which is the most similar measure of mobility to the FMS, was not used as an external criterion. The pilot study reported in Chapter Four demonstrated that the FMS was able to detect change following SEMLS in children with spastic diplegia (Harvey et al 2007). That study examined change after surgery using the FMS, not the responsiveness of the scale.

It was therefore considered necessary to further examine responsiveness of the FMS using the FAQ as an external criterion to determine that it is able to detect clinically important change where that change occurs. Because a useful measure must reflect “no change” as well as it does “change” (Husted et al 2000), it was considered necessary to prospectively show that the FMS is able to detect clinically important change in mobility status as well as show stability where no change occurs. To do this, a group of children with CP following single event multilevel surgery who have been shown to change would be compared with children having botulinum toxin injections where clinical experience suggests that mobility does not change afterwards.
The research question was “what is the responsiveness to change of the FMS for detecting clinically important change in children with CP following orthopaedic surgery and spasticity management”. The aim of this study was to examine the ability of the FMS to detect minimal clinically important difference in mobility status, judged as one level on the FMS, in a group of children with CP having a range of interventions using the FAQ as an external criterion.

The hypothesis was “the FMS is able to detect clinically important differences in functional mobility in children with CP where change in mobility occurs; it will show change in children having SEMLS and will show stability in children having botulinum toxin injections”.

6.2. Method

This study was prospective and longitudinal. Data were collected for a group of children at predetermined stages before and following different interventions involving surgery and spasticity management.

6.2.1. Participants

The participants were a consecutive population-based sample of children with CP who were due to have interventions including spasticity management and orthopaedic surgery. Consecutive population-based sampling was used to ensure a large enough sample size was obtained for a representative group that would show changes in mobility in the children as a group and for smaller subgroups representing different types of interventions. It also ensured that adequate time for follow-up after the interventions was considered. Although a random sample would ensure that the sample obtained represented the target population, it was considered that this was not feasible within the time constraints of the thesis. Also, by recruiting the children through the clinics of The Royal Childrens Hospital where surgical decision-making occurs, the final sample was representative of the population of interest.
6.2.1.1 Inclusion criteria

Inclusion criteria were:

i. children with CP aged 2-18 years
ii. children GMFCS levels I-IV
iii. children attending the CP orthopaedic surgical clinic or gait laboratory of The Royal Childrens Hospital
iv. children scheduled to have orthopaedic surgery or spasticity management between January 2006 and May 2007

The upper age limit was 18 years because the FMS has been developed for children aged 4-18 years. This was explained in Chapter 4 (section 4.2.1). Although the lower age limit for the scale is 4 years, this study did include children aged 2 years and older. This was because a particular group of interest was the younger children who have botulinum toxin injections for spasticity management. This intervention is often used in younger children aged 2-6 years and therefore these younger children were included. Children classified as GMFCS levels I-IV were included. These are the levels that the FMS has been developed for (see Chapter 4 section 4.2.1). The CP orthopaedic surgical clinics and the gait laboratory of The Royal Childrens Hospital are the major clinics where decisions regarding forthcoming surgery for children with CP are made. This is where the children were recruited from. The children were required to be having surgery between January 2006 and May 2007 to allow for adequate time post-intervention for repeat assessments.

The interventions performed on the children examined in the study were:

i. single event multilevel surgery (SEMLS) for gait correction, involving children with diplegia and hemiplegia
ii. single level orthopaedic surgery for deformity correction, for example, foot surgery or hamstring lengthenings, in isolation
iii. two level simple orthopaedic surgery, for example hamstrings and calf lengthening in combination
iv. surgery for hip displacement;
   a. adductor lengthenings with phenol to the obturator nerve and
b. varus derotation osteotomies of the femur (VDRO’s)
v. botulinum toxin injections into specific muscles for spasticity management, at either one, two or three levels and involving:
   a. calves
   b. hamstrings
   c. adductors
   d. psoas

Single event multilevel surgery (SEMLS) refers to the correction of all orthopaedic deformities in one session (Graham and Selber 2003). It can be defined as at least two orthopaedic procedures at different anatomical sites in each limb i.e. a minimum of four procedures (Pirpiris et al 2003). It is now considered to be the most accepted approach to correct the musculoskeletal deformities contributing to gait deviations (Browne and McManus 1987, Nene et al 1993, Abel et al 1999, Fabry et al 1999, Saraph et al 2002, Bache et al 2003, Graham and Selber 2003). The frequently used procedures are muscle-tendon lengthenings, tendon transfers, rotational osteotomies and bony stabilisation procedures (Bache et al 2003). These procedures can also be performed in isolation or in smaller combinations, such as single level or two level simple surgery when the deformities are not as extensive.

Injections of Botulinum toxin A are used to treat spasticity in selected muscle groups by resulting in temporary chemodenervation of muscle and improving muscle length and joint range of motion (Cosgrove et al 1998). Injections are indicated primarily in younger children aged between 2 and 6 years (Cosgrove et al 1998, Graham et al 2000) to prevent the development of fixed contractures (Graham and Selber 2003). Applications in children with CP include dynamic equinus, adductor spasticity causing a scissoring gait and hamstrings spasticity causing crouch gait (Corry et al 1998, Cosgrove et al 1998, Graham and Selber 2003). One aim of the injections is to defer the need for more extensive deformity correction until the child is at an optimal age where the most benefits are obtained from surgical intervention (Bache et al 2003).
Injection of Phenol to the motor nerves is another option in spasticity management (Bache et al 2003). Phenol has a small role as a neurolytic agent for selected motor nerves such as the obturator nerve in the presence of adductor spasticity (Graham and Selber 2003). For children with more advanced hip displacement, derotation osteotomies of the femurs are performed for medial femoral torsion and hip subluxation (Ounpuu et al 2002).

It is not within the scope of this thesis to describe in detail the range of orthopaedic procedures used. For more information on the types of orthopaedic procedures and their indications the reader is directed to the relevant literature (Graham et al 2000, Gage and Novacheck 2001, Bache et al 2003, Graham and Selber 2003, Davids et al 2004, Sussman and Aiona 2004). Relevant articles on the use and indications of botulinum toxin injections are also available (Cosgrove et al 1994, Graham et al 2000, Boyd and Hays 2001a, Boyd and Hays 2001b, Baker et al 2002).

The orthopaedic surgeons responsible for each child’s management prescribed the surgery and performed the surgical operations or injected the botulinum toxin.

6.2.1.2 Exclusion criteria

Excluded were children:

   i. aged younger than 2 years or older than 18 years
   ii. with a diagnosis other than CP
   iii. classified as GMFCS level V
   iv. whose parents were not able to understand due to language or cognitive problems
   v. whose parents were upset or anxious associated with their clinic or gait laboratory appointment.

Children whose parents were not able to comprehend what was required due to language or cognitive problems were excluded. Also excluded were children who themselves or their parents were too upset or anxious at the clinic appointment. This was explained in Chapter 5 (section 5.2.1.2).
6.2.1.3 Sample size

A sample size of 80-100 was aimed for. This was considered to be adequate for exploring the responsiveness of the FMS over different time points because a previous pilot study of the FMS examining change after multilevel surgery showed that change was detected in a sample of 66 children with CP (Harvey et al 2007). Another aim of this study was to compare change in mobility following SEMLS with change following botulinum toxin injections. The sample needed to include at least 20 children having each of those interventions to enable adequate comparison. The sample size obtained was partly driven by the practicalities of recruiting children through the hospital with the clinical availability of participants having interventions. It was also dependent on allowing adequate follow-up time for the subsequent assessments within the timeframe of thesis completion. Based on the number of children who have surgery and botulinum toxin injections per year at The Royal Children’s Hospital, this number was considered to be achievable.

6.2.2. Procedure

The study was approved by the Ethics in Human Research Committee of the Royal Children’s Hospital (reference number 25119A) and The University of Melbourne Human Ethics Research Committee (reference number 060212). Written consent was obtained from each participating child’s parent or legal guardian prior to data collection.

6.2.2.1 Recruitment

Children were recruited through the CP orthopaedic surgical clinics and the gait laboratory of The Royal Children’s Hospital. The primary investigator (AH) obtained the operating, clinic and gait laboratory appointment lists in advance of the children attending the clinics to determine eligibility for entry. Once eligibility was determined, the parents of the children were approached prior to the interventions and invited to participate following a full explanation of the study. Consent forms were signed and a written information sheet provided to those willing to participate. The primary investigator (AH) was responsible for collection of the information pre-operatively and at each post operative time-point.
6.2.2.2 Testing procedure

The data collected for each child at the initial pre-operative time point included:

   i. date of birth, age and gender,
   ii. GMFCS level,
   iii. FMS score,
   iv. Gillette Functional Assessment Questionnaire (FAQ) score,
   v. date of surgery and type of surgery.

The GMFCS level for each child was determined by the primary investigator based on the descriptors for each level (Appendix B). Date of birth, gender and topographical distribution of CP for each child was obtained from the hospital records. The FMS was administered with the child and parents by the primary investigator. The walking scale of the Gillette FAQ is a 10 level ordinal scale that is completed by the parents. A copy of this scale is in Appendix E. It was completed at each assessment with the primary investigator present. The date of surgery and type of surgery was obtained from the hospital record at the pre-operative consultation and checked against the operation report at the first follow-up assessment. The age, GMFCS levels and type of surgery were collected for descriptive purposes. The FMS and FAQ were collected for data analysis of responsiveness after the interventions.

The children were then assessed at regular time points following the interventions. The post-operative follow up time points varied according to the interventions. The children were assessed at:

   i. SEMLS; 3, 6, 9 and 12 months post-operatively.
   ii. one-two level surgery - 3 months and 6 months post-operatively.
   iii. adductor lengthening with phenol to the obturator nerve - 6 weeks, 3 and 6 months post-operatively.
   iv. VDRO’s - 3, 6, 9 and 12 months post-operatively
   v. botulinum toxin injections - 3, 6, 12 and 24 weeks post-injections.

The different time points were determined based on the amount of surgery performed, the expected post-intervention recovery period based on clinical
judgement and the routine post-intervention clinical follow-up that occurs. Children who undergo SEMLS require at least 12 months of rehabilitation and changes in mobility are seen up until 24 months (Harvey et al 2007). In the clinical setting these children are monitored every 3 months for the first 12 months. Children who have surgery to one or two levels tend to recover sooner because of the lesser amount of surgery. Following adductor lengthening and phenol to the obturator nerve, improvements may be seen earlier, thus requiring a 6 week assessment time point. Children who are injected with botulinum toxin are able to return to usual activity as soon as they return home and the effects of the injections can be seen by 3 weeks post-injection with the usual length of time the benefit continues is up to 6 months (Cosgrove et al 1994, Eames et al 1999). These children were assessed at more frequent points earlier after the injections.

At each of the follow-up assessments the information collected was:

1. GMFCS level
2. FMS score
3. FAQ score

The GMFCS classification was obtained while observing the child and questioning the parents. The FMS was administered by the principal investigator. The FAQ was completed by the parents. The information was collected at the usual post-operative appointments in the clinic or the gait laboratory by the principal investigator. It was not possible to blind the principal investigator to the treatment path of the children because post-operative wounds and the age of the child would make it obvious as to which group they were in. Some of the children who had received botulinum toxin injections or single level surgery were not required to attend the hospital for their routine clinical appointments as often as the protocol of the collection of data for this study. To prevent the child and family having to attend the hospital more than clinically necessary, an alternative method of data collection was employed. This involved the principal investigator administering the FMS over the telephone with the parents and mailing the FAQ with a stamped, self-addressed envelope out to the parents who then completed it and returned it.
Wherever feasible the same parent filled in the FAQ and answered the questions for the FMS rating. The principal investigator performed all telephone FMS ratings and the clinic appointment ratings.

6.2.3. Data Analysis

Descriptive statistics for participant characteristics were calculated using means and standard deviations for continuous variables and frequency and percentages for categorical variables. A decision was made by a group of four clinicians with extensive experience of administering the FMS, including one surgeon and three physiotherapists, that a change of one category (up or down) on the FMS was considered a minimal clinically important difference (MCID). This decision was made based on the literature discussed in the introduction of this chapter that clinical experience with a measure is a valid method of determining the MCID where there is no “gold standard” for what represents a real change in clinical status (Jaeschke et al 1989). This MCID formed the basis for reporting the change scores.

The FAQ was used as an external criterion to assess change following the interventions. The FMS and FAQ are both categorical measures. It is therefore not appropriate to use statistical methods such as effect size, standardised response means and mean change as has been used for continuous variables (Terwee et al 2003). It was decided to use change scores (follow-up score minus initial score) as a measure of responsiveness (Deyo and Inui 1984). Change scores on the FMS and FAQ were generated at each post-intervention stage. The percentage of children who either did not change or changed for the better (up) or for the worse (down) were calculated at each time point. These descriptive statistics were calculated for the group as a whole and the results presented.

In addition, Spearman rank correlation coefficients were calculated for the correlation of change scores on each distance of the FMS with the FAQ over each time period for the group as a whole. Spearman’s rho is a non-parametric analog of the Pearson r (Siegel and Castellan 1988). It is appropriate for use with categorical data such as the FMS and FAQ and is a measure of association.
Spearman correlation coefficients were calculated after change scores for the FMS and FAQ were grouped into no change, improved (change up) or deteriorated (change down).

Descriptive change score statistics were also calculated for the group who underwent SEMLS in isolation and compared with the group who had botulinum toxin injections in isolation. This comparison was performed as potential further evidence of the responsiveness of the FMS. Clinical experience suggests that children who have botulinum toxin injections do not change dramatically in their mobility status afterwards. On the other hand, the mobility of children who have multilevel surgery does change significantly, initially for the worse and then for the better throughout the post-operative period (Harvey et al 2007). Not only is it important that the FMS is able to detect change where it actually occurs, it is important to show that it shows no change in mobility status when the child does not change.

The data were stored and organized using the EpiData for Windows program ([http://www.epidata.dk/](http://www.epidata.dk/)). All analyses were performed using Stata [StataCorp 2005 Stata statistical software Release 9.0. College Station; TX. Stata Press.].

6.3. Results

6.3.1. Descriptive results of participants

A total of 96 children were recruited for the study. Nine children dropped out of the study throughout the course of follow-up assessments. This was due to parents not returning the FAQ in the mail. Three children were recruited and subsequently did not have their intervention. This left a sample size of 84 children.

Of the remaining 84, the age range was 2-16 years with a mean age of 8.8 years (SD 3.28). There were 59 males and 25 females. Eight children had hemiplegia, 63 had diplegia and 13 had quadriplegia. The spread of GMFCS levels pre-operatively was; 10 children level I, 31 children level II, 35 children level III
and 8 children level IV. Table 6.1 summarises the descriptive characteristics of the sample.

Table 6.1 Participant characteristics

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>n</th>
<th>84</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (y,m), mean (SD)</td>
<td>Total</td>
<td>8.8 (3.3)</td>
</tr>
<tr>
<td>Sex, n (%)</td>
<td>Male</td>
<td>59 (70%)</td>
</tr>
<tr>
<td></td>
<td>Female</td>
<td>25 (30%)</td>
</tr>
<tr>
<td>GMFCS, n (%)</td>
<td>I</td>
<td>10 (12%)</td>
</tr>
<tr>
<td></td>
<td>II</td>
<td>31 (37%)</td>
</tr>
<tr>
<td></td>
<td>III</td>
<td>35 (42%)</td>
</tr>
<tr>
<td></td>
<td>IV</td>
<td>8 (9%)</td>
</tr>
<tr>
<td>Topography</td>
<td>Hemiplegia</td>
<td>8 (9%)</td>
</tr>
<tr>
<td></td>
<td>Diplegia</td>
<td>63 (75%)</td>
</tr>
<tr>
<td></td>
<td>Quadriplegia</td>
<td>13 (16%)</td>
</tr>
</tbody>
</table>

The numbers of children within each category of intervention were;

i. 24 (29%) with bilateral SEMLS (diplegia)

ii. 25 (30%) with botulinum toxin injections

iii. 35 (41%) with other types of surgery including; unilateral SEMLS, adductor lengthening and Phenol injections, VDRO’s, single and two level surgery.

All children were used to show change for the group as a whole and for correlations of change scores between the FMS and FAQ. The children who had SEMLS and Botulinum toxin injections were separated out to calculate percentage of those who changed at each post-intervention time point.

6.3.2. Descriptive statistics of change scores

The children were grouped according to those who showed no change, those who changed up one or more level (changed for the better) and those who changed down one or more level (changed for the worse). The children were grouped into change up or down based on the earlier decision that change of at least one level on the FMS was clinically meaningful. The percentage of
children in each of those categories at each post-operative time point (compared to baseline) were calculated and these figures are presented in Table 6.2.

This table shows that for the group as a whole there were more than 50% of children who did not change at each time point on the FMS. On the FAQ there was more change at each time point with less than 50% of children showing no change. For the group, there were more children showing deterioration than improvement at the first post-operative time point compared to pre-operatively. There were a relatively even number of children who deteriorated and improved at the second post-operative time point. More children improved than deteriorated at both the third and fourth post-operative time points.

Table 6.2 Percentage of children in change categories for the FMS and FAQ at post-intervention time points

<table>
<thead>
<tr>
<th>Time</th>
<th>1st time-pre</th>
<th>2nd time-pre</th>
<th>3rd time-pre</th>
<th>4th time-pre</th>
</tr>
</thead>
<tbody>
<tr>
<td>Change</td>
<td>0 ↓ ↑</td>
<td>0 ↓ ↑</td>
<td>0 ↓ ↑</td>
<td>0 ↓ ↑</td>
</tr>
<tr>
<td>All children (n=84)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>5</td>
<td>60</td>
<td>33</td>
<td>7</td>
<td>74</td>
</tr>
<tr>
<td>50</td>
<td>57</td>
<td>38</td>
<td>5</td>
<td>67</td>
</tr>
<tr>
<td>500</td>
<td>67</td>
<td>27</td>
<td>6</td>
<td>70</td>
</tr>
<tr>
<td>FAQ</td>
<td>40</td>
<td>47</td>
<td>13</td>
<td>49</td>
</tr>
<tr>
<td>SEMLS (n=24)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>5</td>
<td>29</td>
<td>62</td>
<td>9</td>
<td>54</td>
</tr>
<tr>
<td>50</td>
<td>37</td>
<td>59</td>
<td>4</td>
<td>58</td>
</tr>
<tr>
<td>500</td>
<td>42</td>
<td>50</td>
<td>8</td>
<td>54</td>
</tr>
<tr>
<td>FAQ</td>
<td>25</td>
<td>67</td>
<td>8</td>
<td>52</td>
</tr>
<tr>
<td>BOTOX (n=25)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>5</td>
<td>88</td>
<td>4</td>
<td>8</td>
<td>92</td>
</tr>
<tr>
<td>50</td>
<td>84</td>
<td>12</td>
<td>4</td>
<td>80</td>
</tr>
<tr>
<td>500</td>
<td>96</td>
<td>4</td>
<td>0</td>
<td>100</td>
</tr>
<tr>
<td>FAQ</td>
<td>64</td>
<td>20</td>
<td>16</td>
<td>64</td>
</tr>
</tbody>
</table>

0; no change, ↓; changed down (worse), ↑; changed up (better)

When the children who had SEMLS were compared with those who had Botulinum toxin injections, a definite pattern emerged. For each time point post-operatively there were a greater number of children who showed no change on the FMS and FAQ in the Botulinum toxin group. More children who had SEMLS showed change; particularly at the first time point-postoperatively where more than 50% of children deteriorated on all three FMS distances.
the third post-operative time point (9 months after SEMLS) more children were showing improvement than deterioration. In contrast, many of the children who had botulinum toxin injections showed no change, particularly for the 500 metre FMS distance. The FAQ followed similar patterns, albeit with less children showing no change.

The changes seen in the children who had SEMLS compared to those who had Botulinum toxin can be seen clearly in Figures 6.1-6.6. The bar graphs in Figures 6.1-6.3 show that the mobility status of children as measured by the FMS in many children did not change following Botulinum toxin injections, particularly for the 5 and 500 metre distances. The bar graphs in Figures 6.4-6.6 tell a different story with more children showing change in mobility status following SEMLS, particularly the deterioration (change down) at the first post-operative time point. On these graphs, \textit{no change} represents the percentage of children who did not change compared with pre-operatively. \textit{Up} represents the percentage of children who improved one or more level compared with pre-operatively, for example changed from using a walker to using crutches. \textit{Down} represents the percentage of children who deteriorated one or more level compared to pre-operatively, for example changed from using a walker to using a wheelchair.
Figure 6.1 Change in FMS 5m score for Botulinum toxin group

Figure 6.2 Change in FMS 50m score for Botulinum toxin group

Figure 6.3 Change in FMS 500m score for Botulinum toxin group
Figure 6.4 Change in FMS 5m score for SEMLS group

Figure 6.5 Change in FMS 50m score for SEMLS group

Figure 6.6 Change in FMS 500m score for SEMLS group
6.3.3. Correlations between change scores on the FMS and FAQ

The results of the Spearman rank correlation coefficients for all children are presented in Table 6.3. This table shows that the correlation coefficients were generally low with only six comparisons statistically significant. Change scores on the FAQ were associated significantly with change scores on the FMS for the 5, 50 and 500 metre distances in the first time period post-operatively and for 50 metres at the third and fourth post-operative stages.

<table>
<thead>
<tr>
<th>Change period</th>
<th>FMS distance</th>
<th>Rho</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Change from first post-op stage to pre-op</td>
<td>5</td>
<td>0.28</td>
<td>0.0088*</td>
</tr>
<tr>
<td></td>
<td>50</td>
<td>0.36</td>
<td>0.0007*</td>
</tr>
<tr>
<td></td>
<td>500</td>
<td>0.23</td>
<td>0.04*</td>
</tr>
<tr>
<td>Change from second post-op stage to pre-op</td>
<td>5</td>
<td>0.17</td>
<td>0.1282</td>
</tr>
<tr>
<td></td>
<td>50</td>
<td>0.18</td>
<td>0.1075</td>
</tr>
<tr>
<td></td>
<td>500</td>
<td>0.06</td>
<td>0.6016</td>
</tr>
<tr>
<td>Change from third post-op stage to pre-op</td>
<td>5</td>
<td>0.01</td>
<td>0.5815</td>
</tr>
<tr>
<td></td>
<td>50</td>
<td>0.26</td>
<td>0.03*</td>
</tr>
<tr>
<td></td>
<td>500</td>
<td>0.04</td>
<td>0.7391</td>
</tr>
<tr>
<td>Change from fourth post-op stage to pre-op</td>
<td>5</td>
<td>0.05</td>
<td>0.7279</td>
</tr>
<tr>
<td></td>
<td>50</td>
<td>0.29</td>
<td>0.05*</td>
</tr>
<tr>
<td></td>
<td>500</td>
<td>0.22</td>
<td>0.14</td>
</tr>
</tbody>
</table>

* denotes statistically significant result

Although these showed associations, the correlations were low, with the highest correlation 0.36. The Spearman correlations examined the association between change scores on the two measures. There would be a trend towards low coefficients where there were a higher percentage of children showing no change. This may explain some of the very low correlations.

6.4. Discussion

6.4.1. Responsiveness of the FMS

The results of this study provide some evidence of responsiveness to change of the FMS. The descriptive results of the change scores demonstrated more change in children following SEMLS compared to those having botulinum toxin injections. The correlations of change scores on the FMS with the FAQ were
lower than expected. There are a number of potential reasons why these correlations were low and these will be explored below. Although this study adds to the evidence of responsiveness of the FMS, it has also highlighted issues in assessing and reporting responsiveness, suggesting further investigations are required.

The differences in changes reported for the children having SEMLS and those having botulinum toxin injections are important for demonstrating that the FMS is able to detect both change and stability in the mobility status of children with CP. In order for a measure to be responsive, it is necessary for it to be able to detect clinically important change as well as show no change where status of the concept in question is stable (Husted et al 2000). The results of this study support the ability of the FMS to detect different levels of mobility changes in children with CP after different interventions.

The low correlations of change scores of the FMS and FAQ possibly reflect the differences between the two measures. The walking scale of the FAQ is a one item 10 level ordinal scale where one category is chosen to reflect the child’s usual walking ability. The FMS is a six level ordinal scale with three different items reporting three different levels of mobility according to environmental setting. The FAQ is a parent report measure and is completed by the parent. The FMS is a clinician interpretation of child or parent report and is administered by the clinician asking a few questions of the child or parent. Although they are both measuring mobility, the methods of reporting differ. Correlations were performed for all three FMS items with the FAQ. It is interesting to note that there were higher correlations of the FAQ with the 50 metre FMS distance. This suggests that of the three FMS distances, the 50 metre distance is most closely related to the FAQ.

The greater number of levels to select from on the FAQ may have resulted in a higher variability of categories chosen. This is reflected in the results with an overall smaller percentage of children showing no change on the FAQ compared to the FMS. With more categories to choose from there is a smaller chance of selecting the same category each time. Low correlations of change
scores between the FMS and FAQ were shown in this study despite good concurrent validity of the two measures as shown in Chapter Eight of this thesis. Spearman correlation coefficients of the FAQ with FMS 5 metres was 0.69, 0.76 for the 50 metre distance and 0.79 for 500 metres (see Chapter Eight). This suggests that in stable conditions the two scales are measuring similar constructs, yet in detecting change over time they have differing abilities.

The FAQ was chosen as an external criterion to demonstrate change in mobility. The differences in how and what the scale measures compared to the FMS may suggest that it is not the most ideal external criterion to use. The biggest difference between the two is that children can improve or deteriorate in mobility as measured by the FMS and remain in the same category on the FAQ. This is because the FAQ does not consider the different assistive devices the children use. For example, if a child progresses from using a walker to using crutches around the home and at school following orthopaedic surgery they will go up a level on the FMS (from 2 to 3) for the 5 and 50 metre distances. It is quite likely that their FAQ level will remain the same because the change in assistive device will not be picked up by the FAQ. This implies that although both scales are measuring mobility, their underlying constructs are different and in fact they are measuring different aspects of mobility. There is, however, no other scale or measure that can be considered a “gold standard” for mobility that is equivalent to the FMS to use as an alternative external criterion.

A weakness of relying on an external standard to assess responsiveness is that a new outcome measure, such as the FMS, may be designed specifically because it reflects a different aspect than currently available measures (Husted et al 2000). This may be a reason why studies choosing to use an external criterion to assess responsiveness have previously chosen parental or clinician rating of change as an external criterion over using a different measurement tool.

Other methods of assessing responsiveness that do not require the use of an external criterion, such as effect size and standardised response means, are not appropriate for categorical data. These methods tend to assess the magnitude of treatment effect rather than the ability of the tool to detect clinically important
differences. Assessing measures using the magnitude of treatment effect does not provide information about the quality of the instrument to serve its purpose, that is, to detect clinically important change where it exists (Terwee et al 2003). Studies assessing the responsiveness of other tools that measure activity limitation have used varied statistical techniques and many of them assessed treatment effects rather than responsiveness of the tool (Nordmark et al 2000, Ottenbacher et al 2000, Young et al 2000, Damiano et al 2005, Vos-Vromans et al 2005, Wright et al 2005, McMulkin et al 2007). Some of these studies used a number of different statistical methods comprehensively with continuous data, however these methods do not provide evidence that real change has actually occurred.

This investigation chose methods that assess external responsiveness which reflects the extent to which changes in a measure relate to corresponding changes in a reference measure (Husted et al 2000). In the absence of a gold standard, as is often the case in health measurement, it has been suggested that using patient or clinician judgement as a criterion is adequate (Deyo and Inui 1984). Studies have used correlations between parents and clinicians reports of change with variable results (Daltroy et al 1998, Vos-Vromans et al 2005). Using the perspectives of clinicians and parents can be subjective and potentially introduces biases in reporting. The reliability and validity of such reporting systems was not investigated in these studies. It is possible for an instrument which includes some subjective components to detect differences between interventions and thus demonstrate responsiveness without being a valid measure of overall health status (Guyatt et al 1989). These methods using subjective measures of change were not chosen for the current study because of the lack of evidence of reliability and validity of such methods. Determining the best methods for assessing responsiveness of evaluative tools remains an ongoing process with further work in the area required.

Determining the best methods for examining responsiveness relates to selecting the most appropriate tools for the construct being measured. If there is no change using a particular tool, it does not necessarily imply that no change has occurred in the child. It may indicate that there has been no change in the
specific construct measured by that tool. For example, in this study a large percentage of children who had botulinum toxin injections did not change in mobility status. This was expected because the treatment aims to reduce spasticity in focal muscle groups and clinical experience suggests mobility is not affected. Measures of dynamic range of movement and joint kinematics would more likely demonstrate change without a change in FMS score. Consequently, change not detected does not necessarily indicate a tool is not responsive. It may indicate the tool is not designed to measure the construct in question. When selecting measurement tools to assess the effects of interventions, clinicians must consider what the tool is designed to measure as well as the aims of the intervention. They then should match the tools to these aims. This will be discussed further in the grand discussion in Chapter Nine of this thesis.

The FMS was designed to measure the different assistive devices children use for mobility in different environmental settings. It is particularly useful for the group of children with CP who have SEMLS for gait correction (Harvey et al 2007). In this group the FMS is able to detect both the initial deterioration and consequent improvement in mobility status throughout the post-operative rehabilitation period. This was reported in the pilot study in Chapter Four. For this reason the group having SEMLS were chosen to determine if the FMS was able to detect those changes in comparison to a relatively stable group consisting of children having botulinum toxin injections.

6.4.2. Limitations and recommendations

There are a number of limitations as well as recommendations for further examination of responsiveness of the FMS. Firstly, the sample used was a consecutive sample from one centre only. There was no randomisation of the children recruited. The justification for this sampling method was stated in section 6.2.1. Randomisation would have minimised the potential sampling bias of not representing the population of interest. However, the clinics through which the children were recruited manage all children who require orthopaedic surgery and spasticity management. It was thus felt that recruiting children over
Chapter 6 – Responsiveness

a 12 month period through these clinics would be a fair representation of children.

The sample used comprised children with CP, aged 2-18 years and classified as GMFCS level I-IV. There was a higher representation of children with spastic diplegia as well as those classified as GMFCS levels II or III. This reflects the demographic of children who are appropriate for spasticity management and orthopaedic surgery for gait correction. The results of this study can therefore only be generalised to this age group and this demographic of children. Also, the children within each group were different due to the type of intervention they received. As a result, the children in the spasticity management group were younger and more likely to be susceptible to natural change and development. Further investigations of the responsiveness of the FMS should involve children from different centres; with a more even spread of GMFCS levels that represent the population of children with CP. It is also important to demonstrate that the FMS is able to detect changes in mobility after other treatments such as physiotherapy and other forms of spasticity management as well as changes that occur as the children grow and develop over time that are not related to interventions. This would involve both improvements as children gain motor skills as well as potential deterioration as they develop increased spasticity, muscle contractures and bony deformities.

The FMS is measured on a categorical scale and this limits the range of statistical methods that can be used to analyse the data for responsiveness. Methods using an external criterion (the FAQ) were chosen as well as descriptive statistics on the percentage of children who changed. The results suggest the FAQ is not the most appropriate external criterion because it measures different constructs to the FMS. A more suited external criterion may have resulted in higher correlations of change scores. The FAQ was chosen because, like the FMS, it measure mobility in isolation. In hindsight, consideration of other tools such as the mobility domains of the PEDI (Haley et al 1992) or the ASK (Young et al 1995) may have been more suitable as an external criterion to correlate with the FMS.
As well as considering different options for an external criterion, the results may have been strengthened by using ROC curves for the data analysis. They were not used in this study because of a lack of a defined stable group of participants to which change could be compared. The children having botulinum toxin were chosen as a group that would show less change than those having SEMLS, however this was based on clinical experience only. There is no direct evidence to suggest this group is completely stable in their mobility status following the injections. In hindsight, this study could have used a group of children having no interventions (a stable group) to compare with children having various interventions. Receiver operating characteristic curves could then have been calculated to determine the responsiveness of the FMS. Another option for categorical data is to use regression models to determine external responsiveness (Husted et al 2000).

For the purpose of this study, the MCID of the FMS was determined prior to data collection and analysis and defined as one level up or down on the FMS. This was determined by consensus of three clinicians with extensive experience in FMS administration. In the absence of a “gold standard” of what represents a real change in clinical status (Deyo and Inui 1984), this method was considered appropriate for the current study. Future investigation of what is considered a MCID for the FMS could involve consensus methods using a larger number and range of clinicians from different centres who have some experience of using the FMS.

6.5. Conclusion

The FMS was able to detect changes in mobility in children with CP as defined by one level of improvement or deterioration. The FMS was able to detect more change in children who had SEMLS compared to those who had botulinum toxin injections. Correlations of change scores with an external criterion were low; however this may reflect the properties of the criterion chosen rather than the responsiveness of the FMS itself. This study provides some evidence of the ability of the FMS to detect clinically important change. Further investigations are required into the responsiveness of the FMS to ensure that this essential property of an evaluative measure is evident in a range of clinical settings.
CHAPTER SEVEN: OBSERVATION OF MOBILITY IN CHILDREN WITH CEREBRAL PALSY: A STUDY OF CONSTRUCT VALIDITY OF THE FUNCTIONAL MOBILITY SCALE

7.1. Introduction

The primary aim of this chapter is to describe the methodology and results of an investigation of one aspect of construct validity of the FMS by comparing usual administration of the scale with direct observation. Construct validity refers to the extent to which scores on a particular instrument relate to other measures in a manner consistent with theoretically derived hypotheses concerning the concepts that are being measured (Kirschner and Guyatt 1985, Streiner and Norman 2003). A construct is the extent to which a variable is abstract rather than concrete (Nunnally 1978) and is often thought of as a “mini-theory” to explain the relationships among various behaviours or attitudes (Streiner and Norman 2003).

Construct validation involves a hypothesized relationship between a supposed measure of a construct and a particular observable variable (Nunnally 1978). It involves specifying the domain of observables related to the construct, determining the extent to which the observables tend to measure the same thing, and performing studies to determine the extent to which measures of the construct produce results which are predictable from the theoretical hypotheses concerning the construct (Nunnally 1978). Construct validity can be assessed by testing predefined hypotheses about expected correlations between measures or expected differences in scores between known groups (Terwee et al 2007). It often involves theorizing intuitively how the variables relate to one another and usually comes down to circumstantial evidence for the usefulness of a new measurement method (Nunnally 1978).

For the FMS, the construct is mobility, with particular reference to the amount of assistance required for mobility in different environmental settings. Uniquely,
the FMS measures how the same child might use more than one method of mobility as determined by the environmental setting. The FMS is a clinician interpretation of self-report performance measure and direct observation is not involved. There is an assumption that administration of the FMS through child or parent report will accurately reflect the true performance of the child in his or her usual environments, however this has not been established. Other measures that rely on child or parent report include the FAQ (Novacheck et al 2000), the ASK (Young et al 1995) and the two health related quality of life tools, the CHQ (Landgraf et al 1996) and PODCI (Daltroy et al 1998). These tools use self-report to obtain the perspectives of the child and/or parent while assuming that what is reported reflects what is really happening. Other scales such as the PEDI (Haley et al 1992) and WeeFIM (Msall et al 1994) use a combination of observation and parent or clinician report. Tools that use direct observation only, such as the GMFM (Russell et al 1989), usually measure capability rather than performance and the observation occurs in the clinical setting.

Testing of construct validity of the FMS is required to ensure that it is measuring what is actually occurring in the different environments of home, school and community. It is also necessary to establish that it is based on the different assistive devices children use in those different settings. One way to determine if the scale is measuring what it is intended to measure is to compare the self-reported FMS rating with direct observation of the mobility methods children use in their own environments. Because validation is an ongoing process, one study alone cannot empirically determine construct validity (Streiner and Norman 2003). Other studies within this thesis, reported in Chapters Six and Eight, also contribute to construct validity. These results will be synthesized in the grand discussion. Although a previous study attempted to examine construct validity of the FMS, the authors were examining change in mobility status after surgery rather than testing predetermined hypotheses about theoretical constructs (Graham et al 2004). Results from the examination of concurrent validity in the Graham study add to the evidence of convergence and divergence of the FMS; however the concepts that form the basis of the FMS have not been adequately examined.
The research question of the current study was “does the information obtained from the FMS via self-report of the assistance required in the different settings of the home, school and community accurately reflect the mobility status of children with CP”.

The main aim was to examine one aspect of construct validity of the FMS in a group of children with CP by comparing FMS ratings performed by usual method with direct observation at home and school. This was to determine if the FMS reflects the actual mobility status reported. A secondary aim was to describe the different mobility methods the children used as a group and individually.

The a priori hypothesis was “the mobility status of children with CP observed within the environmental settings of home, school and the community agrees with the self-report rating of the FMS with agreement of at least 0.6 using kappa statistics”. The value of 0.6 was chosen based on guidelines that kappa statistics of 0.61 and higher represent substantial to almost perfect agreement (Landis and Koch 1977).

The secondary hypothesis was that “children with CP use different types of mobility methods in the different environmental settings of home, school and community”.

7.2. Method

7.2.1. Participants
The inclusion criteria for children involved in this study were those:
   i. with CP aged between 4-18 years
   ii. classified as GMFCS levels I-IV
   iii. who were clients of the Royal Children’s Hospital, Victoria, Australia
   iv. who attended Nepean Special School, Victoria, Australia

The first and second inclusion criteria are in accordance with the population the FMS was devised for. This has been justified in Chapter 4 (section 4.2.1). The
children were required to be clients of The Royal Childrens Hospital, Victoria because this was where participants were recruited from. This study involved observing children at home and at school. One special school, Nepean School, was targeted for recruitment of participants. This was to ensure it was feasible to observe enough children with CP at school and at home within the time constraints of the thesis. For the one school utilized it was necessary that the school have a large enough group of children with CP attending it to guarantee adequate recruitment of participants. Using a special school assured this. Of the special schools attended by children with CP from The Royal Childrens Hospital that were appropriate, the staff at Nepean School were the first to consent for the study to take place there. Nepean School is a special school in Victoria, Australia. The children who attend there have significant cognitive or behavioural impairments which preclude them from being integrated into mainstream schools. Many of them also have significant physical impairments. The population of children at the school tends to include more children classified as GMFCS level IV than the usual CP population.

Excluded were children:

i. younger than 4 years and older than 18 years
ii. classified as GMFCS level V
iii. with a diagnosis other than CP
iv. whose parents were not able to understand the information explaining the study or instructions due to language, cognitive or other difficulties.

Children younger than 4 years or older than 18 years and with a diagnosis other than CP were excluded because the FMS was developed for children with CP aged 4-18 years. Children classified as GMFCS level V were excluded also because the scale is not appropriate for use with them. This has been explained more comprehensively in Chapter 4 (section 4.2.1) and in Chapter 5 (section 5.2.1.2). Children whose parents were not able to understand due to language or cognitive difficulties were excluded if they could not fully comprehend what was required.
This investigation was a feasibility study to examine one aspect of construct validity. The study aimed to obtain some preliminary data on construct validity from which further studies could be developed. An ideal sample size would have been more than 72. This is based on an equation of $2c^2$ (Norman and Streiner 2000). Using this equation $c$ is the number of categories within a scale. There are 6 categories in the FMS, therefore $2 \times 6^2 = 72$. Because each child was required to be observed at home and school by a physiotherapist a sample size of 20 was aimed for. This smaller sample size allowed adequate time for recruiting the children through the hospital who attended the school and for the direct observations to occur. It was unrealistic and impractical to attempt to recruit more than this given the amount of time it would take to complete all assessments within the time constraints.

### 7.2.2. Procedures

The study was accepted by the Ethics in Human Research Committee of the Royal Children’s Hospital (reference number 25118A) and the University of Melbourne Human Ethics Research Committee (number 060211). Approval for the study was also obtained from the Department of Education and Training (Victoria), the Regional Director for the Southern Metropolitan Region and the school principal.

#### 7.2.2.1 Recruitment

Recruitment of participants was via a sample of convenience. A sample of convenience was used so that all children who attended Nepean School and were clients of the Royal Children’s Hospital had a chance to participate. It was also necessary to use this method to ensure an adequate sample size was obtained as only one recruitment centre was utilized. This school was selected because many of the children who attended there also attended the CP clinics or gait laboratory of the hospital and the children represented a range of severity of motor impairment as classified by GMFCS levels. The primary investigator obtained a list of eligible children from the physiotherapist employed by the school (JH). The parents of these children were sent a letter in the mail explaining the study and inviting them to participate. A stamped, self-addressed envelope was included in that letter for the parents to return their consent to
participate. These returned forms designated the children to be included in the sample. All children whose parents agreed for them to be involved in the study had signed consent forms.

7.2.2.2 Testing procedure

There were three stages to data collection:

i. obtaining an FMS rating over the telephone as per usual administration of the scale by an independent experienced physiotherapist

ii. direct observation of the child at school by the principal investigator

iii. direct observation of the child at home and in outside the home by one of two Nepean school physiotherapists.

The FMS ratings obtained over the telephone for each child were administered by one person (AF). This person was a physiotherapist working within the gait laboratory of The Royal Childrens Hospital who was experienced in the use of the FMS. She was independent of the study and the school and did not have knowledge of the mobility status of the children. This ensured impartiality. Ratings for all children were performed over the telephone with a parent or guardian and recorded on a form. The person administering and recording these ratings had no further involvement in the study.

Direct observation of the children at school was executed by the primary investigator (AH). She attended the school for one entire school day and directly observed the children moving around and recorded methods of mobility used. The distances and settings observed for each child were:

i. from the bus to the classroom,

ii. within the classroom,

iii. from classroom to classroom and

iv. around the playground.

These four activities were selected because they represent the different settings and situations at school children might be required move around in order to participate within the school environment. A checklist on a recording sheet was
composed for the observer to record the method of mobility the children used for each activity (see Appendix F). The possible different methods included:

i. wheelchair

ii. walker

iii. crutches

iv. sticks

v. holds hands or walls

vi. independent walking (uses rails for stairs)

vii. independent walking (no rails for stairs)

viii. is carried

ix. crawls

These methods were chosen to reflect the categories on the FMS and to represent the typical range of methods children with CP might use to move from one place to another. Also recorded on the checklist were details including date of birth and age, GMFCS level, type of assistive devices used and approximate dimensions of the relevant areas the children moved around. While attending the school the observer attempted to quietly observe and record the children unobtrusively without disrupting or changing their “usual” routine. It was important to record what the children usually did rather than what they could do, so that a record of performance rather than capability was obtained to reflect the purpose of the FMS.

The home observations were performed by the two physiotherapists from Nepean School (JH and MH). These observers were employed because the parents were familiar with them and were more likely to accept them entering their homes. The children themselves would also be more relaxed with their school physiotherapist visiting them at home. Similar to the school observations, the aim of the home visit was to observe how the children chose to move around rather than best effort. It is not uncommon for school physiotherapists to perform home visits and it was felt that it was more likely to obtain a true performance observation utilising the school physiotherapist rather than with an unfamiliar clinician.
Each child was visited at home by one of the physiotherapists who observed the children moving around the home environment and outside the home. A similar checklist with the same options of mobility methods was devised for the home visits for the observers to record the methods used (see Appendix G). The areas within the house recorded were

i. from room to room
ii. within one room
iii. outside the home

Once the child had been observed inside the home, the physiotherapist asked the child to venture outside and travel more than 150 metres where possible and practical, for example, down to the shop or around the block. This information was aimed at providing data to compare with the 500 metre FMS score. All attempts were made to ensure the children were moving around as they would usually choose to, rather than giving their best effort because they were being watched. The observers attempted to take the focus off how they moved, explaining it was the place and distance that were important, not which method they chose.

Each child had one rating performed over the telephone and was observed once at school and once at home. For all three stages of data collection, the various observers and raters were independent of previous information collected by others and did not have access to each others information observed and recorded.

The information obtained from the school and the home observations was used to generate an “observed” FMS score. To generate this, the “within one room” category for the home observation was used for the 5 metre distance and the “outside the home” category was used for the 500 metre distance. At school the “from class to class” category was used for the 50 metre distance. Within the class was not used from the school observation because this is a similar distance to within the home. The playground was not used for the 50 metre distance because those distances were often greater than 50 metres. All methods used were recorded to provide details of the variety and number of methods used in
each environment and the number of different methods used by each child. The “observed FMS” score was compared to the “telephone FMS” score for the data analysis.

7.2.3. Data Analysis

Descriptive statistics of participant characteristics were obtained using means and standard deviations for continuous variables or frequency and percentages for categorical variables. Descriptive statistics were used to illustrate the number and types of various methods of mobility the children used. The number and variety of methods for each child was recorded as well as for the group as a whole.

Kappa statistics with 95% confidence intervals were used to analyse agreement between the “telephone FMS” obtained by usual administration method and the “observed FMS” generated from the observations. The data obtained was categorical, therefore the kappa statistic to examine agreement is the most appropriate method of analysis for agreement (Tooth and Ottenbacher 2004). Data analysis involved the use of unweighted kappa statistics based on the justification in Chapter Five (section 5.2.3) that disagreement of 1 level on the FMS is clinically significant and should be factored into the analysis. The basic or unweighted Kappa considers all disagreements in ratings as equal in seriousness (Tooth and Ottenbacher 2004). Quadratic weighted kappas were also calculated as a comparison.

The data were stored and organized using the EpiData for Windows program (http://www.epidata.dk/). All analyses were performed using Stata [StataCorp 2005 Stata statistical software Release 9.0. College Station; TX. Stata Press.].
Chapter 7 – Construct Validity

7.3. Results

7.3.1. Descriptive results

7.3.1.1 Participant characteristics

Twenty seven children were eligible to enter the study based on the inclusion and exclusion criteria. The parents or guardians of 18 of those 27 children agreed for their child to be part of the study. There was no reply from the parents or guardians of the remaining 9 children. The sample of 18 included 7 females and 11 males. The age range was 8.5 to 17 years (mean 12.7 and SD 2.63). There were 5 children classified as GMFCS level II, 4 as level III and 9 as level IV. Table 7.1 summarises the participant characteristics. The mode of FMS scores based on the telephone ratings was level 1 for 5 metres with 6 children using a wheelchair around the home, level 1 for 50 metres with 8 children using a wheelchair at school and 1 for 500 metres with 15 children using a wheelchair in the community.

Table 7.1 Participant characteristics

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Total number</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>Total number</td>
<td>18</td>
<td>18</td>
</tr>
<tr>
<td>Age (yrs)</td>
<td>12.7 (2.63), 8.5-17</td>
<td>range 8.5-17 years</td>
</tr>
<tr>
<td>Gender, n (%)</td>
<td>male 11 (61%), female 7 (39%)</td>
<td>GMFCS level II</td>
</tr>
<tr>
<td>GMFCS, n (%)</td>
<td>II 5 (28%), III 4 (22%), IV 9 (50%)</td>
<td>GMFCS level III</td>
</tr>
</tbody>
</table>

7.3.1.2 Timing of ratings and observations

The time between FMS ratings performed over the telephone and direct observation at home and school varied. The mean time between the home observation and the FMS rating was 10.2 days (SD 5.44) with a range of 0-17 days. Nine children (50%) had the rating and observation performed within 10 days apart. There was an interval of 11-17 days for the remaining 9 children. The mean time between the school observation and the FMS rating was 20.9 days (SD 6.36) with a range of 14-40 days. Thirteen (72%) of the children had
the rating and school observation performed within 3 weeks apart. There was an interval of up to 40 days for the remaining 5 children.

7.3.1.3 Results from direct observation

The direct observations showed differences in the number and range of mobility methods used within the home, school and community. Within the home 12 children (67%) used one method, 5 children (28%) used two methods and 1 child used three different methods. Within the school 5 children (28%) used one method, 10 children (56%) used two methods and 3 children (16%) used three methods. Outside the home, for longer distances, all children were observed to use only one method. In summary, the majority of children used one method of mobility within the home, two methods at school and one method for longer distances outside the home. Of the total number of 18 participants, 7 used more than one method at home and 13 used more than one method at school.

There were differences in the different types of mobility methods observed within each environment. There were six different methods used within homes, seven within school and four outside the home. None of the children used single point sticks or were carried. Figure 7.1 summarises the frequency of different types of mobility methods used in each setting.

![Figure 7.1 Frequency of mobility methods observed](image-url)
The percentages quoted below are the percentage of total number of different methods for each setting, not the percentage of children, because some children used more than one method. Within the home the most used method was independent walking not requiring rails (28%), followed by crawling (20%) and wheelchair (20%). Other methods included using a walker (12%), holding hands (16%) and independent walking requiring rails for stairs (4%). Within the school the method most used was wheelchair (44%) followed by walkers (21%). Other methods included independent using rails for stairs (11%), holding hands or walls (9%) and to a much lesser extent crawling, crutches and independent with no rails. Outside the home for longer distances the method most used was wheelchair (72%). Independent walking without rails, holding hands and crutches were used by a very small number of children.

There were some subtle differences in number of mobility methods within each environmental setting according to the GMFCS level of the children. This is summarized in table 7.2.

i. **GMFCS II.** Children in GMFCS level II used mainly one method in the home (4 of 5 children) and two methods in the school (3 of 5 children). The home method was predominantly independent walking and the school methods were independent walking or walking holding onto walls or hands.

ii. **GMFCS III.** Children in GMFCS level III used mainly one method at home (3 of 4 children) and three methods at school (3 of 4 children). The methods used at home were independent walking, walker, crawling and wheelchair. The methods used at school were walker, crutches, wheelchair, and holding hands or walls.

iii. **GMFCS IV.** Children in GMFCS level IV used between one and two methods at home (5 used one and 4 used two) and mainly two methods at school (6 of 9 children). The methods used at home were crawling, walker, wheelchair and holding hands or walls. The methods at school were walker and wheelchair.
Chapter 7 – Construct Validity

The school setting for children in GMFCS levels III and IV had the most number of different methods used. All children classified as GMFCS level III used more than one method at school. Children in level III were the only ones who used three different methods. The most consistent was mobility outside the home with children in all GMFCS levels using only one method of mobility, including wheelchair, holding hands, crutches, and independent walking. Both the numbers of children in the total sample and for each GMFCS level were small, particularly levels II and III. The results are of interest but are not conclusive.

Table 7.2 Children (n) in each GMFCS level who used 1, 2 or 3 different mobility methods in each setting

<table>
<thead>
<tr>
<th>GMFCS level</th>
<th>Home</th>
<th>School</th>
<th>Community</th>
</tr>
</thead>
<tbody>
<tr>
<td>II (n=5)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>1 method used</td>
<td>4</td>
<td>2</td>
<td>5</td>
</tr>
<tr>
<td>2 methods used</td>
<td>1</td>
<td>3</td>
<td>0</td>
</tr>
<tr>
<td>3 methods used</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>III (n=4)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>1 method used</td>
<td>3</td>
<td>0</td>
<td>4</td>
</tr>
<tr>
<td>2 methods used</td>
<td>0</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>3 methods used</td>
<td>1</td>
<td>3</td>
<td>0</td>
</tr>
<tr>
<td>IV (n=9)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>1 method used</td>
<td>5</td>
<td>3</td>
<td>9</td>
</tr>
<tr>
<td>2 methods used</td>
<td>4</td>
<td>6</td>
<td>0</td>
</tr>
<tr>
<td>3 methods used</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
</tbody>
</table>

The variability in methods observed in the different settings is best described using some case examples.

i. Example 1. A boy “JD” aged 12 years 3 months who was classified as GMFCS level III was observed to walk around his home independently. At school he was observed walking independently around the classroom, using crutches from class to class and a wheelchair in the playground and from the bus to the classroom. Consequently, at school he used three different methods of mobility.

ii. Example 2. A girl “CM” aged 9 years 5 months classified as GMFCS level III was observed to both crawl and use a Kaye walker at home. At school she used a walker within the classroom and between classes and a
wheelchair around the playground. She had two different methods at home and at school.

iii. Example 3. A girl “KS” aged 15 years and classified as GMFCS level IV walked with adult assistance and crawled at home. At school she used a wheelchair. She therefore used two methods at home and one at school.

Some children were more consistent and used a smaller number of methods for mobility. For example, one boy aged 14 years 4 months and classified as GMFCS level II was observed to walk independently in all areas at home and school, including longer distances. He used only one method in total.

7.3.2. Agreement results

Unweighted and weighted kappa statistics were calculated for the agreement between the FMS rating performed over the telephone (“telephone” FMS) and the “observed” FMS score for each distance. Table 7.3 displays the kappas obtained with confidence intervals and percentage agreement for each FMS distance.

The unweighted kappas were:

i. 0.27 for 5 metres
ii. 0.45 for 50 metres
iii. 0.29 for 500 metres

The weighted kappas using a quadratic weighting system were:

i. 0.71 for 5 metres
ii. 0.76 for 50 metres
iii. 0.74 for 500 metres

Table 7.3 Agreement between "telephone" FMS and "observed" FMS

<table>
<thead>
<tr>
<th></th>
<th>FMS 5m</th>
<th>FMS 50m</th>
<th>FMS 500m</th>
</tr>
</thead>
<tbody>
<tr>
<td>Unweighted</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>k (CI)</td>
<td>0.27 (0.08, 0.46)</td>
<td>0.45 (0.20, 0.70)</td>
<td>0.29 (0.05, 0.53)</td>
</tr>
<tr>
<td>% agreement</td>
<td>39%</td>
<td>61%</td>
<td>72%</td>
</tr>
<tr>
<td>Weighted</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>k (CI)</td>
<td>0.71 (0.41, 1.0)</td>
<td>0.76 (0.31, 1.00*)</td>
<td>0.74 (0.33, 1.00*)</td>
</tr>
<tr>
<td>% agreement</td>
<td>45%</td>
<td>94%</td>
<td>95%</td>
</tr>
</tbody>
</table>

CI = confidence interval  *CI’s truncated to 1.00 at upper end
Using the guidelines for the range of kappas describing the strength of the agreement stated in Chapter 5 (section 5.3.2) (Landis and Koch 1977), the agreement for the unweighted kappas for 5 and 500 metres is fair and for 50 metres is moderate. The weighted kappas show substantial agreement for all three distances. The large confidence intervals around these kappa statistics preclude any conclusion to be drawn about higher or lower agreement between the three distances.

Due to the small numbers in the study, further examination of agreement according to different GMFCS levels, age groups or number of mobility methods used was unable to be performed. The numbers in each group are too small for kappa statistics to be calculated.

7.4. Discussion

7.4.1. Variety of mobility methods observed

The children in this investigation displayed a range of methods of mobility used within the different environmental settings of the home and school. There was variety within the group as a whole with regard to the type and number of methods used and variety within the children. Many of the children used more than one method in either setting and used different methods at home than at school.

This finding is supported by other reports on mobility methods across environmental settings (Palisano et al 2003, Tieman et al 2004). Both of these studies used data from a longitudinal study of children with CP from Ontario and asked parents to report “usual” mobility methods their children used in the home, school and outdoor/community setting. The Palisano study described the mobility methods at one point in time in a random stratified sample of 636 children with CP, GMFCS levels I-V, aged 2-12 years (Palisano et al 2003). Children in GMFCS levels II-IV demonstrated a variety of methods in all settings. There was less variability for those in level I, where children mostly walked independently, and level V, where they were pushed or carried. Children
were more independent at home than school and used more dependent methods in the community compared to school.

Following on from this study, 62 children aged 6-14 years, classified as GMFCS levels II-IV, were examined more closely because they had showed the most variability (Tieman et al 2004). Rather than examine mobility at one point in time, the authors followed the children for 3-4 years to examine change over three time points. At home the most common methods were floor mobility or independent walking. At school the most common method was walking with a walking aid or wheelchair. For the community a wheelchair was the most common method. These results are very similar to the most common methods demonstrated in the current investigation. In the Tieman study, the children used methods requiring more gross motor function at the second time point compared to the first. This suggests they had progressed with their skills. The difference between the second and third time point was not significant. Both the Tieman and Palisano studies provide important information regarding mobility methods using large numbers of children. Both asked parents to choose only one method their child used in each setting and neither study differentiated the type of walking aid used. The current investigation reported all methods the children used in each setting and described all types of walking aids.

7.4.2. Agreement between self-report FMS and direct observation

The agreement between the FMS rating performed over the telephone and that observed at home and at school was lower than expected using unweighted kappa statistics. Agreement was substantial when quadratic weighted kappas were calculated; however unweighted kappas are more clinically meaningful, as discussed in Chapter Five. This is because even one level of disagreement can be considered a clinically meaningful difference for a scale such as the FMS that has only six levels.

There are a number of factors that could have contributed to this agreement being fair to moderate. Some of these including the variability of mobility methods, the issue of self-report and the issue of measuring capability or performance; could suggest that the disagreement is real. Other factors are
methodological issues that suggest agreement could have been improved with enhanced methodology.

One factor that is likely to have influenced the agreement is the number and variety of mobility methods observed in the children. Many children were observed to use more than one method at home and at school. It is possible that this variety created difficulties for the clinicians administering the FMS and for the child or parent reporting. It may have been challenging to choose which FMS rating to assign for each of the three distances when more than one method could have applied. It is likely that the 50 metre school rating was problematic because some children used up to three different methods. Not only did the parent have to make a choice of methods, it is also possible that they were not fully informed of their child’s mobility at school. Most of the children in this study travelled to and from school on the bus without the parents attending the school. The parents may not have given an accurate report of school mobility. Another factor related to the variety of methods used that could have affected agreement was that the children were observed at home and school at one point in time only. On a different day the children may have used other methods.

Another factor that potentially influenced the agreement was the self-report nature of the FMS. Like other self-report scales used in children with CP, such as the FAQ (Novacheck et al 2000), ASK (Young et al 2000), CHQ (Landgraf et al 1996) and PODCI (Daltroy et al 1998), it is assumed that self-report reflects what is truly happening. Self-report may not be the most reliable method of obtaining information regarding activity for these complex children. The agreement in the current study may in fact reflect the true difference between self-report and direct observation.

Studies have examined the reliability of self-report measures. The reliability of family report for the GMFCS was examined in 97 children aged 6-11 years by comparing GMFCS classifications made by clinicians with a questionnaire completed by the family (Morris et al 2004). ICC’s of 0.92-0.97 were found suggesting family report is reliable. It is assumed in the Morris study that the GMFCS classifications made by the clinicians were through observation in the
clinical setting however it is not completely clear. During the development of the ASK, 28 children aged 5-15 years completed one version two weeks apart to assess the reliability of child-report (Young et al 1995). Retest reliability was high with ICC’s of 0.97 and child report was also highly concordant with parent report scores with ICC’s of 0.96. The Young study supports reliability of child report but does not investigate the agreement between child report and direct observation. Studies have examined the reliability of other self report measures, such as the FAQ (Novacheck et al 2000) and the PODCI (Daltroy et al 1998). These studies on the reliability are important; however there is a deficiency in the literature of examining the agreement between self report and direct observation, that is, the validity of the self-reporting method. Without evidence of this agreement it is uncertain whether these measures are reporting what they intend to report.

The FMS is intended to measure performance rather than capability. Despite all efforts by clinicians asking the correct questions to obtain a performance rating, there is the possibility that the parents reported what their child “can do” rather than what they “do do”. Studies have shown differences between capability and performance of activity in children (Young et al 1996, Tieman et al 2004). In the Young study, both the capability and performance versions of the ASK was administered to 28 children with physical disabilities aged 5-15 years. Capability consistently exceeded performance with an 18% difference for total summary scores. In the Tieman study, differences in the performance and capability of mobility across environmental settings was examined in a random stratified sample of 307 children with CP aged 6-12 years. Capability was based on the highest item passed on the GMFM from three items based on mobility. Performance was based on parent questionnaire. Children with similar capabilities demonstrated differences in performance across settings. These studies demonstrate that capability of activities, such as mobility is not the same as performance during daily activities and routines. For the current study, it was possible that some of the parents reported capability on the FMS telephone rating. As a result, agreement would be lower because the observational ratings of the children recorded performance.
Differences in agreement between what is observed and what is reported is an important issue that impacts on how we measure activity in children with CP. If measuring performance reflects function relevant to these children and is more meaningful, then it is likely that self-report is the method of information collection. Even though observations in daily life are more valid than other methods of observation, such as interviews (Nunnally 1978), it is impractical to observe every child seen clinically within all their environmental settings. Observing children within the formal clinical setting is not reflective of their usual everyday settings and therefore it is difficult to measure typical performance (Long 1992). Self-reporting therefore remains the most practical and feasible method of measuring performance.

The nature of the sample recruited for this study may have influenced the lower than expected agreement. The children who attended the school were complex with intellectual disabilities as well as physical disabilities. Many mobility options were dictated by safety related to behavioural and cognitive issues rather than physical factors alone. A sample more representative of the CP population would have more consistent mobility options and possibly be easier to rate. Other methodological factors that possibly influenced the agreement include the small sample size and the time that elapsed between the ratings and observations. There were only 18 children included in the sample due to the feasibility of recruitment, as stated in the methods section. A larger sample size may have resulted in improved agreement with increased power and smaller confidence intervals. The time between the FMS ratings performed over the telephone and the direct observations ranged from 0-40 days. There is a possibility that change occurred in the mobility status of some children that would result in lower agreement between the two.

Also potentially influencing the agreement was the process of creating the “observed” FMS. For the home and school observations there was more than one category where mobility method was recorded that reflected different distances. For example, at school, mobility within the class, from class to class and in the playground was recorded. The problem of which category to choose for the “observed” score was minimized by stating clearly that for the school
rating the principal observer would use the “from classroom to classroom” category to rate the 50 metre distance. Agreement between this “observed” score and what the parent reported may have been different. A similar predicament was present for the home distance where categories of observation included within a room or between rooms. The “observed” FMS used the within the room which may not have concurred with the parental report. Observations of what method the child used outside the home was used to create the 500 metre “observed” FMS. This cannot be truly considered as a community observation because a usual community setting would involve other factors such as crowds, greater distances, time-saving factors and unfamiliar environments.

The investigation reported in this chapter has highlighted some pertinent issues regarding the categories of the FMS. The FMS focuses on independent mobility. The types of walkers commonly used for independent mobility are posterior walkers as illustrated in Figure 7.2. The FMS does not include those walkers requiring adult assistance to place the child into that support the child at the trunk and pelvis as illustrated in Figure 7.3. During scale development these types of walkers were considered to provide too much support for the child to be considered independent.
Chapter 7 – Construct Validity

At the school visit it was noted by the principal investigator that some of the children classified as GMFCS level IV could be independent in these more supportive walkers once they were placed there and strapped in by an adult. Once they were secured they were able to move around the playground at lunchtime on their own, often covering distances greater than 200 metres. This issue will be elaborated on further in the grand discussion.

There are a number of factors that might influence or determine why the same child uses different mobility methods in different environmental settings, or how the environmental setting determines what method is used. This includes the amount of assistance required, safety and liability issues, therapy goals, convenience and child or parent preferences.

Within the home there are shorter distances for the child to travel to get from one place to another and furniture and walls that provide stability or a sense of security. The child who normally uses assistive devices at school might walk on their own at home because they feel safer. Many children choose to crawl at home because they can get from one place to another quickly and independently without having to pull themselves up into standing and onto an assistive device. This provides them with more independence and gives the parents more time within the busy schedule of family life. The distances and logistics of a school setting tend to preclude children from crawling there.

Safety within the school environment and liability issues also potentially influence mobility methods. Many schools are concerned that children might harm themselves moving around within the school setting, particularly with other children rushing around. This is less of an issue in special schools where the environment and support is tailored for children with physical and cognitive impairments. It is possible that children use wheelchairs more at school to ensure safety and limit liability concerns. As an example, one boy in this study aged 17 years and classified as GMFCS level II was observed to walk independently at home. Significant behavioural issues placed him at risk of self-harm at school. As a result, he used a wheelchair for the majority of the time at school, except for within the classroom. Therapy goals at school might also
influence mobility methods used. In this study, many of the children walked from class to class with the supervision of a teacher, physiotherapist or aide as part of their physiotherapy program. Although this was observed to occur regularly, many of the children were unable to do this without supervision or assistance and this would not constitute their “usual” performance.

Convenience and time saving factors might influence mobility methods chosen, particularly at home and in the community. Many parents reported that they would use a wheelchair at the supermarket or shopping centre to save time or because the child tired easily with longer distances. Some families reported that they leave the walker at school because it does not fit easily inside the house. Crawling or wheelchair would then be the methods used at home.

7.4.3. Limitations and recommendations

There are a number of limitations of this study that need to be addressed. The sample size was comparatively small, as stated earlier, and only one school was used for recruiting the participants. Due to the nature of this school there was a bias in the sample of children classified as GMFCS level IV. This is evidenced by the modal frequency of FMS level 1 for each distance. This limits the generalisability of the results as it does not adequately represent the population of children with CP.

The observer in the school setting was the principal investigator who knew some of the children through clinics at The Royal Childrens Hospital. While she was not fully aware of the mobility status of each child, she was not completely impartial. The observers used in the home settings were well known to the children. The advantages and reasons for this were stated in the methods section. The disadvantages of this were that they had some prior knowledge of what the children were able to do which this may have influenced their observations. Having the physiotherapist observe them at home may have influenced how the child chose to mobilize. There was the possibility that they did their “best” performance rather than “usual” in order to impress the physiotherapist. It is very difficult to minimize the effect of the children being watched in a close environment such as the home. This was less of a problem at
school because the observer could blend in more and not be so obvious because of more activity going on around the children.

The range of timing between the “telephone” FMS and direct observation could have allowed for change to occur in the mobility status of some of the children. This has the potential to influence the agreement negatively and provide a source of variation. It would be preferable to have a smaller time period between them to limit the possibility of such change occurring. An interval of 2-14 days is suggested as usual (Streiner and Norman 2003).

This was a feasibility study with the aim to provide information for further research. Further examination of construct validity exploring the same question is required using larger numbers of children representing a more even spread of GMFCS levels recruited from a larger and more varied number of schools. Within this it would be preferable to observe the children directly in the community with completely objective observers and with less time lapsing between ratings and observations.

The study highlighted some issues around the administration of the FMS and how to record other forms of mobility not currently covered by it. There is the potential to modify the scale to improve its’ clinical utility. This will be discussed further in Chapter Nine.

7.5. Conclusion

Fair to moderate agreement was found for each distance of the FMS when comparing direct observation with FMS ratings in a group of children attending a special school. Descriptive statistics from direct observation showed there was variability in the number and type of mobility method used within different environmental settings for the whole group and for individual children. The diversity of mobility methods used, issues relating to self report measures, and methodological limitations of this study are potential explanations for the differences. Further research with larger and more representative samples is required comparing self-report with direct observation for the FMS and similar measures of activity limitation used in children with CP.
CHAPTER EIGHT: CONCURRENT AND DISCRIMINATIVE VALIDITY OF THE FUNCTIONAL MOBILITY SCALE

8.1. Introduction

This chapter explores the concurrent and discriminative validity of the FMS. This further validation of the scale was considered to be important to ensure that it is measuring activity limitation and can distinguish between groups of children with CP based on severity of activity limitation.

Concurrent validity is a form of criterion validity and is sometimes referred to as convergent validity (Streiner and Norman 2003). It involves correlating a new scale with a criterion measure, ideally a ‘gold standard’ which has been used and accepted in the field (Nunnally 1978, Streiner and Norman 2003). The criterion measure is usually another measure of the disorder under study. Examination of concurrent validity of the FMS is required to ensure that it correlates well with other tools that also measure mobility. The correlation aims to show a strong relationship between the FMS and a criterion measure, yet not perfect, otherwise the two measures could be considered too similar.

The other form of criterion validity is predictive validity, which is examined primarily in diagnostic or predictive tests (Streiner and Norman 2003). It allows measurement done in the present to infer something about the future for predicting outcome (Baxter 2005). Predictive validity is important to examine in measurement tools that are designed to discriminate abnormal motor development from normal development such as the AIMS (Piper and Darrah 1994) and the Peabody Developmental Motor Scales (Folio and Fewell 1983). The FMS is an evaluative measure. It has been developed to assess change over time rather than to diagnose or predict. Because of this the examination of predictive validity is not a requirement; however the examination of concurrent validity is important.

The FMS measures functional mobility in children with CP, for which there is no gold standard. There are a number of tools that have mobility sections within
a larger inventory such as the PEDI (Haley et al 1992), the WeeFIM (Msall et al 1994) and the ASK (Young et al 1995). The walking scale of Gillette FAQ (Novacheck et al 2000) focuses on mobility and is also an ordinal scale. It describes the typical walking abilities of children with a consideration of the environment, yet does not consider the assistive devices used. The FMS considers different assistive devices the children use. It also describes mobility within each environmental setting of the home, school and community based on the assistance required. Consequently, the FAQ reports one score or category whereas the FMS reports three. The FAQ is a parent report measure, whereas the FMS is a clinician interpretation of child or parent report. In the absence of a “gold standard” the FAQ was chosen to be the criterion measure to correlate with the FMS for examining concurrent validity. Despite some differences between the two measures, the FAQ is the only tool that also measures mobility in isolation in children with CP.

The GMFCS (Palisano et al 1997) is a classification system that is now used internationally to describe the gross motor functional ability of children with CP. Children are classified according to one of five ordered levels of severity of motor impairment. Correlation of the FMS with the GMFCS was also considered important. Although it is not an outcome measure, it does classify children according to activity limitation and a strong correlation with it would further support the concurrent validity of the FMS.

Discriminative validity involves assessing the ability of a scale to distinguish between two groups, one group that has the trait or behaviour in question and the other that does not (Streiner and Norman 2003). Discriminatory analysis is employed when groups of persons are defined a priori and the purpose of the analysis is to distinguish the groups from one another on the basis of their score profiles (Nunnally 1978). It is sometimes called construct validation by extreme groups (Streiner and Norman 2003). The FMS corresponds to the activity and participation domain of the ICF. It is used clinically in conjunction with classification systems such as the GMFCS (Palisano et al 1997) to classify and measure the activity limitations of children with CP. The validity of the FMS would be further supported by determining if it was able to discriminate severity
of CP as classified by the GMFCS. This would provide evidence that the scale was able to distinguish between groups based on their level of activity limitation.

A preliminary study of the FMS examined aspects of concurrent and discriminative validity in 310 children with CP (Graham et al 2004). Concurrent validity was examined by correlating the FMS with the CHQ, PODCI, Uptimer, energy expenditure and Rancho scale. Spearman rank correlations showed strong correlation of the FMS with all measures ($r>0.71$) except for energy expenditure ($r=0.51-0.55$). A limitation of the Graham study was that the FAQ was not one of the measures involved in the examination of concurrent validity. The Rancho scale (Hoffer et al 1973), sometimes called the Hoffer scale, was developed for children with myelomeningocele. It rates ambulation level on a four point scale; community, household, non-functional ambulators or non-ambulators. Although it has been used in children with CP the scale has not been validated in CP. Uptime (Eldridge et al 2003) records the amount of time children spend in the upright position each day. It uses metric measurement and is not a categorical scale like the FMS. The CHQ and PODCI are both larger inventories that include a section on physical function. The FAQ is the tool most similar to the FMS and examination of concurrent validity with it as a criterion measure is required.

In the Graham (2004) study, discriminative validity was examined by determining the ability of the FMS to differentiate between different walking abilities of the children based on the Rancho grading system (Hoffer et al 1973). The authors stated that the FMS was able to differentiate children with varying degrees of walking ability. However the results were descriptive with no formal statistical analysis. Further examination of concurrent validity with the FAQ and discriminative validity of the FMS is therefore required.
The research questions for this study were:

i. “What is the concurrent validity of the FMS with the FAQ and the GMFCS?”

ii. “Is the FMS able to discriminate severity of activity limitation in children with CP as classified by the GMFCS?”

The aims of the study were to determine:

i. the concurrent validity of the FMS by correlating it with the walking scale of the Gillette FAQ and the GMFCS

ii. if the FMS can discriminate severity of activity limitation of children with CP as classified by the GMFCS

The hypotheses were:

i. The FMS will have a strong positive relationship with the FAQ using correlation coefficients in a sample of children with CP at one point in time

ii. The FMS will have a strong negative relationship with the GMFCS using correlation coefficients in a sample of children with CP at one point in time. This relationship will be negative due to the reverse scaling of the FMS compared to the GMFCS.

iii. The FMS will discriminate severity of motor impairment in children with CP as described by the GMFCS for children classified as GMFCS levels I-IV.

8.2. Method

8.2.1. Participants

Participants involved in this study were recruited for the inter-rater reliability (Chapter Five) and responsiveness to change (Chapter Six) studies. These were sample 2 and sample 3 as illustrated in Chapter One (section 1.3). The current study utilised the data collected in those studies for additional analyses. There were a number of children who had participated in both studies. For these, only the data obtained from the reliability study was used in the current study so that one data set per child was utilised.
The inclusion criteria for the participants were children:
   i. with CP aged 2-18 years,
   ii. classified as GMFCS levels I-IV,
   iii. who attended the CP clinics or gait laboratory of the Royal Children’s Hospital, Melbourne, Australia.

Excluded were children:
   i. with a diagnosis other than CP,
   ii. younger than 2 years and older than 18 years,
   iii. classified as GMFCS level V.

The inclusion and exclusion criteria were justified in Chapter Five (section 5.2.1.2) and Chapter Six (section 6.2.1). The aim of the current study was to determine the concurrent and discriminative validity of the FMS for the sample of children at one point in time.

The sample size was determined by the numbers in each of the combined studies. The sample size of 118 used in the reliability study was a large sample justified on the basis of obtaining a high level of agreement between raters with 95% confidence intervals. The sample size of 84 recruited in the responsiveness study was based on obtaining adequate numbers to ensure a change following interventions would be detected and on clinical availability of the children. A combination of these two samples provided a large number of participants for examining concurrent and discriminative validity.

8.2.2. Procedures
The study was approved by the Ethics in Human Research Committee of the Royal Children’s Hospital (reference numbers 25118A and 25119A) and The University of Melbourne Human Ethics Research Committee (reference numbers 060211 and 060212) as stated in Chapters Five and Six. Written consent was obtained from each participating child’s parent or legal guardian prior to data collection.
8.2.2.1 Recruitment

Recruitment was via the procedures explained in detail in Chapters Five and Six. All children were recruited through the CP clinics or gait laboratory of the Royal Children’s Hospital and were approached by the primary investigator (AH). Following explanation of the study the parent/guardians signed consent forms and were provided with information statements about the study. The data from the studies in Chapter Five and Six that was required for the current study were; GMFCS level, FMS score, FAQ score and descriptive details of the child such as age, gender and topographical distribution of CP.

8.2.2.2 Concurrent Validity

The information required for analysis of concurrent validity for each child was their FMS score, their GMFCS level and their FAQ score. Each child was classified according to GMFCS level by the primary investigator at the initial consultation for recruitment. This is described in Chapter Five (section 5.2.2.3) and Chapter Six (section 6.2.2.3). The GMFCS was developed for children with CP and has been shown to be reliable, valid and stable over time (Palisano et al 1997, Wood and Rosenbaum 2000, Morris et al 2004, Palisano et al 2006, McDowell et al 2007).

The FMS score utilised for the current study was dependent on which study the child was originally recruited for. In the reliability study each child had two FMS ratings. The one used for the current study was that one obtained at the Royal Children’s Hospital by either the physiotherapist or the surgeon. In the responsiveness study the FMS for each child was administered by the principal investigator. All FMS scores for the current study had been administered in person.

For all children the FAQ was completed by the parents at the initial consultation. The FAQ is a 10 level ordinal scale that the parents complete by circling the most appropriate response from 1-10 that best describes their child’s walking ability (see Appendix E). It ranges from “cannot takes any steps” through to “fully independent on all surfaces” within different environmental settings. The second part of the questionnaire involves the parents ticking the
“extra” activities their child can do from a list of 22 items. These items include activities such as walking while carrying an object, hopping on one leg, running and getting off an escalator independently. The FAQ has been shown to have high inter-rater reliability and good concurrent and construct validity for children with walking disabilities (Novacheck et al 2000).

8.2.2.3 Discriminative validity

The GMFCS level and FMS score for each child were used for the discriminative validity analysis. It involved examining the ability of the FMS to distinguish between groups of children based on their GMFCS levels.

8.2.3. Data Analysis

Descriptive statistics for participant characteristics were obtained using means and standard deviations for continuous variables and frequency and percentages for categorical variables. This included age, gender, GMFCS level, and topographical distribution of the participants. The distribution and mode of the FAQ and FMS levels for the group and for children grouped according to GMFCS levels was determined. The mean, standard deviation and range of extra items on the FAQ was also calculated.

8.2.3.1 Data analysis for concurrent validity

Concurrent validity was examined using Spearman rank correlation coefficients (Siegel and Castellan 1988). Data analysis examined the degree of correlation or association between each FMS distance and the FAQ as well as each FMS distance and the GMFCS. The FMS, GMFCS and FAQ are all ordinal scales. Nonparametric statistical analysis is required for this data. Spearman’s rho is a nonparametric analog of the Pearson r (Siegel and Castellan 1988), and is therefore the most appropriate to use with ordinal data to describe the strength and direction of a relationship between two variables (Portney and Watkins 1993). Spearman’s rho is the most frequently used measure of association for ranked data and is derived from the Pearson correlation coefficient (Norman and Streiner 2000).
8.2.3.2 Data analysis for discriminative validity

Discriminative validity was examined using the area under the receiver operating characteristic (ROC) curves (Streiner and Norman 2003). ROC curves can be used to judge the discrimination ability of various statistical methods (Hanley and McNeil 1982). It is derived from the early days of radar and sonar detection for finding the optimal setting where the largest ratio of signal to noise is detected, and has since been used to detect the presence or absence of a state or disease (Streiner and Norman 2003). The sensitivity (true positive rate) and specificity (true negative rate) of a test are derived from 2x2 tables and the ROC curve constructed from these values for each possible cut point of the test (Streiner and Norman 2003). A certain cut point on the curve for the scale or measure in question, in this case the FMS, is the optimal point where there are the most number of true cases (signal) without wrongly labelling cases as “non-cases” (noise).

For the current study ROC curves were created for each distance of the FMS after grouping children into two groups based on their GMFCS levels. Those who essentially were able to walk without assistive devices (GMFCS levels I and II) were labelled as “independent walkers”. This group can be considered as the true cases. Those who required assistive devices to mobilize (GMFCS levels III and IV) were labelled as “assisted walkers”. This group can be considered the non-cases. The aim of the data analysis was to determine if the FMS could distinguish between these two groups and what level of the FMS was the best discriminating level. ROC curves were created for each FMS distance. The points on the curve are the plots of the sensitivity (true positive rate) and 1 minus the specificity (false positive rate) for each FMS level (from 1-6). This will be explained further in the results section.

The area under the ROC curve measures the probability of a correct ranking of a pair as to the disease state in question (Hanley and McNeil 1982). In this case whether the FMS could distinguish between GMFCS I/II (“independent walkers”) and III/IV (“assisted walkers”). The estimate of the area under the curve can be calculated as well as confidence intervals constructed around this
Chapter 8 – Concurrent and Discriminative Validity

figure (Hanley and McNeil 1982). A non-discriminating test has an area of 0.5 and a perfect test has an area of 1.0 (Streiner and Norman 2003).

The data were stored and organized using the EpiData for Windows program (http://www.epidata.dk/). All analyses were performed using Stata [StataCorp 2005 Stata statistical software Release 9.0. College Station; TX. Stata Press.].

8.3. Results

8.3.1. Descriptive results

There were 213 sets of data available with 41 children included in both studies. For these 41 children, only one of their data sets was included. This resulted in 172 children being included in the current study. The mean age of the participants was 9.9 years (SD 3.7 years) with a range of 2-18 years. There were 111 males and 61 females. There were 27 children with hemiplegia, 114 with diplegia and 31 with quadriplegia. The frequency of children within each GMFCS level in the sample was; 19 children level I, 69 level II, 63 level III and 21 level IV. Table 8.1 summarises the participant characteristics.

Table 8.1 Participant characteristics

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<thead>
<tr>
<th>Characteristic</th>
<th>Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age: mean (SD), range (yrs)</td>
<td>9.9 (3.7), 2-18 yrs</td>
</tr>
<tr>
<td>Gender n (%)</td>
<td></td>
</tr>
<tr>
<td></td>
<td>male</td>
</tr>
<tr>
<td></td>
<td>female</td>
</tr>
<tr>
<td>GMFCS n (%)</td>
<td></td>
</tr>
<tr>
<td></td>
<td>I</td>
</tr>
<tr>
<td></td>
<td>II</td>
</tr>
<tr>
<td></td>
<td>III</td>
</tr>
<tr>
<td></td>
<td>IV</td>
</tr>
<tr>
<td>Topography n (%)</td>
<td></td>
</tr>
<tr>
<td></td>
<td>hemiplegia</td>
</tr>
<tr>
<td></td>
<td>diplegia</td>
</tr>
<tr>
<td></td>
<td>quadriplegia</td>
</tr>
</tbody>
</table>

For the group, the modal score for 5 metres on the FMS was 5 with 70 children (41%) rated as walking independently on level surfaces. For 50 metres the modal FMS score was also 5 (64 children or 37%). For 500 metres the modal score was 1 with 91 children (53%) using a wheelchair for community
distances. Figure 8.2 summarises the number and percentage of children in each FMS level for each distance.

Table 8.2 Frequency of children in each FMS category for each distance

<table>
<thead>
<tr>
<th>FMS level</th>
<th>FMS 5</th>
<th>FMS 50</th>
<th>FMS 500</th>
</tr>
</thead>
<tbody>
<tr>
<td>6</td>
<td>25 (14%)</td>
<td>18 (11%)</td>
<td>10 (6%)</td>
</tr>
<tr>
<td>5</td>
<td>70 (41%)</td>
<td>64 (37%)</td>
<td>49 (29%)</td>
</tr>
<tr>
<td>4</td>
<td>16 (9%)</td>
<td>12 (7%)</td>
<td>8 (5%)</td>
</tr>
<tr>
<td>3</td>
<td>8 (5%)</td>
<td>8 (5%)</td>
<td>5 (3%)</td>
</tr>
<tr>
<td>2</td>
<td>18 (11%)</td>
<td>43 (25%)</td>
<td>7 (4%)</td>
</tr>
<tr>
<td>1</td>
<td>11 (6%)</td>
<td>25 (14%)</td>
<td>91 (53%)</td>
</tr>
<tr>
<td>C (crawl)</td>
<td>24 (14%)</td>
<td>2 (1%)</td>
<td>0</td>
</tr>
<tr>
<td>N (does not do)</td>
<td>0</td>
<td>0</td>
<td>2 (1%)</td>
</tr>
<tr>
<td>Total</td>
<td>172</td>
<td>172</td>
<td>172</td>
</tr>
</tbody>
</table>

The modal FAQ score for the whole group was 6 (“walks more than 15-50 feet outside the home but usually uses a wheelchair or stroller for community distances”) with 42 children (24%). The next frequent category was 9 (“walks outside the home for community distances, easily gets around on level ground, curbs and uneven terrain, but has difficulty or requires minimal assistance or supervision with running, climbing and/or stairs”) with 37 children (22%). The mean number of extra items the children were able to perform was 8.1 (SD 5.5) with a range of 0-22. This range covers the whole possible range of extra items.

The mode of FMS level for each distance varied depending on the GMFCS levels of the children. For 5 metres the mode was at FMS 6 (independent on all surfaces) for children classified as GMFCS I, 5 (independent on level surfaces) for level II, crawling for children level III and wheelchair for level IV. For 50 metres the mode was level 6 for children classified as level I, 5 for level II, walking with walkers for level III and wheelchair for level IV. For 500 metres the mode was 5 (walking on level surfaces) for GMFCS levels I and II and wheelchair for levels III and IV. Table 8.3 shows the frequency of children in each FMS level for each distance according to GMFCS level.
Table 8.3 *Number of children in each FMS category for GMFCS level*

<table>
<thead>
<tr>
<th>GMFCS</th>
<th>FMS</th>
<th>6</th>
<th>5</th>
<th>4</th>
<th>3</th>
<th>2</th>
<th>1</th>
<th>Crawl/N</th>
</tr>
</thead>
<tbody>
<tr>
<td>I</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>(n=19)</td>
<td>5</td>
<td>17</td>
<td>2</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td></td>
<td>50</td>
<td>15</td>
<td>4</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td></td>
<td>500</td>
<td>9</td>
<td>10</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>II</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>(n=69)</td>
<td>5</td>
<td>8</td>
<td>60</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td></td>
<td>50</td>
<td>3</td>
<td>60</td>
<td>3</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>0</td>
</tr>
<tr>
<td></td>
<td>500</td>
<td>1</td>
<td>39</td>
<td>7</td>
<td>0</td>
<td>1</td>
<td>20</td>
<td>1 (N)</td>
</tr>
<tr>
<td>III</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>(n=63)</td>
<td>5</td>
<td>0</td>
<td>8</td>
<td>14</td>
<td>7</td>
<td>16</td>
<td>0</td>
<td>18</td>
</tr>
<tr>
<td></td>
<td>50</td>
<td>0</td>
<td>0</td>
<td>9</td>
<td>8</td>
<td>39</td>
<td>5</td>
<td>2</td>
</tr>
<tr>
<td></td>
<td>500</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>5</td>
<td>6</td>
<td>50</td>
<td>1 (N)</td>
</tr>
<tr>
<td>IV</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>(n=21)</td>
<td>5</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>11</td>
<td>6</td>
</tr>
<tr>
<td></td>
<td>50</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>3</td>
<td>18</td>
<td>0</td>
</tr>
<tr>
<td></td>
<td>500</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>21</td>
<td>0</td>
</tr>
</tbody>
</table>

From this table, it is evident that children classified as GMFCS level I and II tended to be at the higher end of the FMS (5 and 6) and walked independently. Children classified as level III displayed more varied mobility abilities from walking independently for short distances on level surfaces through to using all forms of assistive devices. The majority of children in level III and many in level II required the use of a wheelchair for the longer community distances. Some children in level IV used walkers or crutches for shorter distances; however, the majority used a wheelchair for all three distances. These results show the importance of the information obtained when using the FMS along with the GMFCS by providing details of the range of assistance over different distances.

The children who crawled for mobility in the home were separated out from the rest of the sample for the remaining analyses to enable statistical testing of the FMS categories 1-6. In the present form of the FMS, crawling is designated a “C” rather than given a numerical rating. The levels 1-6 all represent different forms of assistance required for mobility. Statistical analysis was therefore performed without the ratings from the children who crawled at home. The group of 24 children who crawled at home had a mean age of 6.6 years (SD 3.7) and range 2-14 years. They showed an even spread of FAQ scores but tended to fall within the 2-6 categories. Of the 24, 18 were classified as GMFCS level III and 6 were level IV.
8.3.2. Concurrent validity results

The Spearman rank correlation coefficients for each FMS distance correlated with the FAQ and GMFCS are presented in table 8.4. This calculation was performed on the sample excluding the children who crawled for 5 or 50 metres or who were rated as “N” (does not do) for 500 metres. After exclusion of these children there were 148 observations for FMS 5 metres and 170 observations for 50 and 500 metres.

<table>
<thead>
<tr>
<th></th>
<th>FMS 5 (n=148)</th>
<th>FMS 50 (n=170)</th>
<th>FMS 500 (n=170)</th>
</tr>
</thead>
<tbody>
<tr>
<td>GMFCS</td>
<td>r = -0.89</td>
<td>r = -0.91</td>
<td>r = -0.74</td>
</tr>
<tr>
<td>FAQ</td>
<td>r = 0.69</td>
<td>r = 0.76</td>
<td>r = 0.79</td>
</tr>
</tbody>
</table>

All p values for these correlations were 0.0000 rejecting the null hypothesis that there was no relationship between the two variables. The negative correlations with the GMFCS reflect the direction of the scale and classification. The FMS categories range from 6 to 1; from independent walking (6) to requiring a wheelchair (1). The GMFCS ranges from I-IV in the opposite direction.

Figures 8.1-8.3 display the correlations using scatter plots between each FMS distance and the FAQ while figures 8.4-8.6 show the scatter plots between the FMS and GMFCS. Because the FMS, FAQ and GMFCS are all measured on a categorical level, the scatter plots were enhanced to ensure that all points were considered. This is displayed on the graphs by the different sized circles on each of the coordinates. This illustrates that for some coordinate points a number of associations are represented.
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Figure 8.1 Scatter plot FMS 5 metres with the FAQ

Figure 8.2 Scatter plot FMS 50 metres with the FAQ
Figure 8.3 Scatter plot FMS 500 metres with the FAQ

Figure 8.4 Scatter plot FMS 5 metres with the GMFCS
The scatter plots for correlation of the FMS with the FAQ (Figures 8.1-8.3) show that there is a positive correlation for all three distances of the FMS. The 5 and 50 metre plots are similar with an increased number of points represented by level 6 on the FAQ and a grouping of points at the higher end on both scales. This suggests that those children who score higher on the FMS for 5 and 50
metres also score higher on the FAQ. The 500 metre distance shows a slightly different pattern with a large number of children who scored 1 on the FMS (use a wheelchair) showing a range of scores on the FAQ. There is also a grouping of scores at the higher end on both scales.

The scatter plots describing the relationship between the FMS and GMFCS (Figures 8.4-8.6) show clear negative relationships for 5, 50 and 500 metres. The negative direction is due to the opposite direction of categorisation for the respective scales. All three plots show a collection of children who score 5 on the FMS and are classified as GMFCS level II. Children classified as GMFCS level III show a range of scores on the FMS, particularly at 5 and 50 metres. Similar to the FAQ scatter plots for 500 metres, many children who score 1 on the FMS (use a wheelchair) show a range of GMFCS levels.

8.3.3. Discriminative validity results

For the calculation of the area under the ROC curve the children were grouped according to “independent walking” or “assisted walking”. There were 84 children in the “independent” group and 88 in the “assisted” group. The data for the children who crawled or “does not do” were omitted for the purpose of statistical analysis because they are not assigned a score between 1-6. Results for the area under the ROC curves are summarized in table 8.5. A non-discriminating test would have an area of 0.5 and a perfect test would score 1.0 (Streiner and Norman 2003). The results for each distance show the FMS to have good ability to discriminate between independent and assisted walkers, particularly for the 5 and 50 metre distances where the figures are close to 1.0. The graphs of the ROC curves are displayed for each FMS distance in Figures 8.7-8.9.

<table>
<thead>
<tr>
<th>FMS</th>
<th>Area under ROC</th>
<th>Standard error</th>
<th>Confidence Interval</th>
</tr>
</thead>
<tbody>
<tr>
<td>5 (n=148)</td>
<td>0.95</td>
<td>0.017</td>
<td>(0.92, 0.98)</td>
</tr>
<tr>
<td>50 (n=170)</td>
<td>0.97</td>
<td>0.015</td>
<td>(0.94, 1.0)</td>
</tr>
<tr>
<td>500 (n=170)</td>
<td>0.87</td>
<td>0.026</td>
<td>(0.82, 0.92)</td>
</tr>
</tbody>
</table>
Chapter 8 – Concurrent and Discriminative Validity

Figure 8.7 ROC curve for FMS 5 metres

Area under ROC curve = 0.9501

Figure 8.8 ROC curve for FMS 50 metres

Area under ROC curve = 0.9733

Figure 8.9 ROC curve for FMS 500 metres

Area under ROC curve = 0.8667
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The Y axis on the curves represents the sensitivity, or the true positive rate. The X axis represents 1 minus the specificity, or the false positive rate. The diagonal line that runs from point (0,0) to (1,1) reflects the characteristics of a test that would have no discriminating ability. The better the FMS is able to discriminate cases from non-cases, or in this case independent walkers from assisted walkers, the closer it will approach the upper left hand corner (Streiner and Norman 2003). The curves for each FMS distance show good discriminating ability of all three distances. The curves for the 5 and 50 metres are closer to the upper left hand corner, suggesting these distances discriminate slightly better than the 500 metre distance.

On the curves, the score that is closest to that upper left corner is the cut-point which minimizes the total number of errors (false positive and false negative) (Streiner and Norman 2003). For the current study this point is the cut-point between two levels on the FMS that best discriminates independent from assisted walkers. Figure 8.7 displays the curve for the 5 metre FMS distance. The point on this curve that is closest to the upper left corner is the cut-point between levels 4 and 5 on the FMS. Therefore, for 5 metres the point between walking using sticks or holding hands or walls and independent walking on level surfaces discriminates whether children are classified into GMFCS I/II (independent) or II/IV (assisted). For 50 metres (Figure 8.8), the closest point is also between levels 4 and 5, however the point between levels 3 and 4 (between using crutches and walking with sticks or holding hands/walls) is also very close. For 500 metres the cut-point is between levels 3 and 4 on the FMS.

8.4. Discussion

The concurrent validity of the FMS was confirmed by the results in this investigation with good correlations for all three distances with both the FAQ and the GMFCS. Discriminative validity of the FMS was also substantiated with its ability to distinguish between “independent” and “assisted” walkers based on the GMFCS levels of the children and with good agreement between calculated and true GMFCS levels.
8.4.1. Concurrent validity

Concurrent validity was examined using an outcome measure, the FAQ, and a classification system, the GMFCS. The highest correlations were between FMS 5 metres and the GMFCS and between FMS 50 metres and the GMFCS. The GMFCS classifies children according to their gross motor function and is based on activity limitation. A strong correlation of the FMS with the GMFCS provides important evidence that it is measuring activity limitation. The highest correlation of the GMFCS with FMS 50 metres suggests it is this distance that clinicians consider when allocating a GMFCS level to a particular child. The FAQ measures walking ability and is the most similar measure to the FMS. It is reassuring that the correlations with the FAQ were high, suggesting they are both measuring the same aspect of activity. It is also reassuring that the correlations were not perfect, and varied between the three subscales of the FMS, ensuring there is no issue of redundancy of the FMS.

The FMS and the FAQ are both measures of mobility. They differ on the construct of assistance required for mobility with the FMS differentiating the assistance required in different environmental settings. The FAQ is rated independent of the assistive devices the child uses. If the correlations were found to be very high it might indicate some overlap between the scales and suggest redundancy of one of the measures. The correlations found show strong yet imperfect relationships between the two measures. This suggests they measure similar domains while being sufficiently different to ensure the clinical utility of both. The FAQ is not considered a gold standard for measuring mobility in children with CP. In the absence of such a gold standard it was considered to be the most similar tool for correlation with the FMS. The FAQ had not been considered in the preliminary work on the FMS (Graham et al 2004). The GMFCS is not an outcome measure, it is a classification system. The good correlation with it provides evidence that the FMS is measuring gross motor function and across the spectrum of severity of motor impairment seen in children with CP.
There is considerable literature on the correlation of outcome measures used for children with CP. Many studies have examined the relationship between these measures without formally examining concurrent validity of one particular outcome measure (Drouin et al 1996, Ottenbacher et al 1999, Schneider et al 2001, Tervo et al 2002, Abel et al 2003, Oeffinger et al 2004, Sullivan et al 2007). The strongest relationships have been found between those tools measuring change in physical function rather than those assessing social and cognitive status (Oeffinger et al 2004, Sullivan et al 2007). Stronger relationships were found between tools where both measure activity limitation than where one measures activity and the other measures body structures and function (Tervo et al 2002, Abel et al 2003).

Other studies examining outcome measures that concentrate on activity limitation in children with CP have supported the concurrent validity of the ASK (Young et al 2000, Pencharz et al 2001), the CHQ (Pencharz et al 2001, McCarthy et al 2002), the FAQ (Novacheck et al 2000), the GMFM (Russell et al 1989), the PEDI (Wright and Boschen 1993, McCarthy et al 2002), and the PODCI (Daltroy et al 1998, Pencharz et al 2001, Vitale et al 2001, McCarthy et al 2002, Damiano et al 2005, Vitale et al 2005). The overall theme for all of the studies is that correlation was highest for like domains, such as the FAQ with the PODCI transfers and mobility domain (Novacheck et al 2000) and for tools that measure the same constructs, such as the WeeFIM and PEDI (Ziviani et al 2001). Correlations were found to be lower between tools measuring different constructs such the FAQ with energy expenditure (Novacheck et al 2000) or between unlike domains of multidimensional scales such as the CHQ and PODCI (Pencharz et al 2001, McCarthy et al 2002).

The preliminary study of the concurrent validity of the FMS showed strong correlations with Uptime, PODCI and CHQ, moderate with the Hoffer scale and lower with energy expenditure (Graham et al 2004). Like the studies examining concurrent validity of other activity limitation tools, it is expected that correlations would be higher with tools measuring similar constructs. Lower correlation with energy expenditure is not unexpected because it is measuring a physiological response of the body to activity as opposed to the FMS that is a
scale measuring assistance required for mobility. As stated earlier, a limitation of the Graham study was that the FAQ was not used as a criterion measure. The current study addressed this deficiency and found good correlation between the two measures of mobility.

### 8.4.2. Discriminative validity

Examination of discriminative validity using the ROC curves showed that the FMS is able to discriminate between “independent” and “assisted” walkers based on their GMFCS levels. The 5 and 50 metre distances had slightly stronger discriminative ability than the 500 metre distance. ROC curves allow for discrimination between two groups only, hence grouping the GMFCS levels into I/II and III/IV. The results provide evidence of discriminating between these two groups, although not between all GMFCS levels. For example the results do not discriminate between levels I and II, levels II and III or levels III and IV. The results provide some evidence of the ability of the FMS to discriminate severity of activity limitation.

Previous investigations have examined the discriminative validity of other activity limitation outcome measures used for children with CP. The ASK was able to discriminate severity of condition (mild, moderate or severe) (Young et al 2000), and Hoffer ambulation level (Pencharz et al 2001) in children with musculoskeletal disorders. The CHQ was not able to discriminate all levels of topographical distribution of CP (McCarthy et al 2002, Vitale et al 2005) and had only a weak ability to distinguish Hoffer ambulation level (Pencharz et al 2001). The GMFM showed a hierarchy of severity of CP (Russell et al 2000). The mobility domain score of the PEDI discriminated topography of CP (McCarthy et al 2002). The PODCI was able to discriminate Hoffer ambulation level (Pencharz et al 2001) and topography with the transfers and mobility domain (McCarthy et al 2002, Vitale et al 2005). The WeeFIM showed only a weak ability to discriminate topography (Msall et al 1994).

These reports all provide evidence of the varied abilities of the tools to discriminate, however the main limitation of all of these studies is that they have not examined the ability to discriminate GMFCS level (Palisano et al 1997).
Chapter 8 – Concurrent and Discriminative Validity

The studies have used the ability to discriminate topographical distribution of CP, Hoffer ambulation level or severity of condition (mild, moderate or severe) as their classification. As stated in Chapter Two, the GMFCS is now used widely to classify the motor function of children with CP. It is important that the GMFCS is considered when examining the discriminative validity of these tools. For some of the above studies, the timing of the study in relation to the timing of the introduction of the GMFCS into the field of measurement in CP explains why it has not been examined. Further examination of discriminative validity using the GMFCS is warranted.

A more recent investigation examined the ability of a number of tools to discriminate GMFCS level in a large sample of 562 children with CP aged 4-18 years, GMFCS levels I-III (Bagley et al 2007). The data was analysed using effect size indices, odds ratios, regression analysis and ROC curves. Overall the study found that measures of physical function were able to differentiate GMFCS levels better than those measuring cognitive function or quality of life. In isolation the GMFM-66 was the best at discriminating between GMFCS I and II and between II and III. Together the GMFM, gait velocity and the WeeFIM mobility domain had the best ability to discriminate between levels I and II. Together the GMFM, WeeFIM, cadence and FAQ had the best ability to discriminate between levels II and III. It is not surprising that the GMFM in isolation had the highest discriminatory ability. This can be explained by the background to the development of the GMFCS, which is based on the GMFM scores of 275 children with CP of differing severity (Palisano et al 1997). It is also not surprising that the physical function measures were better able to discriminate because the GMFCS is based on motor function not cognitive or quality of life. This study is important because it does use the GMFCS, however its main limitation is the children included represent levels I-III only, and the results can only be generalized to this limited population.

The current study used the GMFCS in the examination of discriminative validity. The sample included a broader range of GMFCS levels than the Bagley study with children in levels I-IV represented. The FMS had not been previously examined for discriminative validity using the GMFCS. The
preliminary study examined its ability to distinguish Hoffer ambulation level (Graham et al 2004), however as stated in the introduction of this chapter, no formal statistical analysis was performed. The current study provides good evidence of the ability of the FMS to discriminate one aspect of severity of motor impairment based on the most widely used classification system of motor impairment.

Because the FMS is an evaluative measure, the most important aspect of validity to examine is its ability to detect clinically important change over time (Kirschner and Guyatt 1985). This was examined in Chapter Six. Although it is not imperative that the FMS is able to discriminate all levels of the GMFCS, the results of the current study on concurrent and discriminative validity provide further evidence of the construct validity of the tool by examining the relationship between measures concerning the concept of interest (Nunnally 1978, Kirschner and Guyatt 1985, Streiner and Norman 2003). Together all experiments on validity and responsiveness support the overall validity of the FMS as a psychometrically sound measure of activity.

8.4.3. Limitations and recommendations

There are a number of limitations of this investigation. The sample used was a combination of two cohorts of children recruited for the investigations into the reliability and responsiveness of the FMS. Both of those samples were samples of convenience and the children were recruited consecutively from the clinics and gait laboratory of The Royal Childrens Hospital, Melbourne, Australia. The limitations of using samples of convenience have been discussed previously. The main limitation of such samples is the potential to introduce a biased sample that does not represent the population of interest. Although a random sample would minimise this bias, the clinics from which the children were recruited comprise those children that the FMS is routinely used with. It is therefore likely that the sample closely resembled the population of interest.

The sample used in the current study comprised more males (65%) than females (35%) and children predominantly with spastic diplegia (66%) than hemiplegia (16%) or quadriplegia (18%). The spread of GMFCS levels of the children was
I-IV yet many children were classified as level II (40%) or III (37%). The sample did not include children classified as level V because the FMS was developed for levels I-IV. The findings can only be generalized to children with CP aged 2-18 years classified as GMFCS levels I-IV. Although the sample is biased towards children with diplegia who are classified as GMFCS II-III, this represents the children who attend the clinics and gait laboratory and for whom the FMS is relevant. This was discussed in Chapter Five.

The data collection procedures for the two studies from which the current study evolved were different. The GMFCS levels of all children involved were classified by the primary investigator. The FMS scores, however, were administered by different clinicians. The primary investigator scored the FMS for the children in the responsiveness to change study. Within the reliability study the FMS ratings were made by different clinicians with a total of seven clinicians involved from the hospital setting. This has the potential to introduce variability in the ratings obtained and influence the level of agreement and correlation between the measures. It would be preferable to have the same person collecting all FMS scores for all children involved.

The data analysis of the Spearman rank correlations coefficients and the ROC curves excluded those children who had been classified as “C” (crawls) or “N” (does not do). This was necessary to enable clinically meaningful statistical analysis. It has not yet been determined which is the most appropriate method for analysing these categories of the FMS. A possible recommendation from the thesis might be to modify the FMS as it currently presents.

8.5. Conclusion

The FMS showed good concurrent validity with the FAQ and the GMFCS. The FMS is also able to discriminate between children who walk independently from those who require assistance based on their GMFCS levels. Although the focus of evaluative measures is to detect clinically meaningful change, the results of this investigation provides further evidence to support the overall validity of the FMS.
CHAPTER NINE: GRAND DISCUSSION AND CONCLUSIONS

The studies in this thesis have focused on specific aspects of psychometric testing of the FMS and have critically evaluated measurement tools used to assess activity in children with CP. This final chapter combines the findings to emphasize the major themes to emerge. In addition to synthesizing the findings, this chapter will also discuss the clinical implications of this research and the limitations of the research. Future directions will be presented to include future research using the FMS as well as potential changes to the scale, followed by the overall conclusions.

9.1. Synthesis of findings

There were a number of major themes to emerge from this thesis. These themes encompass; the importance of having clinically useful and psychometrically sound tools available to measure activity, the influence of the environment on assistance required for mobility and the issues around self-reporting and whether it reflects performance.

9.1.1. The importance of robust tools to measure activity limitation

Mobility involves “changing location or transferring from one place to another” (World Health Organisation 2001). It stands to reason that optimal mobility of children with CP will encourage their participation with their peers and family. This thesis has highlighted the need to quantify mobility using reliable and valid tools. It has emphasized the importance of being able to measure the changes in assistive devices utilized by children as they grow or following therapeutic interventions (Harvey et al 2007). The influence of the environment on activity limitation, in particular mobility, constantly emerged throughout this thesis. The ability to be able to quantify this influence is pertinent. The systematic review in Chapter Three showed that despite there being several measurement tools available that assess activity, the FMS is the only one that quantifies the assistive devices required in the different environmental settings of the home, school and community. The ability of the FMS to detect both the deterioration and improvement following complex interventions such as SEMLS, as shown in
the pilot study in Chapter Four, is critical in the clinical setting where the aims of costly interventions is to improve or maintain the mobility status of these children.

The collective results of the studies in Chapters Five to Eight demonstrated that the FMS, which has a high level of face validity, is a psychometrically sound measurement tool. The results of the inter-rater reliability and concurrent and discriminative validity studies were conclusive. The responsiveness to change study provided some important evidence of the ability of the FMS to show change where change occurred and also showed evidence of stability where no change in mobility was expected. Further work in this area is required as will be reported in future directions. The construct validity study was a feasibility study and revealed some very important issues regarding the nature of self or parent reporting. These will be discussed further later in this section. Although the studies were classified as separate psychometric entities, many of the studies provided evidence of the construct validity of the FMS, that is, Chapters Six, Seven and Eight. Validation is an ongoing process and the studies in this thesis have contributed greatly to the validation of the FMS as a clinical and research tool for quantifying mobility.

9.1.2. The influence of the environment on mobility

Another theme to emerge overall was that children with CP use different mobility methods. Chapter Seven found that not only is there a range utilised by children generally, the same child often uses more than one method of mobility across and within environmental settings. There are a number of potential explanations for the variety seen within the group and for individual children. The severity of the motor impairment of the child, as described by their GMFCS level, will play a role in mobility methods used. Children in levels I and II walk independently and therefore walk without assistive devices. Children in level III use assistive devices to ambulate and it is this level where the most variety of methods was seen. Children in level IV use a wheelchair for the majority of the time with minimal walking for short distances only, usually with a walker and adult assistance. Children in level V use wheelchairs for all forms of mobility.
The other significant influence on mobility methods used by children with CP is the environmental setting. This thesis has shown that mobility and the methods chosen can be influenced markedly by the environmental setting in which the child is moving around. Within each setting, some of the factors that have the potential to influence mobility are safety, familiarity with the environment, access and liability issues. Within their home the child is very familiar with the environmental conditions and distances are shorter. They would feel safe and be able to move around with less assistance than at school or in the community, where distances are longer and the environment is less familiar (Palisano et al 2003).

Some of the factors related to the school environment that can influence mobility are liability issues around safety at school, greater time pressure, and the presence of obstacles impeding free movement (Haley et al 1994). Legal issues can determine methods used and in some instances may limit a child’s potential function, as noted through clinical experience from parental report. For example, a child who is able to use crutches might be instructed by the school to use a walker or wheelchair at school due to concerns the child might hurt themself. Without the everyday practise of using the crutches the child’s activity is being compromised. Time pressure for the child at school is particularly pertinent throughout the secondary school years where children are expected to be more independent. The adolescent with CP might choose to use a wheelchair, even electric wheelchair, so that they are able to get to class on time without assistance.

Other determinants of mobility methods used might include time pressures, convenience and choice. Clinical experience suggests that parents will often push their child in a wheelchair when they go shopping because it takes less time and behavioural issues can be better managed. Many children with CP have high equipment needs. For a child who has more than one assistive device, the parent might choose to leave one at school and one at home. For example, the family of a child who uses crutches at home and a walker at school might choose to leave the walker at school for convenience. This could limit the choice for community distances where the only choice is now wheelchair
because the walker is at school. The other determinant is personal choice of the child. For instance, some children might be physically able to use crutches, yet they choose to use a walker because they are faster and more stable with it and can keep up with their peers more easily.

9.1.3. Issues around self-report measures

Another major theme to emerge from the thesis is the issue of whether self-report measures, such as the FMS, really do measure performance and whether they reflect what is truly happening. The results from the investigation in Chapter Seven revealed the question mark around whether self-report methods are a valid way of measuring performance. Performance measures should reflect what the child does in everyday function (Young and Wright 1995). Self-report measures are thought to be effective in obtaining information on typical performance because it can be difficult to obtain this from a formal assessment situation (Long 1992). Clinicians assume that what is seen in the clinic or reported from the parents about the child’s function reflects usual performance (Rosenbaum 2007). It is usually assumed that respondents will answer honestly to items in a questionnaire (Streiner and Norman 2003). There are a number of factors that might influence the validity of self or parent report including; knowledge of the child’s mobility at school, responder bias and wording of questions from clinicians.

Chapter Seven discussed how parents might be less familiar with their child’s mobility at school than the community physiotherapist who visits the child at school. Alternatively the parent is likely to be more familiar with their child’s mobility at home. These differences in familiarity of a child’s mobility in different settings have the potential to influence the accuracy of the report given. Responder bias can also influence self-report measuring. Parents might want to report the capability of their child, that is their best function, if they feel it might avoid an intervention. Alternatively they may rate their child lower if they feel it would result in an intervention. The former situation can be considered “faking good” or social desirability and the latter as “faking bad” or deviation (Streiner and Norman 2003). These issues are also relevant if the child is the respondent. The way the questions are asked can determine what is
reported. A study compared the answers to specific questions of the CHQ, PODCI and PEDI from the parents of 66 children with CP (Wren et al 2007). Differences in wording had an effect on parent’s responses suggesting that wording and context of questions is important.

An example of a different ratings obtained from the same child was found in the reliability study in Chapter Five. One of the ratings was performed by the child’s usual community physiotherapist rating the child at school (who was 16 years old with no cognitive impairments) and a rating of 5, 1, 1 was obtained. The second rating was performed by a hospital physiotherapist who was unfamiliar with the child. It was performed within the clinic setting by asking the father of the child and a rating of 4, 1, 1 was obtained. The difference in scores could be the result of the responders (child report versus parent) or because the community physiotherapist was less familiar with the child’s performance at home and that influenced her rating.

Despite these issues with self-report measures, they remain the most convenient method of measuring performance in activities of children with CP. A more recent suggestion has been that services need to change the balance of focus from the clinic to the home and community to observe the children in their environments (Rosenbaum 2007). It is not always practical for clinicians to observe children in their own environments. They rely on self-report measures to gather that information.

Another issue of using self-report measures is determining who the best responder is. The options include the child, the parent or a proxy. Each will provide a different perspective of the child’s function. Other tools that measure activity involve varying perspectives. The GMFM measures capability through direct observation of the child by the therapist. The FAQ reports the parent perspective. The FMS uses either child or parent report interpreted by the clinician. The ASK reports the child perspective unless they are unable to complete it themselves. The CHQ and PODCI have both adolescent and parent versions. The WeeFIM reports the clinician perspective and the PEDI reports a combination of parent and clinician perspective.
It is now thought that the child’s view should, where possible, be sought directly rather than being inferred from proxy reports such as parents (Colver and Group 2006). However, age or cognitive ability may impair communication (Young and Wright 1995). Children may not be able to convey information regarding their own experience, thus relying on parental and caregiver proxy reports (Simeonsson et al 2003). In a sample of 148 children with CP, 47% were able to self-report using the PedsQL (Varni et al 2005). The reliability of child report was examined in 28 children with musculoskeletal disorders using the ASK (Young et al 1995). Retest reliability of the children was excellent and their scores were highly concordant with parent-reported ASK scores. Child report has been shown to differ from parent report on the PODCI and PedsQL with child report values higher (Oeffinger et al 2007). Because different perspectives may potentially give different responses, it might be clinically beneficial to obtain both the child and parent perspective.

9.2. Clinical implications

The FMS is an evaluative measure that has unique characteristics by quantifying and differentiating the assistance required in the different environmental settings of home, school and community. No other outcome measure provides this information. An evaluative measure must: contain relevant items, be applicable to the population for whom it was developed, be feasible to use, be reliable and valid for that purpose and be responsive to clinically important change over time (Rosenbaum et al 1990). This thesis has provided evidence of these requirements for the FMS.

The FMS has sound psychometric properties for children with CP aged 4-18 years who are classified as GMFCS levels I-IV. Although the FMS was not developed with scientific rigor, the studies in this thesis have validated it and proved that it is a clinically useful, reliable and valid measure that is able to show change in mobility. The FMS is now available for use by physiotherapists and other clinicians to quantify mobility in children with CP and measure changes associated with growth and development and following costly
interventions, such as orthopaedic surgery. It is available as both a clinical assessment tool and for use in research studies.

The FMS is a feasible outcome measure. Feasibility relates to the time needed to perform the measurement, the equipment required and training required for administration (Dekker et al 2005). The FMS can be administered within 10 minutes and does not require any equipment for administration. Formal training is not required, however it is suggested that clinicans read the brochure that has been developed as part of this thesis prior to using the scale because it provides guidelines for administration. The FMS can also be administered over the telephone because it is a clinician interpreted self-report measure. This adds to its feasibility. Outcome measures that are more time consuming to administer and require equipment and/or training, such as the PEDI and GMFM, provide more detailed information but may lose some clinical utility as a result.

Chapter Four showed that the FMS is a clinically useful tool that can measure change in mobility following interventions (Harvey et al 2007). Many children with CP have simple and complex interventions throughout their childhood and it is important to have reliable and valid tools that can measure deterioration and improvement in activities, including mobility, following these interventions. Used in conjunction with other simple ordinal measures and classifications such as the FAQ and GMFCS it is able to provide a good description of activity in children with CP. The FMS is specific to mobility. Other measurement tools available may be less specific, such as the ASK, CHQ, PODCI and PEDI. They give broader picture of activity or health status in general, however there is the potential to lose specificity with that.

Because the FMS is a clinically feasible tool that does not have high training or equipment needs it can be used in many clinical settings. This includes settings where clinicians do not have access to highly technical equipment as would be found in a gait laboratory. This also includes rural settings and developing countries where access to expensive technology may be uneven or restricted (Graham and Harvey 2007). Another advantage of the FMS is it can be used by a range of clinicians, as shown in the reliability study in Chapter Five. It is a
multidisciplinary tool for use by those who assess activity limitation in children with CP, including physiotherapists, orthopaedic surgeons, rehabilitation specialists, physicians and paediatricians.

Chapter Two introduced the ICF in relation to outcome measurement for children with CP. The FMS does not claim to measure all aspects of activity, it focuses on mobility. Used in conjunction with other tools that also assess activity, such as the FAQ, GMFM and ASK, as well as those that examine aspects of body structures and functions and participation, a comprehensive assessment of a child with CP is provided. For example, a child with CP who may require multilevel surgery could be assessed pre-operatively with three dimensional gait analysis, radiology and range of movement measures to cover body structures and functions. The FMS, FAQ, GMFM and ASK could be included to examine activity and a measure, such as the CAPE, to cover participation. The result could be a comprehensive set of tools providing information from all areas of the ICF, including both aspects of performance and capability and both self-report and direct observation. The same set could then be used post-operatively to compare with pre-operative status to examine the effects of the interventions.

 Chapters Three and Six emphasize the importance of matching outcome assessment tools to the aims of the measurement. If the measure selected is used to assess change after an intervention, a consideration of the aims of that intervention is necessary. This ensures that clinicians use the most appropriate tools rather than basing selection on familiarity or popularity. For example, to assess change after Botulinum toxin injections, the most appropriate measure might be measures of spasticity and range of movement because the aim of the intervention is to reduce spasticity and prevent the development of contractures (Graham et al 2000). As shown in Chapter Six, the FMS may not show any change because the mobility status of the child would not be expected to change. Alternatively, the aim of SEMLS is to correct deformity, improve the gait pattern and maintain or improve mobility. The appropriate measures in this case could include three dimensional gait analysis to detect changes in gait pattern, range of movement measures to show change in deformities and tools
such as the FMS to show changes in mobility. Using inappropriate tools for that purpose might lead to inaccurate conclusions about no change in the concept being measured, when in fact change has occurred in a domain not assessed by the measure.

Use of the FMS in the clinical and research settings has gained momentum since its initial development and publication. Studies have used the FMS to show changes in mobility following hamstring surgery in children with bilateral CP (Ma et al 2006). The 50 metre distance of the FMS was used to compare with the Physiological Cost Index in children with CP to evaluate the efficiency of ambulation (Raja et al 2007). The FMS has also been used in children with myelomeningocele to compare the functional mobility of children who have a ventriculoperitoneal shunt with those who have no shunt (Battibugli et al 2007).

It is anticipated that following publication of the results obtained in this thesis that use of the FMS will continue to grow in the clinical and research arenas.

9.3. Limitations

The limitations of the individual studies were discussed in detail within the relevant chapters. The main themes to emerge regarding the limitations of the studies revolve around the sample biases, the sample sizes and the recruitment process.

The samples in the reliability, responsiveness to change and concurrent and discriminative studies had a bias towards children aged between 6-12 years, with spastic diplegia classified as GMFCS levels II and III. This reflects the demographic of children who attend the hospital clinics and gait laboratory. This bias has thus meant that these studies focus on the target population for which the FMS was developed and for whom the scale is used clinically. A sample of convenience was used in the construct validity study and consecutive sampling was used for the remaining studies. As stated within the chapters, because random sampling was not employed there is a possibility of the samples not being representative of the population for which the results are intended. The consecutive sampling from the clinics that children of the target population
attend reduced this potential for bias through selective recruitment, suggesting that the sample is in fact likely to be representative of the target population.

Time constraints and the logistics of ensuring adequate follow-up of participants resulted in the sample for the construct validity study in Chapter Seven being small. This study was intended as a feasibility study and further research into the area of agreement between self-report FMS and direct observation is warranted. Despite the results of that study being inconclusive due to the small sample it did draw attention to some very important clinical issues around the use of self-report measures and the measurement of performance.

The participants were recruited from one centre only. This centre is the major tertiary care facility for children with CP within the state of Victoria, Australia. Although there is the potential that the generalisability of the results are limited due to using only one recruitment centre, the nature of the centre and its catchment area ensures that the results can probably be generalised more widely.

Some of the FMS ratings in the investigations in Chapters Six and Seven were performed over the telephone. The results of the reliability study in Chapter Five showed similar agreement between FMS administration over the telephone and FMS administration in person. Despite this, there is the possibility that ratings with the child present would be different to those obtained over the telephone. It is difficult for clinicians to ignore a child’s function when they can observe it. These clinical observations could influence ratings, particularly if what is observed conflicts with the self or parent report. It would have been preferential for all FMS ratings to be performed in person for the investigations in those chapters to minimise the effect of such potential discrepancies.

All attempts were made prospectively to ensure that the effect of any limitations would be minimised within each study. Although these limitations are present, they are not significant enough to detract from the results obtained in the thesis overall.
9.4. Future directions

The investigations in this thesis have comprehensively examined the FMS and through this process areas for further work can be identified. Future directions resulting from the findings include potential changes to the scale and further research endeavours with the FMS that could advance knowledge into the field of outcome measurement for children with disabilities.

A number of limitations of the FMS itself became evident throughout the various investigations. The scale is called a “mobility” scale, however it focuses on walking and the assistive devices used to ambulate, with one category for the use of wheelchairs. It does not take into account all forms of mobility and therefore could be considered a “walking” scale or an “ambulatory” scale. As was emphasized in Chapter Seven, the scale does not include more supportive walkers that the children require adult assistance to be strapped into because it was developed to focus more on independent mobility. It is possible that these types of walkers could be incorporated as a sub-category of category 2 within the scale because these types of walkers are frequently used in school settings for children classified as GMFCS level IV.

The wheelchair category of the FMS currently makes no distinction between whether children push themselves independently, require an adult to push them or use an electric wheelchair. It would be beneficial if the scale could distinguish between these abilities because each represents differing levels of function of the children. A study examining the anatomy of the dysplastic hip in children with CP devised an assessment scale of locomotor function making these distinctions (Vidal et al 1985). An ordinal scale from 0 to 8 described function ranging from “bedridden with no motor function” to “independent walking inside and outside”. Distinctions for wheelchair use included “without self propulsion”, “self-propulsion for short distances” and “independent wheelchair existence both in and out of the home”. Although the scale has not been examined for reliability or validity, similar distinctions for wheelchair use could be incorporated into and enhance the FMS.
Chapter Seven indicated that more explicit instructions or questions for administration of the FMS may be required to obtain a true performance rating. Currently the brochure provides guidelines for questioning without using set questions. This allows for clinical judgement and reasoning to guide the questioning process. During scale development it was thought that if it was more prescriptive clinicians could miss key information. A more structured questionnaire may be more direct and ensure more reliable answers. The potential trade-off with that approach could be clinicians not using problem-solving skills and missing information with the scale losing some validity in the process.

The investigation in Chapter Seven also showed that within the home and school settings there were different distances that children travel, for example within the classroom or between classrooms at school. This has the potential to create some confusion when rating, and consequently could affect agreement between different clinicians (Chapters Five and Seven). The accuracy of the FMS might be improved if FMS administration stipulated that clinicians consider distances within one room at home (for the 5m FMS distance) and between classrooms at school (for the 50m FMS distance). This would ensure that the distances for each setting were distinct and might improve consistency between ratings.

Feedback from some of the raters in the reliability study in Chapter Five highlighted potential confusion regarding the “4” category of the FMS. Currently “4” represents the use of single point sticks for ambulation. Category “5” represents children who are independent on level surfaces and require the use of a rail for stairs. The asterisk in category “5” states that if the child uses furniture, walls, fences or shopfronts for support the child is rated as “4”. There is the potential for confusion between using rails for stairs which rates as “5” and walls for walking which rates as “4”. Other confusion was reported for the situation where children hold hands with an adult rather than use an assistive device. Clinicians tend to rate that as “4”; however there is a difference in the levels of independence between using walls or holding hands and using single point sticks. Clarification of these issues is required by ensuring that single
point sticks represents a distinct category from holding hands or walls for support.

This thesis has shown that the FMS is a psychometrically sound and clinically useful tool for children with CP. It has the potential, therefore, to also be a useful tool in other populations. Further research of the FMS could involve examining its reliability and validity in adults with CP and children with other conditions such as myelomeningocele and acquired brain injury. This would enable it to be a more generic measure, with the advantage of comparing children with different conditions (Dekker et al 2005). Interesting and important epidemiological studies could then be performed to examine activity limitation and mobility in children with a range of conditions.

The issue highlighted previously of whether self-report tools measure performance of activity limitations in children with CP requires more research. The study in Chapter Seven showed low agreement between self-report and direct observation. Although the numbers in that study were small and there were some methodological limitations, this is an important clinical and research issue. There are a number of other activity limitation measures used in children with CP that also rely on self-report. There has been some research on reliability of self-reporting but little into whether this is a valid method of measuring performance. Further examination of this for the FMS is required and for other self-report measures such as the FAQ, the ASK, the PODCI and CHQ. Researchers and clinicians need to be able to answer the fundamental question of “What are we really measuring?”

The inter-rater reliability of the FMS examined in this thesis used a large number of clinicians from different professional backgrounds. Further investigation of the reliability of clinicians based on clinical experience would be relevant. This could determine whether clinical experience is required to reliably administer the scale by comparing students or newly graduated clinicians with clinicians with more extensive clinical experience. The generalisability and utility of the FMS would be augmented if reliability was found to be good for clinicians with varying levels of expertise.
The FMS is an evaluative measure and proven responsiveness to change is an important property for such measures. It is particularly important that the FMS can detect clinically important change after treatments. The examination of responsiveness in Chapter Six provided some evidence of this for the FMS. To further establish its responsiveness, examinations of different interventions using different statistical methods and a proven stable group of children to compare with is required. In addition, a more scientific approach to establishing the minimal clinically important difference for the FMS by seeking consensus from a larger number of clinicians experienced in its use would be beneficial. The statistical analyses performed in all investigations in this thesis were limited due to the categorical nature of the data. It would be advantageous to better understand and develop statistical methods for expressing change in categorical data when changes in single categories are significant.

9.5. Conclusions

There are four major issues to consider with measurement; specifying the measurement goal (what to measure), ensuring the instrument selected actually measures these concepts, understanding how well the instrument is able to measure these concepts, and how to interpret the outcomes of the instrument (Terwee et al 2003). The aim of this thesis was to provide a psychometrically sound and clinically useful measurement tool that quantifies mobility in children with CP. This aim has been fulfilled with the main conclusions from the investigations listed below.

1. A systematic review was conducted of the psychometric properties and clinical utility of the available evaluative measures of activity limitations for children with CP (Chapter Three). This found:
   i. Eight evaluative outcome measures (ASK, CHQ, FAQ, FMS, GMFM, PODCI, PEDI, WeeFIM) with each examining different dimensions of activity limitation.
   ii. The ASK and GMFM to have the most robust psychometric properties.
iii. The reliability of all tools to be good, although further examination of validity and responsiveness to change is required for the CHQ, FAQ, FMS and PODCI in children with CP.

iv. The GMFM to measure capability whilst all other tools measured performance. The ASK and PEDI quantify both performance and capability.

v. Different environmental settings to be incorporated within all tools except for the GMFM.

vi. The FMS to be the only tool to describe activity with different assistive devices within each of those settings. However it requires further examination to establish inter-rater reliability, validity and responsiveness.

vii. Each tool to have a different purpose for application in CP. A range of tools may be required by clinicians to cover all domains of the ICF for comprehensive assessment of children with CP.

2. Chapter Five examined the inter-rater agreement for each distance of the FMS for children with CP using a large number of raters from different clinical backgrounds and found:

i. Substantial agreement for each of the three FMS distances using a range of clinicians as raters.

ii. There were no significant differences in agreement between raters for different age groups, GMFCS levels or topographical distribution of CP in the sample of children.

iii. Similar agreement between raters who administered the FMS in person and those who administered it over the telephone, suggesting it can be administered by either method.

iv. Although surgeons tended to rate more of the higher functioning children at the upper end of the scale than physiotherapists, the evidence was not conclusive.

v. The FMS can be used as a reliable tool for different clinicians to assess mobility in children with CP.
3. A study investigated the responsiveness to change of the FMS for detecting clinically significant change following orthopaedic surgery and spasticity management (Chapter Six). It found:
   i. The FMS was able to detect changes in mobility in children with CP as defined by improvement or deterioration of one category or more on the FMS scale.
   ii. The FMS was able to detect clinically important change in children who had SEMLS. It also showed minimal change in children who had botulinum toxin injections whose mobility status was not expected to change.
   iii. Correlations between change scores on the FMS with the FAQ were low. This may, however, reflect the properties of the criterion chosen rather than the responsiveness of the FMS itself.
   iv. Some evidence exists of the ability of the FMS to detect clinically important change. Further investigations are required to fully establish the responsiveness of the FMS.

4. A study of construct validity examining whether the information obtained from the FMS via self-report of the assistance required in the different settings of home, school and community concurs with the direct observation of mobility in children with CP (Chapter Seven). It showed:
   i. Fair to moderate agreement between self-report and direct observation for each distance of the FMS in a group of children attending a school for children with physical and cognitive disabilities.
   ii. Variability in the number and type of mobility method used within different environmental settings. The group showed variability and individual children often used more than one method between settings.
   iii. The variability in mobility methods observed may explain the differences between observations and self-report.
iv. Small numbers and methodological limitations of this study highlighted the need for further research into this clinically important issue of whether self-report tools measure performance.

5. A study examined the concurrent validity of the FMS by correlating it with the FAQ and the GMFCS and the discriminative validity of the FMS by discriminating severity of motor impairment in children with CP (Chapter Eight). It revealed:
   i. Concurrent validity was supported by high correlations of the FMS with the FAQ and the GMFCS.
   ii. The FMS was able to discriminate between children who walk independently from those who require assistance based on their GMFCS levels.

These conclusions demonstrate that the psychometric properties of the FMS have been extensively examined in this thesis. Validation is an ongoing process for any outcome measure. Complex multi-dimensional measures, such as the GMFM and PEDI, require more extensive examination over longer periods than simple scales, like the FMS, before they can be accepted into widespread clinical practice. The FMS is a simple scale with high face validity and obvious clinical applicability. Evidence of this is in its growing widespread use nationally and internationally including in the United States of America, Canada, India, the Netherlands, Sweden, Egypt, South America and the United Kingdom. It has also been translated into five languages. The collective results of the studies in Chapters Five to Eight demonstrate sufficiently robust psychometric properties of the FMS for widespread clinical implementation. The thesis has thus established an adequate level of validation for this simple and clinically applicable scale to become incorporated into routine clinical practice.
Finally, to conclude on a more philosophical note with a conversation between Alice and the Cat, from Alice’s Adventures in Wonderland, (Lewis Carroll 1865):

“Would you tell me, please, which way I ought to go from here?”

“That depends a good deal on where you want to get to” said the Cat.

“I don’t much care where..” said Alice.

“Then it doesn’t matter which way you go” said the Cat

...so long as I get somewhere!” Alice added as an explanation

“Oh, you’re sure to do that” said the Cat, “if only you walk long enough”.
REFERENCES


References


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The Functional Mobility Scale (FMS)

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<th>Walking distance</th>
<th>Rating: select the number (from 1–6) which best describes current function</th>
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<tbody>
<tr>
<td>5 metres (yards)</td>
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<tr>
<td>50 metres (yards)</td>
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<tr>
<td>500 metres (yards)</td>
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Rating: 

Independent on all surfaces:
Does not use any walking aids or need any help from another person when walking over all surfaces including uneven ground, curbs etc. and in a crowded environment.

Uses crutches:
Without help from another person.

Independent on level surfaces:
Does not use walking aids or need help from another person.* Requires a rail for stairs.
* If uses furniture, walls, fences, step stools for support, please use 4 as the appropriate description.

Uses a walker or frame:
Without help from another person.

Uses sticks (one or two):
Without help from another person.

Uses wheelchair:
May stand for transfers, may do some stepping supported by another person or using a walker/frame.

Crawling:
Child crawls for mobility at home (5m).

N = does not apply:
For example child does not complete the distance (500 m).

Please see over page for instructions
Appendices

Introduction

The Functional Mobility Scale (FMS) has been constructed to classify functional mobility in children, taking into account the range of assistive devices a child might use. The scale can be used to classify children’s functional mobility, document change over time in the same child and to document change seen following interventions, such as orthopaedic surgery or selective dorsal rhizotomy.

The FMS rates walking ability at three specific distances; 5, 50 and 500 metres. This represents the child’s mobility in the home, at school and in the community setting. It therefore accounts for different assistive devices used by the same child in different environments. Assessment is by the clinician on the basis of questions asked of the child/parent (not direct observation). The walking ability of the child is rated at each of the three distances according to the need for assistive devices such as crutches, walkers or wheelchair. Orthotics which are regularly used should be included for the rating.

The FMS is a performance measure. It is important to rate what the child actually does at this point in time, not what they can do or used to be able to do.

Questions

To obtain the answers that reflect performance, the manner in which the questions are asked of the child/parent is important. The questions we use to obtain the appropriate responses are:

1. How does your child move around for short distances in the house? (5m)
2. How does your child move around in and between classes at school? (50m)
3. How does your child move around for long distances such as at the shopping centre? (500m)

The distances are a guide. It is the environment that is most important.

Qualifiers

The difference between 1-4 is self-explanatory, however the difference between 5 and 6 is less clear.

5 metres: children who require a rail for stairs would be rated as 5 and children who do not require a rail or help would be rated as 6

50 metres: children who can walk on all surfaces including uneven surfaces and steps, particularly at school are rated as 6 and children that require help on these surfaces but can walk on level surfaces without help are rated as 5.

500 metres: children who can walk on all surfaces including rough ground, curbs, steps and in crowded environments in the community without help are rated as 6 and children who walk long distances only on level surfaces and have difficulty walking in crowds are rated as 5.
APPENDIX B

Gross Motor Function Classification System for Cerebral Palsy

Robert Palisano, Peter Rosenbaum, Stephen Walter, Dianne Russell, Ellen Wood, Barbara Galuppi

Introduction & User Instructions

The Gross Motor Function Classification System for cerebral palsy is based on self-initiated movement with particular emphasis on sitting (truncal control) and walking. When defining a 5 level Classification System, our primary criterion was that the distinctions in motor function between levels must be clinically meaningful. Distinctions between levels of motor function are based on functional limitations, the need for assistive technology, including mobility devices (such as walkers, crutches, and canes) and wheeled mobility, and to much lesser extent quality of movement. Level I includes children with neuromotor impairments whose functional limitations are less than what is typically associated with cerebral palsy, and children who have traditionally been diagnosed as having “minimal brain dysfunction” or “cerebral palsy of minimal severity”. The distinctions between Levels I and II therefore are not as pronounced as the distinctions between the other Levels, particularly for infants less than 2 years of age.

The focus is on determining which level best represents the child’s present abilities and limitations in motor function. Emphasis is on the child’s usual performance in home, school, and community settings. It is therefore important to classify on ordinary performance (not best capacity), and not to include judgments about prognosis. Remember the purpose is to classify a child’s present gross motor function, not to judge quality of movement or potential for improvement.

The descriptions of the 5 levels are broad and are not intended to describe all aspects of the function of individual children. For example, an infant with hemiplegia who is unable to crawl on hands and knees, but otherwise fits the description of Level I, would be classified in Level I. The scale is ordinal, with no intent that the distances between levels be considered equal or that children with cerebral palsy are equally distributed among the 5 levels. A summary of the distinctions between each pair of levels is provided to assist in determining the level that most closely resembles a child’s current gross motor function.

The title for each level represents the highest level of mobility that a child is expected to achieve between 6-12 years of age. We recognize that classification of motor function is dependent on age, especially during infancy and early childhood. For each level, therefore, separate descriptions are provided for children in several age bands. The functional abilities and limitations for each age interval are intended to serve as guidelines, are not comprehensive, and are not norms. Children below age 2 should be considered at their corrected age if they were premature.

An effort has been made to emphasize children’s function rather than their limitations. Thus as a general principle, the gross motor function of children who are able to perform the functions described in any particular level will probably be classified at or above that level; in contrast the gross motor functions of children who cannot perform the functions of a particular level will likely be classified below that level.
Appendices

Gross Motor Function Classification System for Cerebral Palsy (GMFCS)

Before 2nd Birthday
Level I  Infants move in and out of sitting and floor sit with both hands free to manipulate objects. Infants crawl on hands and knees, pull to stand and take steps holding on to furniture. Infants walk between 18 months and 2 years of age without the need for any assistive mobility device.
Level II Infants maintain floor sitting but may need to use their hands for support to maintain balance. Infants creep on their stomach or crawl on hands and knees. Infants may pull to stand and take steps holding on to furniture.
Level III Infants maintain floor sitting when the low back is supported. Infants roll and creep forward on their stomachs.
Level IV Infants have head control but trunk support is required for floor sitting. Infants can roll to supine and may roll to prone.
Level V Physical impairments limit voluntary control of movement. Infants are unable to maintain antigravity head and trunk postures in prone and sitting. Infants require adult assistance to roll.

Between 2nd and 4th Birthday
Level I  Children floor sit with both hands free to manipulate objects. Movements in and out of floor sitting and standing are performed without adult assistance. Children walk as the preferred method of mobility without the need for any assistive mobility device.
Level II Children floor sit but may have difficulty with balance when both hands are free to manipulate objects. Movements in and out of sitting are performed without adult assistance. Children pull to stand on a stable surface. Children crawl on hands and knees with a reciprocal pattern, cruise holding onto furniture and walk using an assistive mobility device as preferred methods of mobility.
Level III Children maintain floor sitting often by “W-sitting” (sitting between flexed and internally rotated hips and knees) and may require adult assistance to assume sitting. Children creep on their stomach or crawl on hands and knees (often without reciprocal leg movements) as their primary methods of self-mobility. Children may pull to stand on a stable surface and cruise short distances. Children may walk short distances indoors using an assistive mobility device and adult assistance for steering and turning.
Level IV Children floor sit when placed, but are unable to maintain alignment and balance without use of their hands for support. Children frequently require adaptive equipment for sitting and standing. Self-mobility for short distances (within a room) is achieved through rolling, creeping on stomach, or crawling on hands and knees without reciprocal leg movement.
Level V Physical impairments restrict voluntary control of movement and the ability to maintain antigravity head and trunk postures. All areas of motor function are limited. Functional limitations in sitting and standing are not fully compensated for through the use of adaptive equipment and assistive technology. At Level V, children have no means of independent mobility and are transported. Some children achieve self-mobility using a power wheelchair with extensive adaptations.

Between 4th and 6th Birthday
Level I  Children get into and out of, and sit in, a chair without the need for hand support. Children move from the floor and from chair sitting to standing without the need for objects for support. Children walk indoors and outdoors, and climb stairs. Emerging ability to run and jump.
Level II Children sit in a chair with both hands free to manipulate objects. Children move from the floor to standing and from chair sitting to standing but often require a stable surface to push or pull up on with their arms. Children walk without the need for any assistive mobility device indoors and for short distances on level surfaces outdoors. Children climb stairs holding onto a railing but are unable to run or jump.
Level III Children sit on a regular chair but may require pelvic or trunk support to maximize hand function. Children move in and out of chair sitting using a stable surface to push on or pull up with their arms. Children walk with an assistive mobility device on level surfaces and climb stairs with assistance from an adult. Children frequently are transported when travelling for long distances or outdoors on uneven terrain.
Level IV Children sit on a chair but need adaptive seating for trunk control and to maximize hand function. Children move in and out of chair sitting with assistance from an adult or a stable surface to push or pull up on with their arms. Children may at best walk short distances with a walker and adult supervision but have difficulty turning and maintaining balance on uneven surfaces. Children are transported in the community. Children may achieve self-mobility using a power wheelchair.
Appendices

Level V Physical impairments restrict voluntary control of movement and the ability to maintain antigravity
head and trunk postures. All areas of motor function are limited. Functional limitations in sitting and
standing are not fully compensated for through the use of adaptive equipment and assistive
technology. At Level V, children have no means of independent mobility and are transported. Some
children achieve self-mobility using a power wheelchair with extensive adaptations.

Between 6th and 12th Birthday
Level I Children walk indoors and outdoors, and climb stairs without limitations. Children perform gross
motor skills including running and jumping but speed, balance, and coordination are reduced.
Level II Children walk indoors and outdoors, and climb stairs holding onto a railing but experience limitations
walking on uneven surfaces and inclines, and walking in narrow or confined spaces. Children have
at best only minimal ability to perform gross motor skills such as running and jumping.
Level III Children walk indoors or outdoors on a level surface with an assistive mobility device. Children may
climb stairs holding onto a railing. Depending on upper limb function, children propel a wheelchair
manually or are transported when travelling for long distances or outdoors on uneven terrain.
Level IV Children may maintain levels of function achieved before age 6 or rely more on wheeled mobility at
home, school, and in the community. Children may achieve self-mobility using a power wheelchair.
Level V Physical impairments restrict voluntary control of movement and the ability to maintain antigravity
head and trunk postures. All areas of motor function are limited. Functional limitations in sitting and
standing are not fully compensated for through the use of adaptive equipment and assistive
technology. At Level V, children have no means of independent mobility and are transported. Some
children achieve self-mobility using a power wheelchair with extensive adaptations.

Distinctions Between Levels I and II
Compared with children in Level I, children in Level II have limitations in the ease of performing movement transitions; walking outdoors
and in the community; the need for assistive mobility devices when beginning to walk; quality of movement; and the ability to perform gross
motor skills such as running and jumping.

Distinctions Between Levels II and III
Differences are seen in the degree of achievement of functional mobility. Children in Level III need assistive mobility devices and frequently
orthoses to walk, while children in Level II do not require assistive mobility devices after age 4.

Distinctions Between Levels III and IV
Differences in sitting ability and mobility exist, even allowing for extensive use of assistive technology. Children in Level III sit
independently, have independent floor mobility, and walk with assistive mobility devices. Children in Level IV function in sitting (usually
supported) but independent mobility is very limited. Children in Level IV are more likely to be transported or use power mobility.

Distinctions Between Levels IV and V
Children in Level V lack independence even in basic antigravity postural control. Self-mobility is achieved only if the child can learn how to
operate an electrically powered wheelchair.

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Want to know more? Contact:

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Website: www.fhs.mcmaster.ca/canchild
**APPENDIX C**

**Systematic Review Data Extraction and Quality Assessment Form**

<table>
<thead>
<tr>
<th>Criteria</th>
<th>Description</th>
<th>Quality Assessment</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>1. Study aims</strong> (objective or goal may be stated instead).</td>
<td>Stated yes/no</td>
<td>Adequate/inadequate</td>
</tr>
<tr>
<td>Justified yes/no</td>
<td>Adequate/inadequate</td>
<td></td>
</tr>
<tr>
<td>Aim (write):</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Adequate/inadequate</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

| **2. Subject selection** | | |
| a) Sample size | Stated? Yes/no If yes N=  | Is the sample size adequate for the aim of the study? Yes/no/unclear |
| b) Population in this study | | |
| Is the population described? Yes/no. | | Is it possible to extract only CP from the data? Yes/no/unclear |
| If yes is it CP only or CP mixed with other conditions? Circle other. | | |
| ABI/SB/DD/disability/other (state if other) | | |
| Numbers of each: | | |
| c) Recruitment | Is the recruitment procedure described? Yes/no. | Is the recruitment procedure well-matched for the aims of the study? Yes/no/unclear |
| If yes, where were subjects recruited from? | | Comments (if required): |
| d) Sampling | Is the sampling procedure described? Yes/no | Is the sampling procedure well-matched to aims of the study? Yes/no/unclear |
| If yes circle. Convenience, consecutive, purposive, quota, random, systematic, other (state) | | |
### Appendices

#### d) Inclusion/exclusion criteria

(i) Inclusion list:

Is the inclusion list enough to satisfy the aims of the study? 
Yes/no/insufficient info

(ii) Exclusion list

Is the exclusion list enough to reduce the effects of confounding factors? Yes/no/insufficient info

Comments:

#### 3. Subject characteristics

<table>
<thead>
<tr>
<th>Subject ages</th>
<th>Mean, range</th>
<th>Adequate/inadequate</th>
</tr>
</thead>
<tbody>
<tr>
<td>Subject gender</td>
<td>Ratio</td>
<td>Adequate/inadequate</td>
</tr>
<tr>
<td>GMFCS levels (CP)</td>
<td>Levels</td>
<td>Adequate/partial/inadequate/not applicable</td>
</tr>
<tr>
<td>Topography of CP</td>
<td>Hemi/dipl/quad</td>
<td>Adequate/partial/inadequate/not applicable</td>
</tr>
</tbody>
</table>

Other (if not CP)

Comments (if required)

#### 4 Rater characteristics

If raters are used in this study, who are the raters?

Circle: therapists/parents/doctors/other clinicians/teachers Number of raters

Are the rater characteristics well-matched for the study design?

Yes/no/unclear Comments (if required)

#### 5. Measurement tools

List 2 key tools used in this study (if more than one)

<table>
<thead>
<tr>
<th>1.</th>
<th>2.</th>
</tr>
</thead>
<tbody>
<tr>
<td>Is the tool described in this study? Yes/no</td>
<td>Is the tool described in this study? Yes/no</td>
</tr>
<tr>
<td>If yes state (include whether tool is purely motor/physical function or this is a subsection)</td>
<td>If yes state (include whether tool is purely motor/physical function or this is a subsection)</td>
</tr>
</tbody>
</table>

Does the tool(s) listed appear to have face validity for the study aims?

1. Yes/no
2. Yes/no

Comments (if required):
### Appendices

| c) Target population for this tool | What is the group(s) of children this tool has been devised for? State:  
1. Diagnosis…………………………….  
2. Age group……………………………. | Is this well matched for the population used in this particular study? Yes/no |
| d) Environment | Does tool used incorporate different environments of home/school/community? Yes/ No/ Unclear | N/A Comments (if required): |
| e) Assistive devices | Does tool used distinguish different assistive devices Yes/No/Unclear | N/A Comments (if required): |
| f) Performance or Capability | Does tool measure performance or capability? Circle: Performance/Capability/Unclear | Is this adequately justified in light of the aims of the tool/study? Yes/No/Not applicable |
| g) Tool feasibility | (i) time taken  
If stated, time taken  
……………………………………....  
(ii) equipment required  
Stated/ not stated/unclear. If stated, list  
…………………………………………………….  
(iii) training required  
Stated/not stated/unclear. If stated, list:  
……………………………………………………. | (i) Is the time taken feasible in line with the goal of the tool? Feasible/not feasible/insufficient info  
(ii) Is this requirement feasible in line with the goal of the tool? Feasible/not feasible/insufficient info  
(iii) Is this requirement feasible in line with the goal of the tool? Feasible/not feasible/insufficient info |
| 6. Reliability | Circle which tested  
a) Interrater  
b) Retest/Intrarater  
c) Not tested | Statistical test used and results (write values)  
Is the result adequate to state the tool is reliable? a) Yes/no/insufficient information  
b) Yes/no/insufficient information  
Comments (if required): |
### Appendices

<table>
<thead>
<tr>
<th>7. <strong>Validity</strong></th>
<th><strong>Statistical test used and results</strong> (write values)</th>
<th><strong>Is the result adequate to state the tool is valid?</strong></th>
</tr>
</thead>
<tbody>
<tr>
<td><em>Circle which tested</em></td>
<td></td>
<td></td>
</tr>
<tr>
<td>a) Content</td>
<td></td>
<td>a) Yes/no/insufficient information</td>
</tr>
<tr>
<td>b) Concurrent</td>
<td></td>
<td>b) Yes/no/insufficient information</td>
</tr>
<tr>
<td>c) Construct</td>
<td></td>
<td>c) Yes/no/insufficient information</td>
</tr>
<tr>
<td>d) Discriminative</td>
<td></td>
<td>d) Yes/no/insufficient information</td>
</tr>
<tr>
<td>e) Not tested</td>
<td>Comments (if required):</td>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>8. <strong>Responsiveness to change</strong></th>
<th><strong>Statistical tests used and results</strong> (write values)</th>
<th><strong>Does the result show that the tool can detect important change over time adequately?</strong></th>
</tr>
</thead>
<tbody>
<tr>
<td>Measured? Yes/no</td>
<td></td>
<td>Yes/no/insufficient information</td>
</tr>
</tbody>
</table>

| 9. **Clinical utility** | Described? yes/no. If yes, state: | **Does the clinical utility stated justify its use in this population of children?**
| | | Justified/not justified/insufficient info |

| 10. **Statement of findings** | Is a clear statement of findings described? yes/no | **Is this statement of findings in line with results obtained?** Yes/No/insufficient info
| | Comments: | |

| 11. **Limitations** | Stated yes/no/incomplete | N/A |

| 12. **Main Conclusion** | Stated yes/no. If yes state: | |
APPENDIX D

Systematic Review: Reviewer Guidelines

1. Study aims - write study aim in space provided.
   - Adequate = the authors have stated the aim clearly and fully and have justified the aim of this study in the introduction
   - Inadequate = the aim is not stated clearly and not justified well

2. Subject selection
   Other populations:
   ABI: acquired brain injury, may also be stated as head injury (HI) or traumatic brain injury (TBI)
   SB: spina bifida, may also be stated as myelomeningocele
   DD: (developmental delay)
   Disability: some studies may put all children with differing diagnoses in together
   Sampling
   Convenience sample is one where subjects are chosen on basis of availability. Included within this is
   • Consecutive – all children who present within a set time interval
   • Purposive – subjects handpicked on the basis of specific criteria
   Random – random sampling procedure from a defined accessible population
   Systematic – from an ordered list of subjects a systematic selection method is used eg. every 10th subject
   Quota – guided sampling to ensure that each stratum is represented in the same proportion as the population

3. Subject characteristics.
   GMFCS = Gross Motor Function Classification System – a system of I to V used to classify severity of cerebral palsy
   Topography = distribution of cerebral palsy according to region of body – hemiplegia, diplegia and quadriplegia
   Both of these can be used for CP. If the study sample is not CP alone then this not relevant and other descriptions may be used
   • Adequate – all details of subject characteristics are provided
   • Inadequate – poor description of details – missing information – GMFCS or topography not stated and sample is CP
   • Partial – only some of the information provided eg. GMFCS levels stated generally but not numbers in each level
4. Rater characteristics
Be careful not to confuse using parents or therapists as raters with them being used in the routine administration of the tool/questionnaire (i.e. respondents)

5. Measurement tools.
GF - general function          PF - physical function          M - mobility   (use this for walking also)
QOL - quality of life          GMF - gross motor function
If more than one tool is used in the study and both are of interest, a separate form is required for each tool to assess psychometric properties.
Face validity is whether or not the tool appears to measure what it is supposed to
Performance can be described as what a child actually “does do” in the normal circumstances of everyday life.
Capability is what they “can do” ie. capable of doing in a defined clinical situation.

5. Reliability
Interrater reliability is the degree to which 2 or more raters can obtain the same ratings for a given variable
Retest/intrarater reliability is the extent to which the same results are obtained on repeated administration of the same scale when no change in physical function has occurred. Intrarater is looking at reliability of the same person and retest is looking at reliability of equipment
General guidelines for reliability values: reliability coefficient of <0.40 is poor, between 0.40 and 0.60 is fair or moderate, between 0.60 and 0.80 is good and >0.80 is very good or excellent.

6. Validity
Content validity is the extent to which items in an instrument adequately reflect the content domain being measured
Concurrent validity is the degree to which the outcomes of one test correlates with outcomes on another established criterion (“gold standard”) test when both tests given at the same time. *please write other tools correlated with in the statistics used*
Construct validity is the extent to which a theoretical construct is measured by an instrument
Discriminative validity is the ability of the tool to differentiate between groups that should differ eg. in this situation, can the tool differentiate severity of cerebral palsy

7. Responsiveness to change is the ability of an instrument to detect important change over time or after treatments
*For reliability, validity and responsiveness to change, where possible write results of statistical tests or a general statement if there are too many

8. Clinical utility is the applicability of the tool and the results of this study to the clinical situation and to clinical practice
APPENDIX E
The Gillette Functional Assessment Questionnaire

Please choose the answer below that best describes your child's usual or typical walking abilities. Circle one number which best describes the highest level of walking ability.

<table>
<thead>
<tr>
<th>Number</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Cannot take any steps at all</td>
</tr>
<tr>
<td>2</td>
<td>Can do some stepping on his/her own with the help of another person. Does not take full weight on feet; Does not walk on routine basis.</td>
</tr>
<tr>
<td>3</td>
<td>Walks for exercise in therapy and/or less than typical household distances.</td>
</tr>
<tr>
<td>4</td>
<td>Walks for household distances, but makes slow progress. Does not use walking at home as preferred mobility. (primarily walks in therapy or as exercise).</td>
</tr>
<tr>
<td>5</td>
<td>Walks for household distances routinely at home and/or school. Indoor walking only.</td>
</tr>
<tr>
<td>6</td>
<td>Walks more than 15-50 ft. outside the home but usually uses a wheelchair or stroller for community distances or in congested areas.</td>
</tr>
<tr>
<td>7</td>
<td>Walks outside for community distances, but only on level surfaces (cannot perform curbs, uneven terrain, or stairs without assistance of another person).</td>
</tr>
<tr>
<td>8</td>
<td>Walks outside the home for community distances, is able to get around on curbs and uneven terrain in addition to level surfaces, but usually requires minimal assistance or supervision for safety.</td>
</tr>
<tr>
<td>9</td>
<td>Walks outside the home for community distances, easily gets around on level ground, curbs, and uneven terrain, but has difficulty or requires minimal assistance or supervision with running, climbing and/or stairs.</td>
</tr>
<tr>
<td>10</td>
<td>Walks, runs, and climbs on level and uneven terrain and does stairs without difficulty or assistance.</td>
</tr>
</tbody>
</table>

Please tick all the things that your child is able to do, in addition to walking:

<table>
<thead>
<tr>
<th>Activity</th>
<th>Example</th>
</tr>
</thead>
<tbody>
<tr>
<td>Walk carrying an object</td>
<td>Jumps off a single step&lt;sup&gt;3&lt;/sup&gt;</td>
</tr>
<tr>
<td>Walk carrying a fragile object or glass of liquid</td>
<td>Hop on right foot&lt;sup&gt;4&lt;/sup&gt;</td>
</tr>
<tr>
<td>Walk up and down stairs using the railing</td>
<td>Hop on left foot&lt;sup&gt;4&lt;/sup&gt;</td>
</tr>
<tr>
<td>Walk up and down stairs without needing the railing</td>
<td>Step over an object, right foot first&lt;sup&gt;4&lt;/sup&gt;</td>
</tr>
<tr>
<td>Steps up and down curb independently</td>
<td>Step over an object, left foot first&lt;sup&gt;4&lt;/sup&gt;</td>
</tr>
<tr>
<td>Runs</td>
<td>Kick a ball with right foot&lt;sup&gt;4&lt;/sup&gt;</td>
</tr>
<tr>
<td>Runs well including around a corner with good control</td>
<td>Kick a ball with left foot&lt;sup&gt;4&lt;/sup&gt;</td>
</tr>
<tr>
<td>Can take steps backwards</td>
<td>Ride 2 wheel bike&lt;sup&gt;5&lt;/sup&gt;</td>
</tr>
<tr>
<td>Can manoeuvre in tight areas</td>
<td>Ride 3 wheel bike</td>
</tr>
<tr>
<td>Get on and off a bus by him/herself&lt;sup&gt;4&lt;/sup&gt;</td>
<td>Ice skate or roller skate&lt;sup&gt;6&lt;/sup&gt;</td>
</tr>
<tr>
<td>Skip rope&lt;sup&gt;2&lt;/sup&gt;</td>
<td>Ride an escalator, can step on/off without help</td>
</tr>
</tbody>
</table>

<sup>1</sup>Including first step without crawling. Use of railing permitted. <sup>2</sup>Consistently with successive jumps. Twirling rope by self or by other people <sup>3</sup>Without falling upon landing <sup>4</sup>Without holding on, without falling upon landing <sup>5</sup>Without training wheels <sup>6</sup>Without holding on to objects or another person
### APPENDIX F

**School Observation Checklist**

<table>
<thead>
<tr>
<th>Child’s name:</th>
<th>Date of observation:</th>
</tr>
</thead>
<tbody>
<tr>
<td>School:</td>
<td>Observer name:</td>
</tr>
<tr>
<td></td>
<td>GMGCS level:</td>
</tr>
</tbody>
</table>

Please tick most relevant response below

**How did the child above move:**

<table>
<thead>
<tr>
<th>How did the child move</th>
<th>wheelchair</th>
<th>walker</th>
<th>crutches</th>
<th>sticks</th>
<th>holds hands, or walls</th>
<th>independent (holds rails)</th>
<th>independent (no rails)</th>
<th>carried</th>
<th>crawls</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. from the car to classroom</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>2. within the classroom</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>3. from class to class</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>4. around playground</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Approximate: dimensions of classroom

- distance class to class
- distance playground

Comments:

- ...
- ...
- ...

- ...
- ...
- ...
APPENDIX G

Home Observation Checklist

Child’s name:                                                                 Date and time of observation:
Observer name:                                                                                                           

Please tick most relevant response below

How did the child above move:

<table>
<thead>
<tr>
<th></th>
<th>wheelchair</th>
<th>walker</th>
<th>crutches</th>
<th>sticks</th>
<th>holds hands, or walls</th>
<th>independent (holds rails)</th>
<th>independent (no rails)</th>
<th>carried</th>
<th>crawls</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. from room to room</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>2. within one room</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
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<td>3. outside the home</td>
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</table>

Approximate:
- dimensions of room ........................................................................................................................................
- distance room to room ....................................................................................................................................
- distance moved outside the house ..................................................................................................................
- pedometer ......................................................................................................................................................

Comments: .............................................................................................................................................................

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Appendices

APPENDIX H

Ethics Approvals

The Royal Children’s Hospital, Melbourne

ETHICS IN HUMAN RESEARCH COMMITTEE APPROVAL

<table>
<thead>
<tr>
<th>EHRC REF. No:</th>
<th>26086 A</th>
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<tbody>
<tr>
<td>PROJECT TITLE:</td>
<td>Mobility changes following single event multi level orthopaedic surgery in children with spastic diplegia.</td>
</tr>
<tr>
<td>Documents approved:</td>
<td>FMS brochure</td>
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<tr>
<td>Approved Protocol:</td>
<td>Protocol within Mod 1 dated 18 Apr 2006</td>
</tr>
<tr>
<td>INVESTIGATOR(S):</td>
<td>A Harvey, R Baker</td>
</tr>
<tr>
<td>DATE OF ORIGINAL APPROVAL:</td>
<td>5th July 2006</td>
</tr>
<tr>
<td>DURATION:</td>
<td>12 months</td>
</tr>
<tr>
<td>DATE OF APPROVAL EXPIRY:</td>
<td>5th July 2007</td>
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SIGNED: COMMITTEE REPRESENTATIVE

APPROVED SUBJECT TO THE FOLLOWING CONDITIONS:

ALL PROJECTS
1. Any proposed change in protocol, or any approved documents, or the addition of any documents (including flyers, brochures, advertising material etc) and the reasons for that change or addition, together with an indication of ethical implications (if any), must be submitted to the Ethics in Human Research Committee for Approval prior to implementation.
2. The Principal Investigator must notify the Secretary of the Ethics in Human Research Committee of:
   - Any adverse effects of the study on participants and steps taken to deal with them.
   - Any unforeseen events.
   - Investigators withdrawing from or joining the project.
3. A progress report must be submitted annually and at the conclusion of the project, with special emphasis on ethical matters. Please note: It is the responsibility of the investigator(s) to ensure that the RCH EHRC approval remains current for the entire duration of the project. Investigator's undertaking projects without current EHRC approval put at risk their indemnity, grant and publication rights.
4. All research information collected whilst individual participants are children, must be kept until the individual turns 25 (i.e. 7 years after their 18th birthday).

DRUG TRIALS
5. The investigator(s) must report to the Sponsor and the Ethics in Human Research Committee within 24 hours of becoming aware of any serious adverse event experienced by any subject during the trial.
6. The investigator(s) must ensure that all externally sponsored Clinical Drug Studies have insurance coverage that is current for the entirety of the study.
19 October 2006

Professor M Morris
Physiotherapy
The University of Melbourne

Dear Professor Morris

I am writing to advise you that this project has been registered at this University as approved at another institution:

Project title: Mobility changes following single event multi level orthopaedic surgery in children with spastic diplegia (EHRC 26086A)
Researchers: Professor M Morris, Ms A Harvey, Dr R J Baker
Ethics ID: 0606057

Please note that you will need to submit an annual report to the Human Research Ethics Committee at the end of each year, or at the conclusion of the project if it continues for less than this time. Requests for annual reports will be sent out via Themis.

Yours sincerely

Ms Kate Murphy
Executive Officer, Human Research Ethics
Phone: 83442073, Email: k.murphy@unimelb.edu.au

cc: HEAG Chair – Physiotherapy
Ms A Harvey

Melbourne Research and Innovation Office
The University of Melbourne Victoria 3010 Australia
T: +61 3 8344 2000  F: +61 3 9347 6739
<table>
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<tr>
<th>EHRC REF. No:</th>
<th>25118 A</th>
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<tr>
<td>Approved Protocol:</td>
<td>Protocol within mod 1 v1 dated 17 Nov 2005</td>
</tr>
<tr>
<td>INVESTIGATOR(S):</td>
<td>A Harvey, H Kerr Graham, R Baker</td>
</tr>
<tr>
<td>DATE OF ORIGINAL APPROVAL:</td>
<td>20 December 2005</td>
</tr>
<tr>
<td>DURATION:</td>
<td>24 months</td>
</tr>
<tr>
<td>DATE OF APPROVAL EXPIRY:</td>
<td>20 December 2007</td>
</tr>
<tr>
<td>SIGNED:</td>
<td>COMMITTEE REPRESENTATIVE</td>
</tr>
<tr>
<td>APPROVED SUBJECT TO THE FOLLOWING CONDITIONS:</td>
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**ALL PROJECTS**

1. Any proposed change in protocol, or any approved documents, or the addition of any documents (including flyers, brochures, advertising material etc) and the reasons for that change or addition, together with an indication of ethical implications (if any), must be submitted to the Ethics in Human Research Committee for Approval prior to implementation.

2. The Principal Investigator must notify the Secretary of the Ethics in Human Research Committee of:
   - Actual starting date of project.
   - Any adverse effects of the study on participants and steps taken to deal with them.
   - Any unforeseen events.
   - Investigators withdrawing from or joining the project.

3. A progress report must be submitted annually and at the conclusion of the project, with special emphasis on ethical matters. Please note: It is the responsibility of the investigator(s) to ensure that the RCH EHRC approval remains current for the entire duration of the project. Investigator’s undertaking projects without current EHRC approval put at risk their indemnity, grant and publication rights.

4. The investigator(s) must maintain all records relating to the study for a period of 23 years.

**DRUG TRIALS**

5. The investigator(s) must report to the Sponsor and the Ethics in Human Research Committee within 24 hours of becoming aware of any serious adverse event experienced by any subject during the trial.

6. The investigator(s) must ensure that all externally sponsored Clinical Drug Studies have insurance coverage that is current for the entirety of the study.
10 April 2006

Professor M Morris  
Department of Physiotherapy

Dear Professor M Morris

Re: The reliability and validity of the Functional Mobility Scale (FMS): a new performance measure of mobility for children with cerebral palsy EHRC 25118A

Principal Investigators Professor M Morris, Professor K Graham, Associate Professor R Baker & Ms A Harvey

HREC No. 060211

I am writing to advise you that the above project has been registered at this University as approved by another institution. Please take note of our HREC reference number above.

Please also note that you will need to submit an annual report to the Human Research Ethics Committee at the end of 2006. Requests for annual reports will be sent out in December 2006.

Yours sincerely

Kate Murphy  
Executive Officer  
Human Research Ethics

c-mail: k.murphy@unimelb.edu.au

c.c. Chair, DHEAG, Physiotherapy  
Ms A Harvey

Melbourne Research and Innovation Office  
The University of Melbourne Victoria 3010 Australia  
T: +61 3 9347 6739  
**ETHICS IN HUMAN RESEARCH COMMITTEE APPROVAL**

<table>
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<th>25119 A</th>
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<tr>
<td>PROJECT TITLE:</td>
<td>The responsiveness to change of the Functional Mobility Scale (FMS): a new performance measure of the mobility for children with cerebral palsy</td>
</tr>
<tr>
<td>Approved Protocol:</td>
<td>Protocol within Mod 1 v1 dated 17 Nov 2005</td>
</tr>
<tr>
<td>INVESTIGATOR(S):</td>
<td>A Harvey, H Kerr Graham, R Baker</td>
</tr>
<tr>
<td>DATE OF ORIGINAL APPROVAL:</td>
<td>20 December 2005</td>
</tr>
<tr>
<td>DURATION:</td>
<td>24 months</td>
</tr>
<tr>
<td>DATE OF APPROVAL EXPIRY:</td>
<td>20 December 2007</td>
</tr>
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</table>

**SIGNED:**

COMMITTEE REPRESENTATIVE

28/1/2005

**APPROVED SUBJECT TO THE FOLLOWING CONDITIONS:**

1. Any proposed change in protocol, or any approved documents, or the addition of any documents (including flyers, brochures, advertising material etc) and the reasons for that change or addition, together with an indication of ethical implications (if any), must be submitted to the Ethics in Human Research Committee for Approval prior to implementation.

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   - Investigators withdrawing from or joining the project.

3. A progress report must be submitted annually and at the conclusion of the project, with special emphasis on ethical matters.

   **Please note:** It is the responsibility of the investigator(s) to ensure that the RCH EHRC approval remains current for the entire duration of the project. Investigator's undertaking projects without current EHRC approval put at risk their indemnity, grant and publication rights.

4. The investigator(s) must maintain all records relating to the study for a period of 23 years.

**DRUG TRIALS**

5. The investigator(s) must report to the Sponsor and the Ethics in Human Research Committee within 24 hours of becoming aware of any serious adverse event experienced by any subject during the trial.

6. The investigator(s) must ensure that all externally sponsored Clinical Drug Studies have insurance coverage that is current for the entirety of the study.
10 April 2006

Professor M Morris
Department of Physiotherapy

Dear Professor M Morris

Re: The responsiveness to change of the Functional Mobility Scale (FMS): a new performance measure of the mobility for children with cerebral palsy EHRC 25119A

Principal Investigators Professor M Morris, Professor K Graham, Associate Professor R Baker & Ms A Harvey

HREC No. 060212

I am writing to advise you that the above project has been registered at this University as approved by another institution. Please take note of our HREC reference number above.

Please also note that you will need to submit an annual report to the Human Research Ethics Committee at the end of 2006. Requests for annual reports will be sent out in December 2006.

Yours sincerely

Kate Murphy
Executive Officer
Human Research Ethics

e-mail: k.murphy@unimelb.edu.au

cc. Chair, DHEAG, Physiotherapy
Ms A Harvey

Melbourne Research and Innovation Office
The University of Melbourne Victoria 3010 Australia
T: +61 3 8344 2000 F: +61 3 9347 6739
APPENDIX I

First author publications arising during candidature


Author/s:
HARVEY, ADRIENNE RUTH

Title:
The functional mobility scale for children with cerebral palsy: reliability and validity

Date:
2008-02

Citation:

Publication Status:
Unpublished

Persistent Link:
http://hdl.handle.net/11343/39403

File Description:
The Functional Mobility Scale for children with cerebral palsy: reliability and validity

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