From THE DEPARTMENT OF
WOMEN'S AND CHILDREN'S HEALTH
Karolinska Institutet, Stockholm, Sweden

MANUAL ABILITY CLASSIFICATION SYSTEM, (MACS): DEVELOPMENT, EVALUATION AND APPLICABILITY

Ann-Marie Öhrvall

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”Hur smått är allting som fått ett svar.
Det stora är det som står olöst kvar
när tanken svindlande stannat.”
Bo Bergman

ABSTRACT

The ability to use our hands affects how we carry out almost all daily activities. Children with cerebral palsy (CP) have varying degrees of difficulties using their hands, ranging from minor clumsiness to major problems with any voluntary movements. There has not been any standardized, reliable method available for describing how children with CP use their hands.

The aims of this thesis were to develop a system to classify hand function among children and adolescents with cerebral palsy, to evaluate the validity and reliability of the results, and to investigate whether the use of this classification system could increase knowledge about the ability of these children to use their hands, and how this correlates with self-sufficiency in daily activities.

Study I describes the development of the Manual Ability Classification System (MACS). The central concept "manual ability," is defined as the ability to handle objects in daily activities. The classification system consists of five levels. The MACS levels are based on the self-initiated ability of the children/adolescents to handle objects in their daily environment, i.e. when engaged in activities such as eating, dressing, playing, or doing schoolwork. The criteria for the different levels also include descriptions of the need for help or adaptations. The MACS is described in a brochure available on the Internet (www.macs.nu).

The validity of the MACS content and concept have been evaluated using different methods and from various perspectives in each of the four studies in the thesis. This was accomplished by interviewing parents of children with CP, occupational therapists and physical therapists, and other experts in the field (Studies I and II). In addition, the correlation between MACS and other instruments of hand function was examined, using a questionnaire to measure the degree of difficulty children have using their hands in daily activities (ABILHAND-Kids), as well as a test of manual dexterity (Box and Block Test) (Study III). MACS was also compared with a classification of gross motor function (Gross Motor Function Classification System, GMFCS) (Studies I and IV). The outcome of MACS was then compared with a measure of independence of performance of daily activities (Pediatric Evaluation of Disability Inventory, PEDI) (Study IV). The reliability between different observers (two therapists, or one therapist and a parent, respectively) was reported in Study I.

The overall results show that both parents and therapists found MACS to be a meaningful method of describing how children handle objects in daily life (Study I and II). The comparison between MACS and other instruments, ABILHAND-Kids and the Box and Block Test, showed a strong correlation ($r_s = -0.88$, $p < 0.05$ and $r_s = -0.81$, $p < 0.05$) among the different assessment methods, even though they describe different aspects of hand function. By linking the meaningful concepts of the instruments to categories of the International Classification of Functioning, Disability and Health (child and youth version) (ICF-CY), it was shown that the instruments cover various aspects of activity and participation within ICF-CY. MACS provided a significantly broader representation of activity and participation (linking to seven chapters) than the other two instruments (linking to two and one chapters, respectively) (Study III). A high correlation was also found between MACS and GMFCS ($r_s = 0.77$, $p < 0.05$) (Study IV). Nevertheless, only half of the children were classified into analogous levels of MACS and GMFCS indicating the complementary nature of the instruments. Interrater reliability for MACS was studied in two ways: in part by having two therapists classify 168 children, and in part by having both parent and therapists classify 25 children. In both situations, interrater reliability was excellent (intraclass correlation coefficient 0.98 and 0.96, respectively) (Study I).

Study IV investigated 195 children aged 3–15 years with different types of CP, using the PEDI functional skill scale for self-care and mobility. The results were compared to the children's MACS and GMFCS levels. Stepwise multiple regression analysis verified that MACS was the strongest predictor of self-care (66%), while the GMFCS was the strongest predictor of mobility (76%). Moreover, children in MACS levels I and II demonstrated an age-related increase of skills, achieving complete or almost complete self-sufficiency in self-care, albeit at a later point than children without disabilities. Children with more severely affected hand function, MACS levels III–V, did not achieve self-sufficiency, and no age-related increase of self-care skills was observed. A similar picture was seen with regard to mobility based on GMFCS levels.

In summary, the studies in this thesis show that MACS is a classification system that provides a valid and reliable functional description of manual ability in children and adolescents with CP.

Key-words: cerebral palsy, children, hand function, manual ability, classification,
Manual Ability Classification System (MACS).

SAMMANFATTNING PÅ SVENSKA

Hur vi kan använda våra händer påverkar hur vi utför en mängd olika aktiviteter. Barn med cerebral pares (CP) har svårt att använda sina händer i olika omfattning: hos en del syns bara en viss fumlighet, medan andra har stora svårigheter att utföra även enkla viljemässiga rörelser. Det har saknats ett standardiserat, tillförlitligt sätt att beskriva hur barn med CP använder sina händer.

Syftet med denna avhandling var att utveckla ett system för att klassificera handfunktion hos barn och ungdomar med cerebral pares, att utvärdera kvaliteten och tillförlitligheten av resultaten, och undersöka om användningen av denna klassificering kan öka kunskapen om barns och ungdomars förmåga att använda sina händer, och dess relation till självständighet i dagliga aktiviteter.

I studie I beskrivs utvecklingen av Manual Ability Classification System (MACS). Det bärande begreppet är ”manual ability”, som definierades som förmågan att hantera föremål i dagliga aktiviteter. MACS-klassifikationen består av fem nivåer. Nivåerna baseras på barns och ungdomars självinitierade förmåga att hantera föremål i sin vardagliga miljö, det vill säga när de till exempel äter, klär sig, leker eller utför skolarbete. I nivåkriterierna ingår även beskrivningar av behov av hjälp eller anpassningar. MACS beskrivs i en broschyr tillgänglig på webben (www.macs.nu).

Validiteten av MACS innehåll och begrepp har utvärderats med olika metoder och ur olika perspektiv i avhandlingens alla fyra delarbetena. Det har gjorts genom intervjuer med föräldrar till barn med CP, arbetsterapeuter och sjukgymnaster samt olika experter på området (studie I, II). Vidare har samband mellan MACS och andra mätinstrument för handfunktion undersökts, med hjälp av dels ett frågeformulär som mäter barns svårigheter att använda händerna i dagliga aktiviteter (ABILHAND-Kids), dels ett test som mäter handmotorisk snabbhet (Box & Block test) (studie III). MACS har också jämförts med en klassifikation av grovmotorisk förmåga (Gross Motor Function Classification System, GMFCS) (studie I,IV). MACS har också jämförts med ett mer omfattande instrument som mäter barns självständighet i dagliga aktiviteter (Pediatric Evaluation of Disability Inventory, PEDI) (studie IV). Reliabiliteten mellan olika bedömare (två terapeuter, respektive en terapeut och en förälder) rapporteras i studie I.

Resultaten visar att både föräldrar och terapeuter tyckte att MACS på ett meningsfullt sätt beskrev hur barn hanterar föremål i vardagen. De ansåg att det var lätt att välja MACS-nivå för barnen utifrån MACS nivåbeskrivningar och de förtydligande distinktionerna. Vidare visade jämförelsen mellan MACS och instrumenten ABILHAND-Kids och Box & Block test att det fanns en stark korrelation mellan de olika bedömningsmetoderna (r_s = -0,88, p < 0,05 och r_s = -0,81, p < 0,05), men att dessa ändå beskriver helt olika aspekter av handfunktion. Genom att länka instrumentens meningsfulla komponenter till kategorier i ICF-CY visade det sig att instrumenten omfattar olika aspekter av aktivitet och delaktighet inom ICF-CY. MACS visade sig innehålla en betydligt bredare representation av olika aspekter av aktivitet och delaktighet (länkning till sju kapitel) än de andra två instrumenten (länkning till två respektive ett kapitel) (studie III). Även mellan MACS och GMFCS fanns en hög korrelation (studie I: r_s = 0,79, studie IV: r_s = 0,77, $p < 0,05$). Trots detta klassificeras endast hälften av barnen till motsvarande nivåer inom MACS och GMFCS. Till exempel kan barn som klassats till MACS nivå III bedömas ha grovmotorisk förmåga motsvarande GMFCS nivå I-IV. Detta visar att de båda klassifikationerna beskriver olika dimensioner av hur barn fungerar i vardagen, och att båda behövs för att beskriva den funktionella förmågan hos barn med CP som ett viktigt komplement till information om diagnosen, subdiagnos och dominerande symtom.

Mellanbedömmarreliabilitet för MACS undersöktes på två sätt: dels genom att 168 barn klassificerades av två terapeuter, dels genom att 25 barn klassificerades av både föräldrar och terapeuter. I båda fallen var reliabiliteten mellan bedömarna mycket hög (Intra Class Correlation Coefficient 0,98 respektive 0,96) (studie I).

I studie IV undersöktes 195 barn med olika typer av CP, 3–15 år gamla, med PEDI–delskalorna funktionella färdigheter personlig vård och förflyttning. Resultaten relaterades till barnens MACS- och GMFCS-nivåer. Stegvis multipel regressionsanalys verifierade att MACS var den starkaste prediktorn av personlig vård (66 %) och att GMFCS var den starkaste prediktorn av förflyttning (76 %). Vidare sågs en tydlig åldersrelaterad utveckling av förmåga i personlig vård hos barn med MACS-nivå I och II, de blev helt eller nästan helt självständiga men senare än barn utan funktionsnedsättningar. Barn med sämre handfunktion, MACS-nivå III–V, nådde inte självständighet och ingen åldersrelaterad utveckling kunde ses. Liknande förhållanden sågs för förflyttningsförmåga utifrån GMFCS-nivå.

Sammanfattningsvis visar studierna i denna avhandling att MACS beskriver hur barn och ungdomar med CP kan hantera föremål i vardagen på ett funktionellt, giltigt och pålitligt sätt. MACS-nivåerna beskriver och särskiljer tydligt barnens och ungdomarnas olika grad av svårigheter, och de stämmer väl överens med andra mätinstrument av både handfunktion och utförande av dagliga aktiviteter. MACS kan användas i kliniskt arbete när terapeuten kan sätta rimliga och uppnåbara mål tillsammans med familjen, för att underlätta kommunikation mellan föräldrar, personal och myndigheter samt av forskare som vill beskriva barngrupper och relatera resultat till barnens förmåga att använda sina händer.

LIST OF PUBLICATIONS

This thesis is based on the following original papers, referred to in the text by their Roman numerals.

I. Eliasson A-C, Krumlinde-Sundholm L, Rösblad B, Beckung E, Arner M, Öhrvall A-M, Rosenbaum P. Parents' and therapists' perceptions of the content of the Manual Ability Classification System, MACS.
Developmental Medicine and Child Neurology 2006, 48: 549–554.

II. Öhrvall A-M, Eliasson A-C. Parents' and therapists' perceptions of the content of the Manual Ability Classification System, MACS. Scandinavian Journal of Occupational Therapy. 2010;17:209-216

III. Öhrvall A-M, Krumlinde-Sundholm L, Eliasson A-C. Exploration of the relationship between the Manual Ability Classification System and hand function measures of capacity and performance. (in manuscript)

IV. Öhrvall A-M, Eliasson A-C, Löwing K, Ödman P, Krumlinde-Sundholm L. Self-care and mobility skills in children with cerebral palsy, related to their manual ability and gross motor function classifications. Developmental Medicine and Child Neurology, 2010, DOI: 10.1111/j.1469-8749.2010.03764.

CONTENTS

LIST OF ABBREVIATIONS

ADL	Activities of Daily living
AHA	Assisting Hand Assessment
ANOVA	Analysis of Variance
BFMF	Bimanual Fine Motor Function
CFCS	Communication Function Classification System
CP	Cerebral Palsy
CV	Coefficient of Variation
GMFCS	Gross Motor Function Classification System
ICC	Intraclass Correlation Coefficient
ICF	International Classification of Functioning, Disability and Health
ICF-CY	International Classification of Functioning, Disability and Health: Children and Youth version
MACS	Manual Ability Classification System
QUEST	Quality of Upper Extremity Skills Test
PEDI	Pediatric Evaluation Disability Inventory
SCPE	Surveillance of Cerebral Palsy in Europe
VABS	Vineland Adaptive Behavior Scales
WHO	World Health Organization

1 INTRODUCTION

This thesis describes the creation, evaluation, and use of a classification system for quantifying ecological aspects of hand function for cerebral palsy (CP), the Manual Ability Classification System (MACS; (Eliasson et al., 2006b). MACS uses five levels to describe how children and adolescents 4–18 years of age, with cerebral palsy (CP), handle objects in daily life. The rational and underlying frames of reference and the process of development, including evaluation of validity and reliability, as well as the usefulness and applicability of MACS, will be discussed in this thesis and its included articles.

Classifications are useful to describe and to group characteristics, for example, of people within a heterogeneous diagnostic group, where subgroup differentiation is warranted because of varying presentations. Classifications do not include detailed descriptions, and they are not intended to measure or detect change. Instead, the purpose of a classification system is simply to describe common characteristics of a group that are thought to have meaning (validity).

The value of a useful classification could be exemplified by the Gross Motor Function Classification System (GMFCS), developed in 1997 (Palisano et al., 1997). The GMFCS describes the variety in gross motor functioning among children with CP in five levels, from level I, where the child walks without restrictions, to level V where self-mobility is severely limited, even with use of assistive technology ((Palisano et al., 1997, Palisano et al., 2008, Rosenbaum et al., 2008). Since its first publication, the GMFCS has been used extensively, both to describe a child's gross motor function in clinical practice and as a descriptive stratification system to explore the distribution of function, for example, in population-based registers of children with CP, as well as in research studies. The GMFCS has been shown to increase communication between professionals, as well as to assist in identifying best practices for children at different performance levels. The success of the GMFCS is reflected by its use in (probably) all research studies including children with CP since its introduction.

As the GMFCS became increasingly used, an equivalent system for manual function was requested, since it cannot be taken for granted that manual ability follows that of gross motor function in children with CP. On the contrary, we know that some children have more severe dysfunction affecting their lower limbs than their upper limbs, and vice versa. The possibility to group children with CP according to level of hand function could, together with information about the gross motor function level, give a fuller and more function-oriented view of the common use of different CP subgroup diagnoses. Thus, there was need for a function-oriented classification of hand function, an equivalent to the GMFCS.

A group of Swedish researchers, in collaboration with one of the developers of the GMFCS, took on the challenge of developing a system for classifying hand function. MACS (Eliasson et al., 2006b) classifies hand function based on how children use their hands to handle objects in daily life. Five levels are described, from level I, which comprises children who can handle objects easily and successfully, to level V, typically children who have difficulty performing even simple actions with the hands. It is the self-initiated manual ability and need of environmental adjustments, either assistance or adaptations to perform the activities, that are classified by MACS (Eliasson et al., 2006b). Distinctions between levels are described to enhance the determination of the most appropriate level for the child; see Table I. MACS describes the child's typical manual ability as used in different environments, like home, school, and community settings, not the maximal capacity in a test situation. The child's overall ability when handling objects in activities should be classified, rather than each hand being assessed and classified separately. When choosing the most appropriate MACS level for the child, the level should be selected based on the child's actual performance in daily life. Thus, this information must be obtained by asking parents, or someone who knows the child well, about how and which objects the child typically handles.

TABLE I. The structure of the Manual Ability Classification System, with five levels of description and distinctions between the levels (accessed at www.macs.nu/files/MACS_English_2010.pdf)

	Levels	Distinctions
I	**Handles objects easily and successfully** At most limitations in the ease of performing manual tasks requiring speed and accuracy. However, any limitations in manual abilities do not restrict independence in daily activities	**Between Levels I and II** Children in Level I may have limitations in handling very small, heavy or fragile objects which demand detailed fine motor control, or efficient coordination between hands. Limitations may also involve performance in new and unfamiliar situations. Children in Level II perform almost the same activities as children in Level I but the quality of performance is decreased, or the performance is slower. Functional differences between hands can limit effectiveness of performance. Children in Level II commonly try to simplify handling of objects, for example by using a surface for support instead of handling objects with both hands.
II	**Handles most objects, but with somewhat reduced quality and/or speed of achievement.** Certain activities may be avoided or be achieved with some difficulty; alternative ways of performance might be used but manual abilities do not usually restrict independence in daily activities.	**Between Levels II and III** Children in Level II handle most objects, although slowly or with reduced quality of performance. Children in Level III commonly need help to prepare the activity and/or require adjustments to be made to the environment since their ability to reach or handle objects is limited. They cannot perform certain activities and their degree of independence is related to the supportiveness of the environmental context.
III	**Handles objects with difficulty; needs help to prepare and/or modify activities.** The performance is slow and achieved with limited success regarding quality and quantity. Activities are performed independently if they have been set up or adapted.	**Between Levels III and IV** Children in Level III can perform selected activities if the situation is prearranged and if they get supervision and plenty of time. Children in Level IV need continuous help during the activity and can at best participate meaningfully in only parts of an activity.
IV	**Handles a limited selection of easily managed objects in adapted situations.** Performs parts of activities with effort and with limited success. Requires continuous support and assistance and/or adapted equipment, for even partial achievement of the activity.	**Between Levels IV and V** Children in Level IV perform part of an activity, however, they need help continuously. Children in Level V might at best participate with a simple movement in special situations, e.g. by pushing a simple button or occasionally hold undemanding objects.
V	**Does not handle objects and has severely limited ability to perform even simple actions.** Requires total assistance.	

The decision should be based on handling of age-appropriate objects commonly used in daily activities such as playing, eating, dressing, drawing, or writing. Hand function is complex and is influenced by many different components; the focus in MACS is on how activities are done, rather than why the children cannot perform them.

1.1 FRAMES OF REFERENCE

In the following sections I will describe the frames of reference that underpin MACS. The theory of occupational performance describes manual ability based on the interactional dynamic interplay between different components affecting performance. The development of MACS has been influenced by the activity and participation components of the International Classification of Functioning, Disability and Health (ICF). It is however, important to note that MACS does not describe underlying factors such as the body function components of the ICF framework.

1.1.1 Occupational performance

To be able to do different kinds of activities is central in a person's life. People are engaged in doing tasks almost constantly during the day, some activities that they must do and some that they want to do. The *doing* is a central issue in occupational therapy, and to be engaged in occupations makes up life (Kielhofner, 2008, Wilcock, 1999). Although *doing* is commonplace, it's not at all clear how it should be defined and described. Kielhofner (2008) divides *doing* into three levels: occupational skills, occupational performance, and occupational participation. Skills are defined as the smallest observable, goal-directed actions needed to complete an everyday activity (Fischer, 2009, Kielhofner, 2008). Examples of skills are reaching, grasping and releasing. Occupational performance is defined as the actual *doing* or performance of specific daily life activities, such as eating, dressing, and toileting. Occupational participation refers to engaging in play or activities in daily living that are part of one's sociocultural context, desired and/or necessary to one's well-being (Kielhofner, 2008).

Christiansen points out that we become who we are by what we do, that participation in activities affects our identity. To feel competent in carrying out activities provides self-esteem (Christiansen, 1999). A dynamic interaction between the person's characteristics, the occupation performed, and the environment form the occupational performance (Kielhofner, 2008, Law et al., 1996) The Person-Environment-Occupation (PEO) model (Law et al., 1996) can be used when identifying factors in the person, the task, and the environment that facilitate or hinder the performance of occupations. Skilled performance is achieved when there is a balance between the person's capabilities, the task requirements, and the support of the environment. The PEO model has a dynamic systems theory approach and can be used when describing the child's manual ability. In the MACS classification, the underlying reasons for a manual dysfunction (e.g., sensory, motor, or cognitive impairments) are not directly considered; instead, personal, environmental, and occupational factors are all embedded in the descriptions of different levels of manual ability, and the focus is on how the individual actually uses his/her hands when handling objects in daily life. Thus, MACS classifies occupational performance aspects of hand function.

1.1.2 International Classification of Functioning, Disability and Health (ICF)

A person's daily activity can be described by the dynamic relationship between the person's functioning, the disability, and the contextual factors, by using the International Classification of Functioning, Disability and Health as a framework (World Health Organization:(WHO, 2001a)). ICF describes functioning in relation to health as both a framework and taxonomy of health and disability. ICF comprises two parts, where part one covers the functioning and disability, including body functions and structures, activity and participation, and part two covers contextual factors, including environmental factors and personal factors (Figure 1). Body functions are defined as physiological functions of body systems, body structures refer to anatomical parts of the body, and impairments are problems in this component. Activity is defined as the execution of a task or action by an individual, and participation as involvement in a life situation. Difficulties in these components are described as activity limitations and participation restrictions. Although activity and participation are defined as separate terms, they are listed jointly in the classification. Environmental factors include the physical, social, and attitudinal environments that can have an impact by either facilitating or hindering a person's functioning. Personal factors form the background of an individual's life and living. Each component, except the personal factors, is further divided into chapters with categories, the units of the classification, arranged in a stem/branch/leaf scheme (WHO, 2001a).

A recently expanded version of ICF is now available, the International Classification of Functioning, Disability and Health, Children and Youth version (ICF-CY) with details capturing developmental aspects of functioning and focus on learning and environmental factors with a child perspective (WHO, 2007). ICF-CY provides a common conceptual approach and congruent terminology that offers the possibility to improve communication between different users, such as clinicians, researchers, and politicians (Lollar and Simeonsson, 2005). The conceptual framework of ICF-CY can be used in discussion with parents and children to enhance their understanding of different interventions and active participation in decision-making (Darrah, 2008, Rosenbaum and Stewart, 2004). ICF-CY can also be used to evaluate the content in different outcome assessments, by linking the different concepts in the assessments to ICF codes (Cieza et al., 2002, Cieza et al., 2005). This improves clinicians ability to choose outcome measures that measure the component that has been the target for the intervention.(Cieza et al., 2005, Krumlinde-Sundholm, 2008). The ICF linking procedure is used in Study III of this thesis.

In the components of activity and participation two qualifiers are used: capacity and performance. Capacity describes an individual's ability to execute a task or an action at the highest probable level of functioning that a person may reach in a standardized environment, as when tested in an arranged clinical setting. Performance describes what an individual does in his or her current environment. Capacity refers to what the individual can do at his/her best, and performance describes how the activities usually are performed (Lollar and Simeonsson, 2005, WHO, 2007). In the MACS classification the aspects of manual ability described are found within the activity and participation component of the ICF-CY, and in particular, the aspect of performance has a central role.

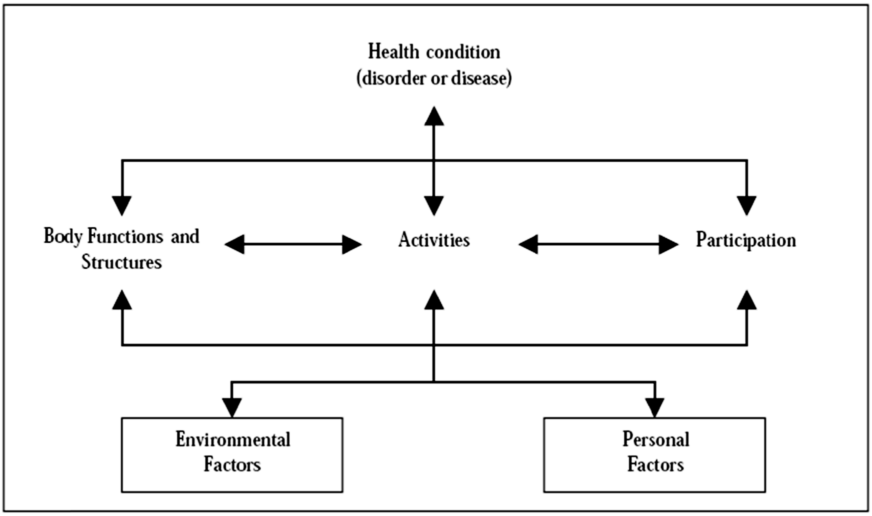

Figure. 1. The conceptual framework of the International Classification of Functioning, Disability and Health ((WHO, 2001b), accessed at www.who.int/classifications/icf/en/).

1.2 CEREBRAL PALSY

Cerebral Palsy is the most common cause of neurodisability in childhood. It is an umbrella term for different injuries in the immature brain.

1.2.1 Definition

A new definition of CP was suggested at a consensus meeting in 2006 with authorities in the field (Supplement, DMCN (2007). It describes CP "as a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to nonprogressive 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 behavior, by epilepsy, and by secondary musculoskeletal problems." Using the framework of the ICF (WHO, 2001a) this definition of CP describes the impairment on the level of body function and structure and its consequences in the components of activity and participation. Since this new definition covers individuals with a broad range of clinical presentation and activity limitations, it is proposed to further use subdiagnoses and functional classes to categorize them (Rosenbaum et al., 2007).

1.2.2 Classifications of CP

The heterogeneous group of children with CP can be subdivided according to the classification described in the reference and Training Manual of the Surveillance of Cerebral Palsy in Europe (SCPE) and is in accordance with the classification described by Rosenbaum et al. (2007). The dominant type of neuromotor abnormality should be classified as spastic, dyskinetic, or ataxic. The spastic type is subdivided according to the distribution of the clinical presentation, as bilateral when both sides of the body are involved, and as unilateral when one side is involved ((SCPE), 2002). Spastic unilateral CP was earlier described as hemiplegia, and spastic bilateral type was earlier described as diplegia and tetraplegia/quadriplegia, depending on the severity (Hagberg et al., 2001). The definitions of diplegia and tetraplegia/quadriplegia varied between countries, which made it impossible to compare children in different studies (Rosenbaum et al., 2007).

However, the functional consequences of the motor disorder are not described by the dominant type of muscle tone or movement abnormality and need to be described by separate classifications to give a fuller picture of the child's functional abilities. For that purpose, the Gross Motor Function Classification System is recommended to describe gross motor function, and for hand function, for example, the Manual Ability Classification System was suggested to be used (Rosenbaum et al., 2007).

To date there are three classifications available for children with CP, with the focus on functioning in daily life and with evidence of validity and reliability. They describe children's functional abilities and are developed to cover levels suitable for the whole spectra of CP, from very mild to very severe disabilities. The first developed was the Gross Motor Function Classification System describes gross motor function in five levels and with distinctions between the levels to enhance the rating. For hand function, the Manual Ability Classification System was developed with the same structure as GMFCS (Eliasson et al., 2006b). A new classification of communication has also recently been published, the Communication Function Classification System (CFCS); it has the same structure as the other two (Hidecker et al., 2011). The combination of these three classifications describes the children's functional performance and is an important complement to the diagnosis of CP (Chiarello et al., 2011, Hidecker et al., 2011, Rosenbaum et al., 2008). The latter classification system has just recently been published, and was therefore not used in the studies of this thesis.

1.2.3 Prevalence and aetiology

In Sweden, the prevalence of CP is reported to be 2.16/1000 live births. These figures are based on the National Cerebral Palsy Follow-Up Program (CPUP) Database in Sweden 2010, comprising 1307 children born 2000–2005 (CPUP; (2011)). The distribution of subdiagnoses showed that unilateral spastic CP was seen in 35% of the children, bilateral spastic CP in 43%, dyskinetic CP in 14%, and ataxia in 6% of the 885 children who had an established subdiagnosis. Gross motor function was classified in 1277 children, and the distribution found was that 45% of the children were classified as GMFCS level I, 15% as level II, 9% as level III, 15% as level IV, and 16% as level V (Figure 2).

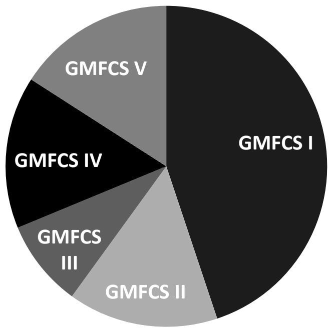

Figure 2. Distribution in different GMFCS levels

Manual ability was classified in 1220 children, showing about one-third with high ability to handle objects (31% at MACS level I) and 24% with somewhat lower ability (MACS level II), see Figure 3. More reduced ability to handle objects was seen for 15% who were classified as MACS level III, 14% as MACS IV, and 16% as MACS V.

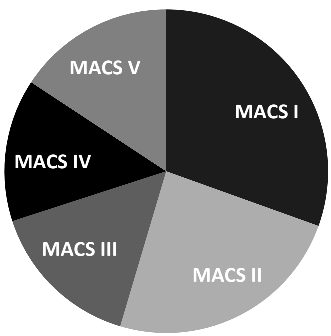

Figure 3. Distribution in different MACS levels

As a comparison, the prevalence of CP among children born in Europe was about the same: 2.08/1000 live births ((SCPE), 2002).

Today it is established that multiple factors, often in combination, can affect the developing brain and cause disturbances that result in a diagnosis of CP. Intrauterine infection, inflammation, multiple pregnancy, genetic factors, ischemia, and hypoxia are example of such factors. Neuroimaging techniques can be used to detect brain abnormalities in about 80% of the children diagnosed with CP (Krageloh-Mann and Cans, 2009). In bilateral CP the most common abnormality is periventricular white matter lesions (60%), and this is more common in children born prematurely than in children born at term. In unilateral CP periventricular white matter lesions account for around 35% of the cases and cortical and deep grey matter lesions account for another 35% of the cases. In children with unilateral CP are periventricular white matter lesions more common in preterm children while cortical and deep grey matter lesions are more common in children born at term. In dyskinetic CP the most common cause is cortical and deep grey matter lesions (54%)(Krageloh-Mann and Cans, 2009).

A recently published population-based study from western Sweden describes gross motor function (GMFCS level) in relation to neuroimaging findings in 160 children born 1999–2002, (Himmelmann and Uvebrant, 2011). GMFCS and MACS were used as functional classifications; however, MACS results were not presented in relation to the neuroimaging findings. The most common finding was white matter lesion, and 76% of the children with these lesions had a mild motor impairment classified as GMFCS level I or II. Two-thirds of the children with basal ganglia lesions and maldevelopment had severe limitations in gross motor function classified as GMFCS level IV or V, respectively. Children with cortical/subcortical lesion were classified as either level I or level V; most of them were born at term. Accompanying impairments, epilepsy, visual impairment, and learning disability were more common in children with cortical/subcortical and maldevelopment lesions than in children with periventricular white matter lesions.

1.3 OCCUPATIONAL DEVELOPMENT

Children's primary occupation is play. The child's occupation is supported by sensory, motor, cognitive, and social skills. Cultural, social, and physical contexts, as well as the child's age, highly influence the child's activity repertoire. Children's occupation arises through the interaction between the child and the environment, which offers the child opportunities for engagement in occupations (Case-Smith, 2010, Humphry, 2002). The culture and the family context are important for when and how the child learns everyday activities. Humpry (2002) argues that children are born with innate traits that guide them to cultural learning by imitating other people's purposeful occupations. The child's tendency to imitate others' behavior becomes a social and purposeful activity by observational learning before the end of the first year (Esseily et al., 2010, Humphry, 2002). Children also learn through play with other children and adults. Occupational development is also influenced by parental facilitation of an occupation, access to opportunities in the natural environment, and the child's motivation (Wieseman et al., 2005). The caregiver promotes the child by deciding which objects and people the child should interact with, and supports some activities while hindering others. Co-occupation is a part of the child's occupational development where the child and the care-giver are intimately involved in the creation of the occupation. The occupational performance is dependent of both persons interactional engagement (Price and Stephenson, 2009).

Self-care is, like play, an important area of children's occupation. The child practices many self-care activities during play, as well as by taking part in everyday occupations. During the pre-school years children develop independence in basic self-care activities. Learning of more complex tasks like tying shoes and doing or undoing buttons takes longer to master (Haley et al., 1992, Henderson, 2006, Henderson and Eliasson, 2008). The young child performs self-care as co-occupations of the caregiver and the child (Price and Stephenson, 2009, Shephard, 2010). Through co-occupation the young child develops a repertoire of occupations necessary for self-care development. Children with disabilities like CP use co-occupation at older ages than typically developed children, and some need to interact with the parent or caregiver in all activities throughout life (Shephard, 2010). Routines are important, and with them, the parent or caregiver helps the child to organize the activities of self-care. Cultural values and family patterns influence the routines. Frequently repeated practice is needed until performance becomes a habit and children are able to master more and more of the activity by themselves. In all play and self-care the handling of objects is important for mastering the different activities (Case-Smith, 2010, Dellatolas et al., 2005, Henderson and Eliasson, 2008, Pehoski, 2006, Shephard, 2010).

1.4 HAND USE

Hand use is essential for the performance of almost all daily activities; using the hands makes it possible to interact with and explore the environment. Although hands are often used in an automatic way, the ability to use the hands in an optimal way while performing activities is a skill that takes time to develop. In the following section, the complexity of hand use, and how the mastering of this complexity is developed, will be described.

1.4.1 Complexity of hand use

The ability to use the hands for skilled performance when handling objects in daily activities is complex. Skilled performance is dependent on many different components, both intrinsic to the child, such as cognition, perception, motivation, sensory-motor system, and muscles and skeletal system, as well as self-efficacy or external variables, such as physical environment, social environment like family dynamics, and cultural influences (Eliasson, 2005, Majnemer and Mazer, 2004). The ability to control the body in space, postural control, is important for the development of reaching and grasping during daily life activities (Shumway-Cook and Wollacott, 2001). Cognition is very important in hand function. The child must understand the point of using the hands in purposeful actions, and the ability to solve problems is important when performing and learning new tasks. Motivation is also crucial when learning new tasks and this is highly related to attention (Eliasson, 2005, Rao, 2006).

Handling objects in daily life is an interaction of the properties of the object, the nature of the task to be performed, and the object-related hand movements. Reach, grasp, and manipulation are important hand actions, which are used in different combinations during the tasks, depending on which object is to be handled. Smooth and precise handling of objects requires coordination and force control when grasping and lifting objects using a precision grip (Forssberg et al., 1991, Forssberg et al., 1992, Eliasson, 2006).

In each activity we commonly learn different ways to perform the activity, where innate handedness influences which hand we prefer to use in more skilled manipulation (Kimmerle et al., 2003, Fagard, 2006). For example, we always brush the teeth with the same hand and strike the match with one hand while holding the matchbox with the other. Thus, we give each hand a certain role in each activity. The hand roles are an important aspect of hand use in daily activities, and most activities are commonly performed bimanually (Henderson and Eliasson, 2008, Kimmerle et al., 2003, Krumlinde-Sundholm et al., 2007) . In bimanual handling of objects the two hands can have identical or different roles in different activities or part of them, depending on what the tasks require and the properties of the object. Examples of symmetrical hand use are catching a large ball or skipping rope. Asymmetrical use of the hand is seen when holding the object with one hand, while manipulating it with the other, for example, opening a jar, zipping-up a jacket, or slicing bread. Bimanual use of the hand requires skillful coordination between the hands and adjustment of the grip (Eliasson, 2006, Steenbergen and Gordon, 2006).

1.4.2 Development of hand use

The development of hand skills starts rapidly during the child's first year, but skilled hand use is not acquired until the early teenage period (Eliasson, 2005, Eliasson, 2006, Pehoski, 2006). Nagy and colleagues (Nagy et al., 2005) showed that newborn infants, immediately after birth, can imitate movements. Reaching towards a moving object was seen in infants from birth, and at about 5–6 months they were skillful at reaching for and grasping a moving object (Fagard et al., 2009, von Hofsten and Ronnqvist, 1988). The repertoire of activities performed starts to increase when the child learns to grasp at about 4 months of age. Following this, children learn to transfer objects between the hands, and bimanual manipulation is observed. During this type of manipulation, objects are explored with the fingers of one hand while the other hand holds the object. At the end of the first year children can manage to handle and manipulate several objects at the same time. Other important skills that developes during the latter part of the first year are the ability to use the pincer grip, and thereafter, the ability to release objects in a controlled manner (Charles, 2008, Pehoski, 2006). In the following years, children begin to manipulate objects within the hand, which involves independent finger movements. In-hand manipulation allows more efficient placement of the object in the hand, which is a prerequisite for effective manipulation of objects in daily activities (Exner, 1990, Exner, 1997, Exner, 2010, Pehoski, 2006, Pont et al., 2008). Children begin to master more complex in-hand manipulation during the preschool period, when they achieve more control of isolated finger movements and refinement of the force control during grasping.

The four-year-old child can handle a lot of different objects, for example writing with crayons, using the whole hand when grasping the crayon and both wrist and arm movements to excute and control the movement. The seven-year-old child in school uses a more mature grasp of the pen and can adjust performance during writing with small and precise finger movements. During this time they also improve their movement speed and accuracy when writing letters. In the teenage period the speed of movements increases while the variability decreases, and this is characteristic for older children's object manipulation. Older children are also able to skillfully adjust the grip size to the size of the object (Exner, 2010, Kimmerle et al., 2003, Pehoski, 2006)

1.4.3 Hand use in children with cerebral palsy

Children with CP have, to different degrees, limitations in using the hands when performing daily activities. Hand function in children with CP can vary from a slight clumsiness when handling very small or brittle objects to difficulties performing even simple movements, like playing at the computer by touching a big button. Hand function can be affected by increased muscle tone, muscle weakness, slow performance, insufficient coordination, and deformities of the joints (Arner et al., 2008, Uvebrant, 1988). Another factor affecting hand use is impaired sensibility, which is crucial for the regulation of the force of the grasp and in guiding the manipulation (Krumlinde-Sundholm and Eliasson, 2002, Pehoski, 2006). Children with CP have difficulties when grasping and their precision grip is affected by impaired grip-lift synergy (Forssberg et al., 1999) and difficulties with grading the grip force (Eliasson et al., 1995, Eliasson et al., 1992). This results in strategies of using more force than needed. Children with unilateral CP have also problems releasing objects because of problems in grading the velocity in the movement, resulting in an abrupt putting down of the object and prolonged and uncoordinated release of the grasp (Eliasson and Gordon, 2000). Most of the research about different aspects of hand function and treatment has been carried out on children with unilateral CP. In children with unilateral CP it is possible to compare the limited function in the disabled hand-arm with performance in the unimpaired or less impaired hand (Eliasson, 2005).

In a group of 367 children, aged 4–14years, high ability to use the hands with independence in handling age-relevant objects in daily life (MACS I–II) was seen in 87% of children with unilateral CP and in 63% of children with bilateral CP. In contrast was high ability only seen in 20% of the children with dyskinetic CP (Arner et al., 2008).

Children with spastic unilateral CP are often classified as MACS levels I and II, and only a few as level III (Arner et al., 2008). They have one affected hand and one well-functioning hand. In recent years some studies have shown that many children with unilateral CP also have some degree of motor involvement in the unaffected hand (Arnould et al., 2007, Dellatolas et al., 2005). The coordinated use of both hands together is affected. Often children with unilateral CP use the unaffected hand in most activities, and the affected hand in activities that demand two-hand use (Krumlinde-Sundholm et al., 2007, Skold et al., 2007). The pace in the affected hand is slower, influencing the bimanual hand use (Dellatolas et al., 2005). Hand use performance in the affected hand can vary from slightly reduced ability to only being ab le to use the hand as a support in activities. Although some children have very limited use of their affected hand they manage to be independent in most everyday activities with the help of alternative strategies.

Children with bilateral CP are distributed across all MACS levels, showing a big variation in the ability of these children to use their hands (Arner et al., 2008). They have involvement in both hands, which could vary from a slight difficulty handling small objects to no possibility of using the hands, at all (Arner et al., 2008). The children with bilateral CP show a slower performance. Some of them have also more involvement of one side of the body, which further affects the bimanual use of the hands (Dellatolas et al., 2005).

Children with dyskinetic CP often have severe limitations when performing activities, and most of them (71%) are classified as MACS level IV or V (Arner et al., 2008). On this level children need assistance during activity performance. Manual ability is influenced by involuntary fluctuating muscle tone, with difficulty holding and manipulating objects and performing goal-directed movements.

Children with ataxic CP are classified as MACS levels I to IV, showing a variety in ability to handle objects in daily life, from handling objects easily to only performing parts of the activities (Arner et al., 2008).

1.4.3.1 Development of hand skills in cerebral palsy

Knowledge about the natural history of development in hand function is limited in children with CP. Only a few studies are available, and they include almost exclusively children with spastic unilateral CP; as well, the samples are small. Fedrizzi and colleagues (Fedrizzi et al., 2003) followed spontaneous hand use and quality of grip in a group of 31 children with unilateral CP from 4 years of age until 11. Improvement in grip pattern was only shown with a weak and not significant trend, and the use of the hand was stable over time. Another study by Hanna and collaborators (Hanna et al., 2003) re-calculated data from a treatment study with 51 children aged 16–60 months at baseline: 20 children with spastic unilateral CP and 27 with spastic bilateral (quadriplegia) CP (Law et al., 1997). Growth curves were constructed from the repeated data collection (four times over 10 months). They showed development owing to severity of hand function measured by Peabody Developmental Motor Scales, fine motor skills (Folio and Fewell, 1983). Children with mild impairments show relatively good development in contrast to children with severe impairment, who show a negative trend at early age. When data were calculated from the Quality of Upper Extremity Skills Test (QUEST)(DeMatteo et al., 1992), an impairment-based measure of quality movements, the results showed minor improvements up to 2–4 years of age, according to severity.

In another study, development in higher ages was demonstrated by a follow-up with 10 children with 5 with unilateral and 5 with bilateral CP measured twice, 13 years apart (Eliasson et al., 2006a). The children were 6–8 years of age at the first data collection and at the second, 19–21 years of age. Manual dexterity was measured with the

Jebsen Taylor Hand Function Test (Jebsen et al., 1969, Taylor et al., 1973), and an evaluation of the precision grip in an experimental grip-lift task measuring fingertip forces was done. The result showed a decrease of 45% in time used for picking up the objects in Jebsen Taylor Test. The overall time for completion of the grip-lift task decreased 22% from the first to the second session, and also, reduced negative load force was demonstrated (less pushing down of the object before lifting) in the second session. This study showed a development to a more efficient grasping, with improvements in higher ages, irrespective of the initial severity of hand function, in contrast to earlier described studies where the severity predicted the development.

Holmefur and colleagues (Holmefur et al., 2010) showed in a longitudinal study how the use of the affected hand when performing bimanual tasks changed over time in children with unilateral CP over the period from 18 months to 7 years of age. The 43 children were divided into two groups according to their score on the Assisting Hand Assessment (AHA)(Krumlinde-Sundholm et al., 2007, Krumlinde-Sundholm and Eliasson, 2003)), one group showing a higher ability and one group a lower ability at the age of 18 months. In general, the children improved their scores over the time span. The group with a relatively higher score at 18 months of age improved more rapidly and reach a higher ability than the other group. This demonstrates that the AHA score at 18 months might be useful for prediction of development. The children with a lower score at 18 months typically did not spontaneously use the affected hand for grasping or holding at this age. At the age of 7 they had reach 90% of their average ability level. Children with a higher AHA score at 18 months could perform bimanual play, but with some difficulties. At the age of 3 they had reached 90% of their average ability level and could use the affected hand as a fairly useful assisting hand.

1.5 CLASSIFICATION SYSTEMS

1.5.1 Classification versus test and questionnaires

A classification is a system that makes it possible to group, divide, or arrange describing features into classes, according to common characteristics. Classifications have importance in everyday life, as well as in research situations, both for surveying a large quantity of facts/phenomena and for comprehending them. The usefulness of the classification depends on the principle of division, how understandable and clear the descriptions are and how meaningful different classes are (Krumlinde-Sundholm, 2008, Law et al., 2005). Classifications do not contain detailed descriptions and are thus not likely to detect change.

In habilitation services classifications are used in addition to tests and questionnaires, with different purposes. It is important to distinguish between these concepts. A test is based on a standardized situation and should be evaluated for accuracy of measurements in the specific target group. It is much more detailed than a questionnaire, and the procedure is typically described in a test manual. Usually a test needs to be administered by a professional who can interpret the results and findings, which are expected to be the basis for further treatment planning. A test should be sensitive for change and discriminate differences between abilities in a group of people. A questionnaire is a standardized instrument, with the aim to capture a person's own view of their performance, but the answers are based on the surveyed perceptions of the respondent's ability. All these types of instruments are important. They are not interchangeable, because they describe different perspectives and are used for different purposes.

1.5.2 Classification for the upper extremities

A commonly used classification for the upper extremity describes limitations in motor skills in children with CP on a 3-level scale: mild, moderate, and severe (Claeys et al., 1983). Mild dysfunction usually means that the child can use a pincer grasp, moderate that the child can use a whole-hand grip, and severe that the child in general has no possibility to grasp. Each hand can be assessed separately, but it is not clear where the distinctions are between the different levels. Moreover, this classification only takes into account an individual grasp, which is only one of many parameters that are important for the ability to use the hands in everyday activities. It is not standardized and the validity and reliability for the target group have not been evaluated.

Classifications for upper extremity in children with CP have two common purposes: one is grading the severity in function and the other is grading the deformity in position of a joint (McConnell et al., 2011). A classification that grades severity is the Bimanual Fine Motor Function (BFMF), classifying the function of each hand separately in five levels for children aged 5 to 8 years (Beckung and Hagberg, 2002). Evidence for validity and reliability has not been presented. The House functional scale and the Modified House functional scale are classifications with the purpose of evaluating hand function in the affected hand in children with unilateral CP and evaluating upper limb function, respectively (House et al., 1981, Koman et al., 2008). The grip in each hand is observed during activities and evaluated individually. Evidence of criterion-related validity is presented for the modified version of the scale, where relation to the Melbourne Assessment is investigated ($r = 0.84$). Excellent interrater and intrarater reliability, for children with both unilateral and bilateral spastic CP, were demonstrated in one study (Koman et al., 2008). and another study showed moderate intrarater and fair interrater reliability for children with unilateral CP (Waters et al., 2004).

There are classifications of deviating hand posture in children with CP: Zancolli classification (Zancolli and Zancolli, 1981), and House thumb classification (House et al., 1981). Zancolli classification evaluate the ability to extend the wrist and fingers; House thumb classifications classify thumb deformity in the affected hand. Reliability for both Zancolli and House are presented.

All classifications described above are commonly used in habilitation services. They evaluate each hand separately and classify what the child does when asked to perform in a clinical situation, and measure capacity according to ICF. They do not describe how the person uses the hands in daily activities. They do not use an occupational performance perspective, and the evidence of validity is very limited - just one classification has evidence for validity. The BFMF has to date not been extensively investigated with regards to validity and reliability. Classifications can be divided, based on how they are described and can be used, into two types. One type has general descriptions; these are used to divide a group into levels by described characteristics, such as GMFCS, MACS, BFMF, and CFCS. These classifications are supposed to be stable over time. The other type has descriptions of a very small area, for example, the position of the thumb, for example, House thumb classification. They are used to evaluate change in positions over time or after hand surgery, and are not supposed to be stable over time. There is a problem with the Modified House classification, which has different items to be done in a standardized way and scores added up on the basis of the performance. It thus meets the requirements of a test and is not a classification.

MACS fills a gap by adding a new perspective with a functional approach. There is a need to describe hand function in children with CP according to the manual ability in daily activities in a valid and reliable way, both in clinical practice and as a grouping variable in databases and in research, that is, a classification describing the child's performance, what the child can do, at the activity and participation level of ICF. Since CP is a heterogeneous group, it has been very difficult to study the typical development of hand function in this group of children. It has to date also been difficult to study the effect of treatment, since it has been impossible to compare groups of children because there was no commonly used classification across studies that could describe the children's manual ability.

1.6 DEVELOPMENT OF INSTRUMENTS

Development of an instrument, a test, or a classification is the process through which it is planned, constructed, evaluated, and modified (American Educational Research Association, 1999). The first step in developing an instrument is to clarify the construct and corresponding content. Furthermore it is important to evaluate the instruments validity and reliability related to the intended target group.

1.6.1 Validity

Validity is a fundamental concept in test development/evaluation. It refers to the degree to which evidence and theory support the interpretations of test scores entailed by proposed uses of the tests (American Educational Research Association, 1999). The validation of a new instrument is an ongoing process, and new instruments need to be validated from a number of different aspects (American Educational Research Association, 1999, Streiner and Norman, 2008). Validity has traditionally been categorized into three parts: content, criterion, and construct, where the criterion validity was divided into concurrent and predictive validity; see Table II. It has recently been recommended that the concept of validity should be changed and now be described under one overarching framework: construct validity (American Educational Research Association, 1999, Cook and Beckman, 2006, Goodwin, 2002, Streiner and Norman, 2008). This approach is based on the assumption that an instrument's scores are only useful if they reflect the underlying construct, and evidence should be collected to support this relationship. Different types of evidence should be collected and evaluated by the test developer. According to Standards (1999), five different types of validity evidence have been described (see Table II). A test/questionnaire is never valid in itself, but rather the interpretation of the test outcomes can be valid in relation to specific purposes. In this thesis we have collected evidence of validity concerning the MACS outcomes related to test content (Studies I and II), relation to other variables (Studies I, III, and IV), internal structure (Study I), and response processes (Study II) when using MACS.

Table.II.Comparison of validity concepts described in different editions of Standards (Goodwin, 2002)

1974	1985	1999
Content validity	Content-related evidence	**Evidence based on test content**
Criterion validity	Criterion-related evidence	**Evidence based on relation to other variables**
Construct validity	Construct-related evidence	**Evidence based on internal structure**
		Evidence based on response processes
		Evidence based on the consequences of testing

1.6.2 Reliability

Reliability refers to the degree to which test scores for a group are consistent over repeated applications of a measurement procedure (Standards, 1999). The stability of two occasions could be calculated from the same raters (intra-rater reliability), from different raters (interrater reliability), and from repeated test occasions (test-retest reliability). It commonly is reported as a reliability coefficient ranging from -1 to +1. It can be calculated by different methods, for example, intraclass correlation coefficient ICC (Shrout and Fleiss, 1979) and Cohen's weighted or unweighted kappa (Cohen, 1960, Cohen, 1968). The ICC and kappa are equivalent, but measurements are estimated in different ways. The interrater reliability was evaluated for MACS in Study I, both when two therapists classified the child and when a parent and a therapist classified the child.

2 AIMS OF THIS THESIS

The overall aims of this thesis were to develop a system to classify hand function among children and adolescents with cerebral palsy, to evaluate the validity and reliability of the results, and to investigate whether the use of this classification system could increase knowledge about the ability of these children to use their hands, and how this correlates with self-sufficiency in daily activities.

The specific aims of the four studies were:

Study1

To describe the development of MACS and provide evidence of its validity and reliability.

Study II

To investigate the content validity based on parents' and therapists' descriptions of the children's ability to use their hands in daily manual tasks and to relate that information to their choice of MACS level and the comprehension of the MACS concept.

Study III

To investigate the relationship between children's ability to handle objects in daily life, expressed through MACS, and hand function measures of capacity and performance, using ICF-CY as a framework to explore the uniqueness of the assessments.

Study IV

To investigate the acquisition of self-care and mobility skills in children with CP at different ages, in relation to their MACS and GMFCS levels.

3 MATERIAL AND METHODS

In this thesis the first study described the conceptual base and development of MACS and demonstrated the evidence for validity, based on content and reliability. The next two studies investigated validity from two different perspectives: Study II with a qualitative approach including interviews with parents and therapists, and Study III comparing the outcomes of MACS with results of other instruments measuring hand function. Study IV was a cross-sectional explorative study, where MACS and GMFCS levels were related to children's self-care and mobility skills (Table III).

TABLE III. Overview of the four studies

Characteristics of the studies	Study I	Study II	Study III	Study IV
Study design	Instrument development and evaluation, cross-sectional	Qualitative	Cross-sectional, content comparison using ICF-CY as frame of reference	Cross-sectional
Analysis	Descriptive, ICC	Content analysis	Descriptive, Spearman rank correlation, ANOVA, linear regression, CV linking to ICF-CY	Descriptive, Spearman rank correlation, Multiple regression analysis, ANOVA

ICC =Intraclass Correlation , ICF-CY =International Classification of Functioning, Disability and Health Children and Youth version, ANOVA =analysis of variance , CV =Coefficient of Variation

3.1 PARTICIPANTS

Study I

In the reliability part of the first study a total of 174 children with CP participated (Table IV). It consisted of two samples, one with 168 children between 4 and 18 years (98 boys, 70 girls) and one with 25 children. The children represent all levels of MACS and GMFCS, showing a broad representation of functional abilities in the study group. Out of the sample of 168 children, 148 were recruited from six cities in Sweden and 20 from one city in Australia. The group of 25 children was recruited in the Stockholm region in Sweden, and these children also participated in Study II. Nineteen out of the 25 children were also included in the sample with 168 children, thus six children were added, making the total number of children in the study 174.

Study II

Twenty-five children aged 8 –12 years (13 boys and 12 girls) with CP participated in this study. The children were represented by their parents and a therapist, either an occupational therapist or a physiotherapist. Caregivers, instead of the parents, represented three of the children, as these children were living in a children's residential care center. The children were selected using purposeful sampling (Creswell, 2000) covering a wide range of functional abilities. The group consisted of 3 children with spastic unilateral CP, 19 with spastic bilateral CP, and 3 with dyskinetic CP (Table IV).

Study III

A convenient sample of children with cerebral palsy was recruited by occupational therapists at local habilitation services mainly located in the area of Stockholm County. Ninety-one children from 5 to 17 years old, MACS levels I–V, participated in the study (Table IV). Attempts were made to cover a wide range of variation, both of functional abilities and of different subdiagnoses of CP.

TABLE IV. Characteristics of the participants

	Study I	Study II	Study III	Study IV
Number of participants	174	25	91	195
Age, range	4-18	8-12	5-17	3-15
Gender				
Boys	100	13	50	122
Girls	74	12	41	73
CP subdiagnosis				
Spastic unilateral	51	3	34	70
Spastic bilateral	94	19	45	100
Dyskinetic	19	3	8	16
Ataxia	6		4	5
Unspecified	2			
MACS				
I	25	5	14	55
II	65	7	37	66
III	28	5	20	33
IV	27	6	10	14
V	25	2	10	27
GMFCS				
I	47			90
II	35			32
III	28			29
IV	33			21
V	27			23

CP = cerebral palsy, MACS = Manual Ability Classification System , GMFCS = Gross Motor Function Classification System

Study IV

In this study 195 children with CP, aged between 3 and 15 years, participated (Table IV). All MACS and GMFCS levels were represented in the group. The sample was recruited in two ways. From a region-based health and quality follow-up register (HEFa), data from 116 of the children were included, and a convenient sample of 79 children was recruited by occupational therapists at the habilitation services in the county of Stockholm. The HEFa register includes children with CP from three county council areas in the south of Sweden: Kalmar, Östergötland, and Jönköping. Forty-five percent of the region's population of children with CP was included in the register at the time of data collection.

OVERALL PARTICIPATION IN STUDIES

There were some children who participated in more than one study. Table V shows how the number of children overlapped between studies. Only 3 children participated in all studies.

Table V. Number of participants overlapping between studies

Study I (n = 174)				
Study II (n = 25)	25			
Study III (n = 91)	3	9		
Study IV (n = 195)	3	3	36	
	Study I	**Study II**	**Study III**	**Study IV**

3.2 DATA COLLECTION AND METHODS

TABLE VI. Overview of included classifications and measurements.

Instruments	**Study I**	**Study II**	**Study III**	**Study IV**
MACS	X	X	X	X
GMFCS	X			X
ABILHAND-Kids			X	
Box and Block test			X	
PEDI				X
ICF-CY			X	

MACS =Manual Ability Classification System, GMFCS = Gross Motor Function Classification System, PEDI = Pediatric Evaluation of Disability Inventory, ICF-CY = International Classification of Functioning, Disability and Health Children and Youth version

Study I

PROCESSES OF DEVELOPMENT AND VALIDATION OF THE CLASSIFICATION

A research group with representatives from three universities in Sweden, in collaboration with McMaster University in Hamilton, Canada, was assembled. This expert group consisted of six researchers with long experience (minimum 20 years) in hand function, with different professional backgrounds and experience of research. The construct development of the classification was an ongoing process for more than 3 years, through workshops and conferences. The concept, the ability to handle objects in daily life, was based on a literature review, clinical experience, and analysis of videos in which children with different degrees of disability performed daily activities. The descriptions of the children's different abilities were defined in five levels. The descriptions were reviewed and changed by a continuous process, until consensus was reached in the expert group. When the first field version was available of the classification for children with CP, ages 8 to 12 years, the external validation process started, by presenting the classification to professionals and parents.

PROCESS OF TESTING THE RELIABILITY OF THE CLASSIFICATION

Data were collected by pairs of therapists classifying the same child, and by parents and therapists classifying the same child. The therapists and parents received a short introduction before reading the MACS leaflet, followed by the rating process, where they were asked to select the most appropriate MACS level to describe the child's ability to handle objects in daily activities. GMFCS levels were collected to describe the children's gross motor ability.

Study II

Data were collected through qualitative interviews with parents of children with CP and with therapists. The interviews took place on separate occasions, with a week or less between the two data collection sessions. First, a short introduction to MACS was given to the parent or the therapist; then, after reading the leaflet, they chose the MACS level that best represented the child's performance. Subsequently, a semi-structured interview was performed (Kvale, 1996). The focus of the questions was the respondent's thoughts about the classification. Questions were organized under the following themes:

(i) motivations for choosing one specific MACS level rather than another
(ii) examples of activities requiring manual ability, which the children liked to do
(iii) thoughts about the meaningfulness of using MACS to describe the children's hand function
(iv) opinions on the overall usefulness of MACS.

The interviews were tape-recorded.

Study III

MACS was compared with two other instruments measuring hand function, ABILHAND-Kids and the Box and Block Test (Arnould et al., 2004, Mathiowetz. et al., 1985). The instruments were selected to reflect different aspects of hand function: both the child's typical performance and the best capacity. The choice of instruments was based on the ICF's definition of performance and capacity (WHO, 2001a). Parents completed the ABILHAND-Kids questionnaire. ICF-CY was used as a reference framework in a content comparison of the instruments (WHO, 2007). The contents of each instrument were linked to the most appropriate ICF-CY code representing the categories of ICF-CY, by using standardized linking roles (Cieza et al., 2002, Cieza et al., 2005). The linking was done independently by two raters who knew ICF-CY well. The content comparison between MACS, ABILHAND-Kids, and the Box and Block Test was performed on the different levels of ICF-CY.

Study IV

Pediatric Evaluation of Disability Inventory (PEDI), MACS, and GMFCS data were collected by health professionals experienced in the use of the classifications and PEDI (Haley et al., 1992). The Swedish version of PEDI was used (Nordmark et al., 1999). The functional skill scales were used to assess self-care (73 items) and mobility (56 items).

3.3 CLASSIFICATIONS AND MEASURES

All classifications and measures included in this thesis will be presented in the following text.

3.3.1 Classifications

3.3.1.1 MACS—Manual Ability Classification System

MACS is a five-level classification system that describes how children with CP use their hands when handling objects in daily activities, on the basis of their self-initiated manual ability and need of environmental adjustments, either assistance or adaptations to perform the activities (Eliasson et al., 2006b). Accessed at www.macs.nu. See Table VII and Table I, under Introduction.

TABLE VII. General headings for each level of MACS (Eliasson et al., 2006)

MACS	**Manual Ability Classification System**
I	Handles objects easily and successfully
II	Handles most objects but with somewhat reduced quality and/or speed of achievement.
III	Handles objects with difficulty; needs help to prepare and/or modify activities.
IV	Handles a limited selection of easily managed objects in adapted situations.
V	Does not handle objects and has severely limited ability to perform even simple actions.

MACS describes the child’s manual ability in different environments, such as home, school, and community settings, not the maximal capacity in a test situation. The child’s overall ability when handling objects in activities should be classified, rather than assessing and classifying each hand separately. The most appropriate MACS level for the child should be determined by the child’s actual performance in daily life, and the selection of level must be made by asking the parents or someone who knows the child well. The decision should be based on the handling of objects commonly used at the child’s age, in daily activities such as playing, eating, dressing, drawing, or writing. The identification chart could guide the decision process (accessed at www.macs.nu/files/MACS_identification_chart_eng.pdf). Hand function is complex and is influenced by many different components. The focus in MACS is on how activities are done, rather than why the child cannot perform them.

3.3.1.2 GMFCS—Gross Motor Function Classification System – Expanded and Revised

The GMFCS describes the self-initiated gross motor function in five levels (Table VIII). Each level is described in five different age-bands: before 2 year of age, 2 to 4 years, 4 to 6 years, 6 to 12 years, and 12 to 18 years (Palisano et al., 2008). The levels capture the age-related characteristics of the child's gross motor function. The classification should be based on the child's typical performance in home, school, and community settings.

TABLE VIII. General headings for each level of GMFCS (Palisano et al., 2008)

GMFCS	Gross Motor Function Classification System
I	Walks without limitations
II	Walks with limitations
III	Walks using a hand-held mobility device
IV	Self-mobility with limitations; may use powered mobility
V	Transported in a manual wheelchair

3.3.1.3 ICF-CY—International Classification of Functioning, Disability and Health (children and youth version)

The International Classification of Functioning, Disability and Health (children and youth version) (ICF-CY) is a classification system for describing functional ability and disability in relation to health (WHO, 2007). With ICF as a conceptual model, it is possible to categorize different instruments and describe the functional level that different instruments measure, whether that is the body function level, or body structure, or activity, or participation. The linking rules (Cieza et al., 2002, Cieza et al., 2005) provide this opportunity to link and compare meaningful concepts in outcome measures by using ICF as a frame of reference. For a more detailed description, see Figure I under Introduction.

3.3.2 Measures

3.3.2.1 PEDI—Pediatric Evaluation of Disability Inventory

PEDI is a norm- and criterion-referenced measure that evaluates functional skills and caregiver assistance in the domains of self-care, mobility, and social function in children aged 6 months to 7.5 years (Haley et al., 1992). Environmental modifications are also rated for the different activities. It can be used for evaluating older children, if their functional abilities are less than expected for a 7.5-year-old child (Haley et al., 1992). The child is assessed by structured interviews with the parents. The functional skills scale measures whether the items can be performed or not in most situations (score 1 = child has capability). The summary scores of the functional skill scale and caregiver assistance scale in the three domains can be converted to normative standard scores and scaled scores. Normative scores are available for children 6 months to 7.5 years. The scaled scores range on a continuum from 0 to 100, where 0 represents no ability and 100 represents full capability to perform the functional skill items in a particular domain. PEDI is available in a Swedish version used in Study IV, with results from the functional skill scales self-care and mobility presented in scaled scores (Nordmark et al., 1999). The PEDI manual contains information about ages when typically developed children master different skills. They reach a scaled score of 100 at around 6 years of age for self-care and 4 years for mobility. The word "capable" in this study means capable of performing the activity in most situations, as defined in the PEDI administration manual (p. 86).

3.3.2.2 ABILHAND-Kids

ABILHAND-Kids is a questionnaire measuring manual ability in daily life activities, in children with CP (Arnould et al., 2004). A response scale in three levels - easy, difficult, and impossible - is used to measure the child's ability to independently manage 21 daily activities that require use of the upper limbs. Parents complete the ABILHAND-Kids questionnaire by estimating their child's ease or difficulty in performing the 21 activities independently. An online analysis (accessed at www.rehab-scales.org) gives the possibility to convert the child's raw score into a logit measure in a Rasch analysis, the interval level data ranged from -6.75 to 5.98 logits. The ABILHAND-Kids logits were used in Study III. ABILHAND-Kids reflects the child's typical performance (Arnould et al., 2007).

3.3.2.3 Box and Block Test

The Box and Block Test measures gross motor dexterity (Mathiowetz. et al., 1985). It consists of a box divided into two compartments by a 15 cm high partition, and a lot of 2.5 cm cubes. The box is positioned in front of the child with the blocks in the compartment near the dominant hand. The child is asked to transfer as many blocks as possible from one compartment to the other in 60 seconds, first with the dominant hand, then with the other. The total number of cubes transported with each hand is registered. In Study III the results for the left and right hands were used as mean values. The Box and Block Test is a standardized test that presents hand-function in the perspective of the best capacity.

3.4 DATA ANALYSIS

A variety of different statistical analyses were performed (Table III).

QUANTITATIVE ANALYSIS

The interrater reliability was analyzed by calculating the intraclass correlation coefficient (ICC), in a one-way random effects model. This is suitable when different raters assess the children using average measures, according to Shrout and Fleiss (1979). Total agreement was also estimated between therapists rating the same child, presented as a percentage (Polit and Hungler, 1999).

One-way analysis of variance (ANOVA) and post hoc test (Tuckey's) were used in Studies III and IV. Differences between mean values of ABILHAND-Kids and the Box and Block Test at the different MACS levels were investigated in Study III. The data were not normally distributed, but the residuals showed normal distribution. In Study IV evaluation of differences in mean scaled scores of PEDI self-care and mobility at the different MACS and GMFCS levels was estimated.

Regression analysis was performed in two studies. In Study III a linear regression analysis was performed in order to investigate how much of the result in ABILHAND-Kids and the Box and Block Test was explained by MACS level. In Study IV the influence of MACS and GMFCS, respectively, on self-care and mobility outcomes was estimated by a linear regression analysis. Multiple regression analyses (stepwise forward) were performed in the same study to evaluate how much MACS, GMFCS, and age together contributed to the result of self-care and mobility. Self-care and mobility scaled scores were used as dependent variables and were each compared with MACS, GMFCS, and age. MACS and GMFCS were treated as categorical predictors, and age as a continuous predictor. The normal plots of residuals were almost linear, for both models, endorsing the validity of the analysis.

The coefficient of variation (CV) was calculated for ABILHAND-Kids logits and Box and Block test scores to compare the variation in the material (Study IV). ABILHAND-Kids logits were transformed to a percentage scale 0–100, for use in this calculation.

Spearman's rank correlation was used to investigate the relationship between MACS and GMFCS in Studies I and IV, as well. The relation between MACS and ABILHAND-Kids, as well as between MACS and the Box and Block Test, were estimated by Spearman rank correlation in Study III. Non-parametric statistics were used, since MACS and GMFCS represent ordinal data, and the material was not normally distributed. P-values of <0.05 were considered statistically significant.

QUALITATIVE ANALYSIS

The qualitative data analysis in Study II was influenced by a content analysis method, directed content analysis, where the goal is to validate or extend conceptually a theoretical framework or a theory (Hsieh and Shannon, 2005).

The first author started by thoroughly listening to all the taped interviews. The data reduction was performed by transcribing the parts of the interviews that related to the four themes from the interview guide; then the data were separately analyzed for each theme. Data according to the *first theme*, *the explanations given for the choice of MACS levels*, were before the analyses divided into two categories: those whose raters agreed on the scoring and those whose raters did not. Analyses were then done separately. For those whose assessments of the *MACS level did agree*, the text was organized according to the different levels of MACS. The explanations of the children's performance given by parents and therapists showed no major differences and were used as one body of information. For those children whose *MACS levels did not agree,* the reasons were analyzed and described. The *second theme*, *the examples concerning the children's performance*, was used to deepen the understanding of the participants' decision to choose a specific level. Data were also used to identify other explanations not included in the leaflet but nevertheless important for decision-making. The findings from both groups, those whose assessments agreed and those whose did not, were compared with the description in the leaflet. The *third and fourth themes, concerning the meaningfulness and the usefulness of the MACS*, were organized separately for parents and therapists. The analyses were performed by the first author and were discussed continuously with the second author, until the data could be understood as a coherent and meaningful whole (Hsieh and Shannon, 2005, Kvale, 1996).

4 ETHICAL CONSIDERATIONS

All studies in the thesis were approved by the Ethics Research Committee at the Karolinska Hospital or the Regional Ethics Review Board at Karolinska Institutet in Stockholm, Sweden. The first study included data from other parts of Sweden and Australia and was approved by the regional ethics committees in Gothenburg, Umeå, and Lund, as well as in Melbourne, Australia.

Informed consent was obtained from the parents of the children.

5 RESULTS

Findings from the four studies will be presented together under the following themes: evidence of validity and reliability of MACS, and self-care and mobility skills in children with CP.

5.1 EVIDENCE OF VALIDITY

5.1.1 Development – the construct and the scale

An inventory of available tests and classifications of hand function was first constructed, which together with the researchers' experiences formed the basis of the discussion about the concept of the classification (Study I). Discussions continued until consensus was reached in the expert group. It was decided that the classification should highlight the ability to handle objects, have a functional perspective, and focus on children's ability to use their hands to handle objects in activities of daily life such as eating, dressing, and playing. The classification should focus on children's typical performance, not the maximum capacity. After the concept was defined, the group worked with the formulation of the five levels, how they would be expressed and distinguished from each other. The expert group defined the content of the levels based on their experience. Videos of children with different subtypes of CP who performed various manual tasks in their natural environments, mainly their homes and schools, were analyzed. The descriptions of the levels and their distinctions were discussed and revised, until consensus was reached in the expert group. A proposal for a classification of hand function, the Manual Ability Classification System, with five levels, and distinctions between levels for children with CP aged 8–12 years, was ready for testing, the first field version.

5.1.2 External processes for validation

EVIDENCE OF TEST CONTENT AND RESPONSE PROCESSES

The first version of the classification was presented to professionals within pediatric rehabilitation and to parents (Study I). Comments and suggestions about MACS, collected from professionals at national and international conferences, were brought back to the expert group and processed. The refinement of the wording in the descriptions of the levels and the distinctions between levels was a continuous process until the reliability study started.

Both parents and therapists expressed that the concept was easy to understand and that MACS provided a good description of how children with CP use their hands when handling objects in daily life (Study II). The descriptions in MACS were clear and easy to understand. The structure outlined in the brochure - the levels and the distinctions between the levels - facilitated the choice of level. The parents and therapists highlighted that some words in the level descriptions, like quality, speed, and independence, could be interpreted differently by different people. This may influence the choice of level and it seems that MACS could be improved by the clarification of these terms.

Most of the children were rated at the same level by parents and therapists. When they described how they chose a level, they frequently used the expressions from the MACS leaflet. They also described in their own words characteristics of the child's performance that were not mentioned in the MACS leaflet.

Only seven out of 25 children were classified at different levels by parents and therapists. The disagreement was distributed across all levels, but it differed by only one level between subjects' estimates of the child's ability. The reasons for choosing different levels were:

(i) children's performance varies in different environments, for example, at home and in school

(ii) whole-day performance versus a limited selection of daily activities were described;

(iii) different degrees of complexity of the activities were described.

These findings confirm what is described in the leaflet, that the MACS level must be determined by asking a person who knows the child's performance well.

When discussing the overall usefulness of the classification system, parents suggested that MACS could be used for several purposes, for example, in communicating with the social services and the local council to discuss the need for support and personal assistance, and in describing to newly employed staff working with the child what the child can do. The therapists expressed similar ideas, but also emphasized the ability to use MACS in the clinic for planning of treatment and goal setting. Therapists emphasized that MACS provides them with a structure of thinking. Parents stressed the advantage of describing what the children actually do, instead of focusing on their limitations.

EVIDENCE BASED ON RELATIONS TO OTHER VARIABLES

In Study III, when comparing MACS with one instrument measuring performance, ABILHAND-Kids, and one measuring capacity, the Box and Block Test, a strong relation between MACS and both instruments was found (MACS–ABILHAND-Kids r_s = -0.88 $p < 0.05$, MACS–Box and Block Test (r_s = -0.81 $p < 0.05$). Children classified in higher functional levels of MACS could master more activities in ABILHAND-Kids and transport more blocks in the Box and Block Test (Fig. 1, Study III). A significant difference in the children's performance in both ABILHAND-Kids and the Box and Block Test was shown between the different MACS levels (ABILHAND-Kids $F(4{:}86) = 103.86$ $p < 0.001$, Box and Block Test $F(4{:}86) = 59.18$ $p < 0.001$). This implies that there is a meaningful difference in hand function between the different MACS levels. The estimation of the coefficient of variation demonstrates a somewhat higher value in the Box and Blocks Test (CV = 58) compared to ABILHAND-Kids (CV = 46) results. This illustrates that children show a remarkably high capacity when performing the Box and Block Test, even if the manual ability is at a lower functioning level. MACS explained a lot of the total variance of the results of both ABILHAND-Kids (82%) and the Box and Block Test (72%) showed by linear regression analysis.

In the content comparison between MACS, ABILHAND-Kids, and the Box and Block Test, the ICF-CY was used as a frame of reference (Study III). All the concepts found in the three instruments, 37 in MACS, 31 in ABILHAND-Kids, and 2 in the Box and Block Test, were linked to categories in the component of activity and participation. MACS covered categories in seven chapters, ABILHAND-Kids in two chapters, and the Box and Block Test in only one chapter (Table II, Study III). Focusing on second-level categories in the ICF-CY, MACS covered the broadest bandwidth, with 15 categories, compared to ABILHAND-Kids (8 categories) and the Box and Block Test (1 category) (Table II, Study III). The only second-level category represented in all three instruments was fine hand use (d440, chapter 4, Mobility). For MACS and ABILHAND-Kids, an additional seven categories were similar, showing that they capture the same contents. There were no categories addressed only to ABILHAND-Kids or the Box and Block Test. For further understanding of the description of each category of ICF-CY, see the World Health Organization website (accessed at www.who.int/classifications/icf/en/; WHO, 2011).

MACS was also compared with PEDI in Study IV. A strong relationship between the children's ability in functional skills in self-care and mobility and their manual ability and gross motor function was found. The children who were rated to higher functioning levels in MACS and GMFCS were more capable in both the self-care and mobility domains of PEDI than children of low functioning levels, demonstrated by high correlations, shown in Table X (Fig. 1, Study IV).

TABLE X. Correlation coefficients between PEDI, MACS and GMFCS, $p < 0.05$

PEDI	MACS	GMFCS
Functional Skill Scale Self-care	-0,76	-0,71
Functional Skill Scale Mobility	-0,76	-0,83

The children's performance in self-care differed significantly between the different MACS levels ($F_{4,190} = 92.9$; $p < 0.001$), and performance in mobility differed significantly between the GMFCS levels ($F_{4,190} = 148.5$; $p < 0.001$).

A strong correlation between MACS and GMFCS was demonstrated in both Studies I and IV ($r_s = 0.79$, $r_s = 0.77$; $p < 0.05$). The total agreement showed that only half of the children demonstrate analogous levels of function on GMFCS and MACS in both studies (49% and 52%). The distribution of the MACS and GMFCS levels in the children in Study IV is shown below in Table IX (see also Table V in Study I, Eliasson et al., 2006). For example, in both studies the group classified as MACS level III comprised children in four different GMFCS levels. This finding indicates that GMFCS and MACS give complementary information from an individual perspective and are not interchangeable with each other.

TABLE IX. The distribution of children's MACS and GMFCS levels in study IV

	MACS levels					
	I	II	III	IV	V	Total
GMFCS levels						
I	**47**	38	5	-	-	90
II	6	**13**	13	-	-	32
III	2	13	**10**	4	-	29
IV	-	2	5	**9**	5	14
V	-	-	-	1	**22**	23
Total	55	66	33	14	27	195

5.2 EVIDENCE OF RELIABILITY

INTERRATER RELIABILITY BETWEEN THERAPISTS

Between therapists the ICC was 0.97 (95% confidence interval [CI] 0.96–0.98) for the whole group, indicating excellent agreement (Table III, Study I). The total agreement was 84.5%. When ICC was calculated for the different ages, there were similar results (Table IV, Study I). The group of 20 children from Australia was first analyzed separately and no differences in the understanding of the MACS were found; however, ICC was slightly lower at 0.91 (95% CI 0.77–0.96). This group of children was then included in the analysis of the whole group.

INTERRATER RELIABILITY BETWEEN PARENTS AND THERAPISTS

Between parents and therapists the ICC was 0.96 (95% CI 0.89–0.98), demonstrating excellent agreement for the 25 children aged 8–12 years. The total agreement was 72%. The raters' disagreement was just one level for seven children; five of them were rated on a higher level by the parents, and two by the therapists. The disagreement was distributed across all the five levels of MACS.

5.3 SELF-CARE AND MOBILITY SKILLS IN CHILDREN WITH CP

In Study IV the purpose was to investigate self-care and mobility skills in children with CP at different ages in relation to their manual ability (MACS) and gross motor function (GMFCS) levels. Self-care and mobility skills were evaluated by PEDI. To investigate how MACS and GMFCS respectively contributed to the outcome of the children's self-care and mobility, a linear regression analysis was accomplished. MACS explained 66% and GMFCS 56% of the variance in self-care, and the corresponding values for mobility were 70% and 76% (Table II, Study IV). How much MACS, GMFCS, and age together influenced the self-care and mobility scores was investigated by multiple regression models, in forward stepwise analysis. (Table III, Study IV). MACS, age, and GMFCS contributed to 80% of the variation in self-care scores and 87% in mobility scores. MACS was the strongest predictor of self-care and GMFCS the strongest predictor for mobility. These results justify the decision to evaluate the relationship between MACS and self-care, and GMFCS and mobility.

An age-related increase of self-care was seen in children in MACS levels I and II, verified by the strong correlation between age and self-care in these levels ($r_s = 0.85$ and $r_s = 0.76$; $p < 0.05$; Fig. 3a–b). Many children at these two levels achieved a maximal score on PEDI, but much later than for typical developed children. For children at MACS level I this was at 9 years of age. Only a few children at level II reached a scaled score of 100, and this was from the age of 12. Children with typical development reached a maximal score at the age of 6.5 years. Also in mobility scores there was an increase according to age, mainly in GMFCS level I ($r_s = 0.74$; $p < 0.05$; Fig. 4a). In the other levels the relationship to age was not so clear. A maximal score was observed in GMFCS level I from the age of 6. However, in level II only half of the group achieved a maximal score, and that was not until the age of 12 or older. This is also later than in typically developing children, who reach a maximal score at about 3.5 years. These findings show that children with CP have different levels of ability and different rates of development in self-care and mobility skills, depending on their level of MACS and GMFCS. It also shows that children with CP have a prolonged development, especially in self-care skills, even if they have fairly good manual ability.

6 DISCUSSION

The results in this thesis show that the Manual Ability Classification System can be used to describe manual ability according to five different levels in children with CP. MACS is built on a unique construct. Different types of evidence for validity and reliability show that MACS captures manual ability in the whole spectrum from mild to severely limited ability to handle objects in everyday life. The correlation to GMFCS (Palisano et al., 2008) is high, although only about 50 percent of the children were assessed at the analogous level in both systems. This indicates that the two constructs are not interchangeable, but describe children's functioning in a complementary way. By dividing the children according to MACS and GMFCS levels and analyzing self-care and mobility skills, an age-related increase in self-care and mobility was found, but only among children classified in MACS level I and II and GMFCS I, respectively .

6.1 MACS CLASSIFIES THE ACTUAL DOING

The concept "manual ability" in MACS is defined as the ability to handle objects in daily activities. This reflects the child's ability according to ICF's definition of performance (WHO, 2001a, WHO, 2007). Performance is what an individual does in his or her current environment. This is different from the individual's maximum capacity in a test situation, which typically measures what they actually can do. Distinguishing between performance and capacity is an important aspect for occupational therapists, physiotherapists, and other health professionals to consider when meeting the children with CP. The child's best capacity is usually what is found when tested in different clinical settings, but it might differ from what the children actually do in their daily environment. In MACS, we ask for the performance in daily activities. Therefore, it is quite important to ask a person who knows the child well about which objects the child is handling and how he/she usually handles them. Therapists might know the children very well, but they typically observe a limited range of the child's daily activities, while parents see the child's performance from a whole-day perspective. Although there was high reliability between parents and therapists, the difference found for a few raters in Study II may be explained from this perspective.

When applying an ICF perspective to MACS, environmental factors also become important. Different factors in the surrounding environment affect manual ability. This becomes especially evident for children in MACS levels III–V. They need various environmental adjustments and technical aids to fulfill their activities. The importance of the environment was described by both the parents and therapists in Study II. For example, children in MACS level IV need exact positioning of the object to be able to perform parts of the task. As such, their performance was dependent on the skills of the person who was assisting them. Children on level III typically need tools such as special forks or glasses for eating and drinking. This close relation between the task, the person, and the environment is also supported in different theoretical models of occupational performance (Kielhofner, 2008, Law et al., 1996, Townsend and Polatajko, 2007).

Another important finding from Study II was that the child's individual inner drive and motivation was a crucial factor for which classification level that was obtained. Parents of some children described that the children could do a lot of activities, but they never took the initiative to do them. The children's ability to handle objects is not only related to motor function; other body function aspects also influence their ability. However, the underlying reasons for a manual dysfunction are not described in MACS. Hand function is complex, but in MACS we are not trying to explain the reason for not doing things; we classify the child's actual doing.

6.2 EVIDENCE OF VALIDITY

Validation of an instrument is an ongoing process. Validity refers to "the degree to which evidence and theory support the interpretations of test scores entailed by the proposed uses of tests" (American Educational Research Association, 1999, Cook and Beckman, 2006, Goodwin, 2002). In other words, validity describes how well one can legitimately trust the results of a test or a classification as interpreted for a specific purpose. All four studies in this thesis have contributed to the body of evidence concerning the validity of MACS. The concept of manual ability has been evaluated from a broad perspective with both qualitative and quantitative methods. So, with regard to validity, what is known about MACS as a classification system, and how does this knowledge impact the usage of MACS?

EVIDENCE BASED ON TEST CONTENT

Evidence of content validity refers to the extent to which a measure represents all facets of a given construct (American Educational Research Association, 1999). In interviews with parents of children with CP and therapists treating these children, it was confirmed that the wordings and descriptions of the different MACS levels corresponded well with the children's manual ability in daily life. Both parents and therapists perceived that MACS gave a relevant description of different levels of hand function, which strengthens the fact that the classification system captures a valid description of the child's performance and that the levels describe meaningful differences of performance.

EVIDENCE IN BASED ON RELATION TO OTHER VARIABLES

The relation between MACS results and other variables was investigated in two ways: by comparing MACS results with three other instruments measuring aspects of hand function and functional performance (Studies III and IV), and by comparing MACS results with those of another classification for children with CP, the GMFCS (Studies I and IV). In Study III it was shown that the outcomes of the ABILHAND-Kids and the Box and Block Test differed significantly between the five different MACS levels. In Study IV, children with high functioning levels in MACS were found to be more capable than children in low functioning levels in self-care skills measured by the Pediatric Evaluation of Disability Inventory, with significant differences between all classification levels ($p < 0.001$).

Relation to other variables of capacity and performance

The results of the Spearman rank order correlations for MACS and the two other instruments evaluating hand function were high, and similar for both instruments (r_s = -0,88 $p < 0.05$ and r_s = -0,81 $p < 0.05$). Thus, from the correlation coefficients it was not possible to detect differences between the relations of the instruments, even though they measure different aspects of hand function in terms of capacity and performance. The correlation between MACS and ABILHAND-Kids, which measures performance, were as high as between MACS and Box and Block, measuring best capacity. The approximately equally high correlations may be a result of the way rank correlation is calculated. A rank correlation is a mathematical approximation where one compares values in pairs (Campbell et al., 2007). A high value of correlation indicates that the scales of the different instruments are constructed in the same manner, measuring from low to high abilities. The children perform similarly in the different instruments; for example, a child rated in MACS level I most probably has a higher score on both ABILHAND-Kids and the Box and Block Test than a child assessed at lower ability MACS levels. Thus, the correlation coefficient describes the relationship according to rank order, but does not describe the relationship regarding the content of the instruments. Therefore, in Study III, the instruments were also compared using ICF-CY as a frame of reference. The result showed that all the instruments captured aspects of hand use in the domain of activity and participation. It demonstrated that the content of MACS related to seven chapters in the ICF-CY, thereby showing a broader description of hand use than ABILHAND-Kids with content represented in two chapters, and the Box and Block Test with content represented in one chapter. The linking to ICF-CY provided a possibility to compare the content of the different instruments. However, the linking procedure has some limitations, since the linking rules by Cieza (2005) do not in a totally clear way describe how to establish the meaningful concepts in the instruments (Xiong and Hartley, 2008). This part of the linking process involves a great deal of interpretation. We determined how to extract the concepts during the linking process in a consensus group before starting the linking process. This was done to increase the reliability.

MACS and GMFCS describe various constructs

When evaluating the relationship between MACS and GMFCS results, the correlation was also high ($r_s = 0.77$, $p < 0.05$). This result could be interpreted as indicating that they both measure the same construct. However, in both Study I and Study IV, cross-tabulation of children's MACS and GMFCS levels shows that it was just half of the children who had analogous levels in both classifications (Table V in Study I and Table IX under Results). An example is children at MACS level III who are represented in four different GMFCS levels. This is an important finding, showing that MACS and GMFCS are not interchangeable, although the correlation between them was high. There is a need of both classification systems to describe the children's functioning, because they describe various constructs (Rosenbaum et al., 2008). Other studies have shown similar results regarding the relation between the two classification systems (Akpinar et al., 2010, Gunel et al., 2009, Imms et al., 2009, Morris et al., 2006b, van Meeteren et al., 2010).

The evidence of test content and relations to other variables, demonstrated that the descriptions of the levels clearly differentiate between the children's manual abilities and that MACS could discriminate and appropriately rank order manual ability according to the severity of the disability. Other studies have also compared MACS to other variables and found significant differences between different MACS levels, this evidence further strengthen the content validity (Akpinar et al., 2010, Kuijper et al., 2010, Smits et al., 2010, van Eck et al., 2010, van Meeteren et al., 2010).

6.3 EVIDENCE OF RELIABILITY

Excellent interrater reliability scores were obtained both when two therapists rated the child (r_s=0,97) and when the child was rated by their parent and a therapist (rs=0,96) (Study I). This is in line with other studies (Akpinar et al., 2010, Kuijper et al., 2010, Mutlu et al., 2010b). The interrater reliability has also been investigated in young adults with CP with excellent interrater reliability (van Meeteren et al., 2010). All these results indicate that the MACS distinguished well between different levels of manual ability for children, adolescents as well as adults.

This result also demonstrate that parents and therapists are rating equally when classifying the child's manual ability, which show that they have a similar view of the child's performance when handling objects. However our sample was small (n=25) but even studies with larger samples show the same excellent interrater reliability (Akpinar et al., 2010, Mutlu et al., 2010b). Another study by Morris and colleagues (2006) has also evaluated the reliability between ratings of parents and therapists but with a somewhat lower interrater reliability (r_s=0.73-0.85). One explanation could be that different procedures were used when introducing MACS to the raters. Morris and collaborators simply sent the MACS brochure by post, whereas in study I we gave a brief introduction to the MACS concept before the parents and therapists did the rating.

Interrater reliability has been investigated in other studies in different countries, that is, Great Britain, the Netherlands, Turkey, and Australia. The high reliability coefficients show that the level descriptions fit for the classification of children's performance in different cultures (Akpinar et al., 2010, Kuijper et al., 2010, Morris et al., 2006b, van Meeteren et al., 2010).

The above described evidence of reliability show that MACS levels for children with CP are consistent over repeated applications. It is important to highlight that reliability and validity are properties of the measurement outcomes and not of the instrument itself. Thus, MACS is a valid and reliable classification for children, adolescents and adults with CP. However, it may also be applicable for use with other diagnostic groups although only after investigation of validity and reliability for the new target group.

6.4 CLASSIFICATIONS ARE NOT TESTS

It is important to distinguish between classification and tests. MACS, like GMFCS and CFCS, is a classification system, and not a test. These systems are designed to group people into classes and/or categories, according to common characteristics. MACS groups children with CP according to different levels of manual ability, GMFCS according to gross motor function, and CFCS according to communication ability. Tests are instruments that use a standardized procedure and are evaluated for their ability to accurately measure parameters of behavior within a specific population. Since classification levels are crude and do not include detailed descriptions, they do not intend to detect change. Clasifications can, therefore, not be used to evaluate intervention.

Furthermore, classifications are, as described earlier, intended to be stable over time. Children may develop their manual ability over time and be more skilled in handling objects in older ages, but within their own MACS level. Naturally, a child whose performance is on the borderline between two levels may, after intervention or with growth, change levels. Stability over time has been evaluated for GMFCS, and a high stability was demonstrated. Almost three-quarters of the children remain in the same level over time, which has been important for understanding how children with different GMFCS levels develop (Hanna et al., 2008, Palisano et al., 2006, Rosenbaum et al., 2008). The stability of MACS has so far only been investigated in one study and was found to be good (Imms et al., 2010). Children were rated with MACS by caregivers twice, with a year in between. Further evaluation of the stability of MACS over a longer time period is needed. If MACS is shown to be stable to about the same extent as GMFCS, it could also be used for predictive purposes and contribute to a more differentiated way to understand how children with CP develop.

6.5 INDEPENDENCE IN DAILY ACTIVITIES RELATED TO MANUAL ABILITY

In the last study in this thesis children's self-care ability measured by PEDI was analyzed in relation to their manual ability. An interesting finding was that children with CP have a prolonged development in self-care compared to typically developing children, even if their hand function was fairly good (MACS levels I–II). Surprisingly, it took as long as three additional years before most of the children in MACS level I reached the maximum scaled score in this sample. This can be understood from a different perspective: one explanation is the complexity of self-care activities, which require both gross motor and executive skills, in addition to hand use. Another reason for not reaching a maximum score could be that certain tasks require high level of fine motor skills, for example, doing or undoing buttons and zippers. A third explanation is that these children need extensive practice to master more complex activities than children without CP (Gordon and Duff, 1999). This need for extensive practice was also confirmed by parents in Study II. Furthermore, the construct of the PEDI scale must also be discussed in relation to the results. The Rasch conversion of raw scores to scaled scores implies that, at the end of the scale, a few raw scores missing makes a big difference in scaled scores. In self-care, for example, if only three of the 73 items are not mastered, the child reaches a scale score of 80 out of the total of 100 scaled scores.

The finding that children with more severely affected manual ability (MACS levels III–V) had no age-related increase in self-care might be less surprising. Even if they improve in their co-occupation, i.e. to do more parts of the activities together with the parents, they do not reach independency and PEDI is not sensitive to detect change of of only parts of an activity (Shephard, 2010). Another explanation is that these children have a very slow pace of performance and consequently limited ability to perform the activities on a daily basis, which implies that they do not get much experience. Also, the children's "inner drive" influences the actual doing described by the parents in Study II.

MACS was a stronger predictor of self-care than GMFCS, while GMFCS was the strongest predictor of mobility. Gross motor function has earlier been found to be an important prerequisite for both self-care and mobility (Voorman et al., 2006, Østensjø et al., 2003). When using the GMFCS, the same pattern in development of children at higher functional levels has earlier been found; none of the preschool children reached independence in self-care investigated by Östensjö and collaborators (2003). When using another scale of PEDI, the caregiver assistance scale, in relation to MACS the same association was found: children in high functioning levels were more capable of self-care than children in low functioning levels, although they were not analyzed according to age (Kuijper et al., 2010). The association between MACS and self-care seems also to be true for older adolescents, using the assessment Vineland Adaptive Behavior Scales (VABS) subscales for daily living skills (van Eck et al., 2010). In our study it was shown that 66% of the variance could be explained by MACS, and that 80% was described by MACS, GMFCS, and age together. Therefore, 20% is influenced by other factors. However, it is important to highlight that MACS is a description of the child's manual ability and not an assessment of ADL. If self-care is the main interest, an ADL assessment must be used. A limitation of this study, as well as the other studies, is that cross-sectional data were used. Further research with repeated measures of self-care ability in a longitudinal study is needed to describe the children's development in self-care skills.

6.6 USABILITY AND APPLICABILITY OF MACS

The usability of MACS refers to how practical and easy MACS is to use, and applicability refers to how relevant MACS is when used for this group of children. The evidence of validity and reliability presented in this thesis imply that MACS can be used in both research and clinical practice. It also seems to be applicable, since it already is used in different ways.

For research it has been used in several studies. It has been cited 228 times since the first publication in June 2006 (Google Scholar, 7 Sept 2011). It can be used to compare results from different studies. It has been used for subdiagnosis, such as in children with unilateral CP, when describing the development of hand function of children in different MACS levels in a longitudinal perspective (Holmefur et al., 2010). It has been used for stratification of groups to be included in studies (Arner et al., 2008, Carnahan et al., 2007, Gong et al., 2010, Mutlu et al., 2010a) and in a population-based register (CPUP). MACS has also been used together with other instruments measuring both hand function and daily activities (Akpinar et al., 2010, Gunel et al., 2009, Morris et al., 2006a, van Meeteren et al., 2010, van Meeteren et al., 2008), and to relate the children's MACS level to ability in self-care (Kuijper et al., 2010, Smits et al., 2011, Voorman et al., 2006, Østensjø et al., 2003). Although not validated for older persons or children below 4 years, MACS has been used for young adults (Nieuwenhuijsen et al., 2009, van Eck et al., 2010, van Meeteren et al., 2010, van Meeteren et al., 2008).

MACS seems to attract people in different countries and cultural settings, since it has been translated into many different languages. Today, the brochure can be downloaded in 23 different language versions from the website. When parents are using the MACS brochure, the ability to read it in the native language has been found to be of importance (Akpinar et al., 2010, Imms et al., 2010). To improve its usability, the brochure was revised in 2010, and a "decision tree" or flowchart for decision making had been developed. The aim is to simplify the rating process, and the flowchart can be downloaded from the website in several languages (accessed at www-macs.nu). An instructional video is now available for purchase describing MACS and the rating process. Although MACS is available in a lot of different languages, cross-cultural validation has only been investigated in Turkey (Akpinar et al., 2010, Imms et al., 2010). This might be a limitation, though the cross-validation is important to show whether the instrument's construct fits the culture. In clinical practice MACS can be used in communication between parents and professionals. It can be used together with the child and the parents in decision making for treatment. It can be useful for deciding on realistic and achievable goals. Parents found it applicable for communication with policymakers and social services, to describe what type of assistance the child needed. Parents also highlighted the positive experience with an assessment classifying the child's possibilities rather limitations. In summary, since the evidence of validity and reliability is supported by the parents' opinions, it would be possible to say that MACS is both usable and applicable.

6.7 METHODOLOGICAL CONSIDERATION

MACS has been evaluated and compared to a number of other instruments during the development process. Validation of an instrument is an on-going process and MACS needs to be evaluated in comparison with instruments describing the performance perspective that MACS is constructed to capture. It would have been interesting in study I to compare the level distribution in MACS to another criterium-based measure developed for children with CP. When MACS was developed no suitable test measuring performance were available that could have been used as a gold standard to compare with.

In study II the analysis was influenced by directed content analysis. The directed content analysis approach can be used to validate or conceptually extend a theoretical framework or theory. This analysis approach has been referred to as a type of deductive category application. During the data reduction of the interviews in study II it was decided to transcribe only parts of the collected data because there was quite a large number (n=50) of interviews included in the study. The parts that were transcribed were those that dealt directly with the previously defined themes and these were transcribed verbatim by the interviewer. This choice of method could be a possible limitation because important information related to the themes might by mistake be omitted from the transcriptions. To ensure that this would not happen, all interviews were listened through several times. Another potential limitation in study II was the interviewers pre-understanding of the field which always to some extent influence the direction of the interviews. This, however, could also be seen as a strength of the study since this meant that the interviewer had the experience needed to ask adequate follow up questions to get a deep understanding of the child's hand use in daily activities.

The linking procedure used in study III has some limitations, since the linking rules by Cieza (2005) do not in a clear way describe how to establish the meaningful concepts in the instruments. It does not describe how to extract the meaningful concepts in a classification This part of the linking process involves a great deal of interpretation (Xiong and Hartley, 2008). To overcome this, it was decied to treat MACS as a one item measure and we determined how to extract the concepts during the linking process in a consensus group before starting the linking process. This was done to increase the reliability.

In Study IV the study group consisted of registry data in combination with a convenient sample. Although the convenient sample was recruited with the aim of achieving a large variation of ages, diagnosis and classification levels, there are few children in some of the MACS and GMFCS levels within a few of the age groups. This can of course be regarded as a limitation of the study, but when comparing the percentage of children included in each level of the classifications (MACS and GMFCS) in the study to the population-based CPUP data base the percentage of children within each category can be viewed as representative of the population of children with CP living in Sweden.

In the sample in Study IV, children aged 3 years were included although MACS is designed for children from 4 years of age. All 3 year olds that were included in the study were going to turn 4 within six months of participating in the study i.e., all children included were older than 3 years and 6 months. Since MACS describes children's age related handling of objects we do not think the extended age range, including children that had not turned 4, influences the results regarding the number of children within each MACS level.

7 CONCLUSION AND CLINICAL CONSIDERATIONS

In conclusion, the studies in this thesis demonstrated that MACS is a classification that provides a description of manual ability in a valid and reliable way, for children and adolescents with CP, aged 4 to 18 years. Manual ability is defined as the child's self-initiated ability to handle objects in everyday life. MACS is built on a unique construct and describes what the children actually are doing instead of focusing on their limitations.

The MACS levels differentiate effectively between the severity of children's/ adolescent's manual abilities, shown by the test content's relation to other variables as well as by an excellent interrater reliability. MACS has proved to have strong correlation to other instruments of both hand function and performance of daily activities. Content analysis with ICF-CY as a frame of reference shows that MACS contains a broader representation of different aspects of activity and participation compared to the instruments ABILHAND-Kids and Box and Block Test. MACS can be used consistently by both parents and therapists demonstrated by an excellent interrater reliability by ratings made by two therapists as well as by parents and therapists.

When investigating predictors of independence of daily activity performance, MACS was found to be the best predictor of self care skills while GMFCS was the best predictor of mobility, as measured by the PEDI. A significant age-related increase in self-care and mobility was found, but only among children classified in MACS levels I and II and GMFCS I respectively. Many children at these levels reached full or almost full independence, but at a later age than children without disabilities.

MACS and GMFCS described different dimensions of children's everyday functioning. This was demonstrated by the fact that despite a high correlation between the two classification systems, only half of the children were classified at analogous levels. Thus, both classifications are needed to describe the functional consequences of CP, as a complement to diagnosis, subdiagnosis and dominant symptoms.

The implications of these results are that MACS can be used for both clinical practice and research. It is already commonly used in research for description of groups and stratification within the heterogeneous diagnosis of CP. In clinical practice MACS can facilitate the communication between parents and professionals. At an individual level

MACS can be used together with families to enhance discussions of intervention planning. By using MACS it might be easier to provide realistic and achievable goals.

Future research about the stability of MACS levels over time is needed. If MACS is stable over time it will be possible to predict future development of manual ability given the child's early classification level. The application of MACS in children under the age of four years needs also to be investigated.

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