

The importance of motor functional levels from the activity limitation perspective of ICF in children with cerebral palsy

Akmer Mutlu^a, Pelin Pistav Akmese^b, Mintaze Kerem Gunel^a, Sevilay Karahan^c and Ayse Livanelioglu^a

Our purpose in this study was to evaluate performance and capacity as defined by Gross Motor Function Classification System (GMFCS) and Manual Ability Classification System (MACS) from the 'activity limitation' perspective of International Classification of Functioning, Disability, and Health (ICF) and to investigate the relationship between the two classification systems in different subtypes of cerebral palsy (CP). This prospective cross-sectional study was performed on 448 children with CP ranging from 4 to 15 years of age. Activity limitations were studied with the GMFCS for gross motor function and MACS for manual ability. The Spearman's correlation coefficient, contingency coefficient, and Cramer's V coefficient were used to assess the strength and significance of the association between GMFCS and MACS. The overall agreement between GMFCS and MACS was found to be 41%. The agreement was 42% in spastic children, 40% in dyskinetic children, 50% in ataxic children, and 28% in mixed type children. The overall κ value was $\kappa=0.235$ ($P<0.001$). The κ coefficient was 0.252 in spastic children, 0.245 in dyskinetic children, 0.318 in ataxic children, and 0.023 in mixed type children. All the κ coefficients except the value for the mixed type were found to be significant. The usage of two different classification systems, GMFCS and MACS, to describe the capacity and performance in children with CP as defined by the ICF provides an easy and quick classification tool for indicating 'activity limitations' of ICF in children with CP. The next step in research should be to highlight the other domains such as participation restrictions in these children.

Ziel dieser Studie war die Evaluierung der Leistung und Leistungsfähigkeit laut Definition des GMFCS (System zur Messung und Klassifikation motorischer Funktionen) und des MACS (System der Klassifikation der manuellen Fähigkeiten) aus der Perspektive der 'Einschränkungen der Aktivitäten' der internationalen Klassifikation der Funktionsfähigkeit, Behinderung und Gesundheit (ICF) sowie die Untersuchung der Beziehung zwischen beiden Klassifikationssystemen in unterschiedlichen Subtypen der Zerebralparese (CP). An dieser prospektiven Querschnittsstudie nahmen insgesamt 448 Kinder mit CP im Alter von 4 bis 15 Jahren teil. Einschränkungen der Aktivität wurden anhand der Systeme GMFCS für körpermotorische Fähigkeiten und MACS für manuelle Fähigkeiten untersucht. Der Korrelationskoeffizient nach Spearman, der Kontingenzkoeffizient und der Cramer-V-Koeffizient wurden zur Beurteilung der Intensität und Bedeutung der Verbindung zwischen

GMFCS und MACS herangezogen. Insgesamt stimmten die Systeme GMFCS und MACS zu 41% überein. Die Übereinstimmung lag bei Kindern mit spastischer CP bei 42%, bei Kindern mit dyskinetischer CP bei 40%, bei Kindern mit ataktischer CP bei 50% und bei Kindern mit einer Mischform der CP bei 28%. Der Gesamtwert κ lag bei $\kappa=0.235$ ($P<0.001$). Der κ -Koeffizient lag bei Kindern mit spastischer CP bei 0.252, bei Kindern mit dyskinetischer CP bei 0.245, bei Kindern mit ataktischer CP bei 0.318 und bei Kindern mit einer Mischform der CP bei 0.023. Alle κ -Koeffizienten mit Ausnahme des Wertes für die Mischform der Zerebralparese erwiesen sich als signifikant. Die Verwendung von zwei unterschiedlichen Klassifikationssystemen - GMFCS und MACS - zur Beschreibung der Leistungsfähigkeit und Leistung von Kindern mit CP laut ICF-Definition ist ein Klassifikationstool, mit dem sich Einschränkungen der Aktivitäten bei Kindern mit CP leicht und schnell aufzeigen lassen. Als nächstes sollte die Forschung andere Domänen wie beispielsweise die Einschränkungen bei der Partizipation (Teilhabe) dieser Kinder am Leben in der Gesellschaft hervorheben.

Nuestro propósito en este estudio fue evaluar el rendimiento y la capacidad, según el Sistema de Clasificación de la Función Motora Global (SCFMG) y el Sistema de Clasificación de la Destreza Manual (SCDM), en lo relativo al dominio limitaciones para la ejecución de actividades de la Clasificación Internacional del Funcionamiento, de la Discapacidad y de la Salud (CIF), e investigar la relación entre dichos dos sistemas de clasificación en los distintos subtipos de la parálisis cerebral infantil (PCI). En este estudio transversal prospectivo participaron 448 niños de entre 4 y 15 años de edad con PCI. Para la exploración de las limitaciones para la realización de actividades se utilizaron el SCFMG, en el caso de la función motora global, y el SCDM, en el caso de la destreza manual. Para determinar la magnitud y significación de la asociación entre el SCFMG y el SCDM se utilizaron el coeficiente de correlación de Spearman, el coeficiente de contingencia y el coeficiente V de Cramer. La concordancia general entre los resultados del SCFMG y del SCDM fue del 41%. La concordancia fue del 42% en los niños que padecen espasticidad, del 40% en los niños con discinesias, del 50% en niños con ataxia, y del 28% en niños con cierta combinación de dichas afecciones. El valor general del coeficiente κ fue de $\kappa=0.235$ ($P<0.001$). Este coeficiente fue de 0.252 en los niños que padecen espasticidad, de 0.245 en los niños con discinesias, de 0.318 en niños con ataxia, y de 0.023 en niños con cierta

combinación de dichas afecciones. Los valores del coeficiente κ , excepto en el caso de los niños con el tipo mixto de la enfermedad, resultaron significativos. El uso de dos sistemas distintos de clasificación, el SCFMG y el SCDM, para determinar el grado de capacidad y de rendimiento, según se definen en la CIF, en niños con PCI constituye una herramienta de clasificación de uso fácil y rápido para determinar el grado de limitaciones para la ejecución de actividades, según la CIF, en niños con PCI. El siguiente paso en la investigación de estos niños sería explorar los otros dominios de esta clasificación, tales como las restricciones de estos niños para participar en actividades cotidianas.

Notre objectif dans cette étude était d'évaluer la performance et la capacité telle qu'elles sont définies par le système de classification GMFCS des fonctions motrices (*Gross Motor Function Classification System*) et le système de classification MACS des capacités manuelles (*Manual Ability Classification System*) du point de vue de la limitation de l'activité selon la classification internationale ICF des fonctions motrices, du handicap et de la santé, et d'étudier la relation entre les deux systèmes de classification dans les différents sous-types d'infirmité motrice cérébrale. Cette étude prospective transversale a été réalisée sur 448 enfants âgés de 4 à 15 ans souffrant d'infirmité motrice cérébrale. Les limitations de l'activité ont été étudiées avec la classification GMFCS pour la fonction motrice globale et la classification MACS pour l'habileté manuelle. Le coefficient de corrélation de Spearman, le coefficient de contingence et le coefficient V de Cramer ont été utilisés pour évaluer la portée et la signification des associations entre GMFCS et MACS. Le taux de corrélation global entre GMFCS et MACS a été mesuré à 41%. La corrélation était de 42% chez les

enfants handicapés moteur, 40% chez les enfants dyskinétiques, 50% chez les enfants ataxiques et 28% chez les enfants de type mixte. La valeur κ globale était $\kappa=0.235$ ($P<0.001$). Le coefficient κ était de 0.252 chez les enfants handicapés moteur, 0.245 chez les enfants dyskinétiques, 0.318 chez les enfants ataxiques et 0.023 chez les enfants de type mixte. Tous les coefficients κ , à l'exception de la valeur correspondant au type mixte, ont été jugés significatifs. L'utilisation de deux systèmes de classification différents, GMFCS et MACS, pour décrire la capacité et la performance chez les enfants atteints d'infirmité motrice cérébrale telle que définie par l'ICF fournit un outil de classification simple et rapide pour indiquer les limitations de l'activité de l'ICF chez les enfants souffrant d'infirmité motrice cérébrale. L'étape de recherche suivante sera de mettre en évidence les autres domaines, tels que les restrictions à la participation, chez ces enfants. *International Journal of Rehabilitation Research* 00:000–000 © 2010 Wolters Kluwer Health | Lippincott Williams & Wilkins.

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^aDepartment of Physical Therapy and Rehabilitation, Faculty of Health Sciences, Hacettepe University, ^bCelal Bayar University, Salihi Vocational School, Department of Child Education, Salihi, Manisa and ^cDepartment of Biostatistics, Faculty of Medicine, Hacettepe University, Samanpazari, Turkey

Correspondence to Akmer Mutlu, PT, PhD, Faculty of Health Sciences, Department of Physical Therapy and Rehabilitation, Hacettepe University, 06100 Samanpazari, Ankara, Turkey
Tel: +90 312 305 25 25 x164; fax: +90 312 305 20 12;
e-mail: akmer@hacettepe.edu.tr

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Introduction

International Classification of Functioning, Disability, and Health (ICF) provides a common language for describing health, functioning, and disability that is increasingly gaining worldwide acceptance. Although it is acknowledged that no model is perfect, the ICF model does represent the current global opinion on health and disability, and it is already being used in many countries for multiple purposes. ICF focuses on each disease such as musculoskeletal conditions, internal medicine conditions, cerebral palsy (CP) and stroke in terms of disease-specific assessment (World Health Organization, 2001; Allet *et al.*, 2007; Bürge *et al.*, 2008; Jönsson *et al.*, 2008; Snögren and Sunnerhagen, 2009). ICF has also been used as a unifying framework for the conceptualization of rehabilitation. A number of reports have described how to use ICF in rehabilitation practice (Ustün *et al.*, 2003; Stucki *et al.*, 2008; Rauch *et al.*, 2008; Kuipers *et al.*, 2009).

The ICF model can be used to guide clinical thinking and delivery of services to children and youth with CP and their families. Children with CP have many neurological

deficits that interfere with motor function and daily activities. These impairments include neuromuscular and musculoskeletal problems such as spasticity, coactivation of agonist–antagonist muscles, muscle weakness, and limited range of motion affecting gross and fine motor function. Additional neuroimpairments such as learning disability, epilepsy, visual impairment, and hydrocephalus frequently coexist and these impairments do not directly reflect the activity limitations and social participation although they do affect activities and participation in society (Aicardi and Bax, 1998). The aim of physiotherapy in children with CP is to normalize movement patterns, reduce neurological signs, and minimize the development of secondary impairments (Mayston, 2001; Arpino *et al.*, 2009). This approach is based on the assumption that increases in 'motor impairments' lead to a decrease in 'activity and functional capacity' and 'participation and social roles'. However, there are only a few articles on this assumption. We believe this may be because of three factors: (i) the range of clinical types and affectation in CP and the personal, environmental, family-related, social and cultural factors acting together to create an even

more heterogenous structure; (ii) the variety of treatments, and (iii) the differences in growing maturation and lifestyle.

In the clinical context of pediatric neurorehabilitation, appropriate and accurate tools are essential to measure the most relevant outcomes for 'activity limitations' and 'participation restrictions' (Rosenbaum and Stewart, 2004). The Gross Motor Function Classification System (GMFCS) has been rapidly accepted into clinical practice and research (Morris and Bartlett, 2004a), and has been shown to be directly related to restrictions in activity and participation (Beckung and Hagberg, 2002). The GMFCS classifies the child's movement ability whereas the Manual Ability Classification System (MACS) represents the child's manual ability. A few recent studies on the MACS have shown that it is a valid and reliable classification tool although it has not been as widely used as the GMFCS to date (Eliasson *et al.*, 2006; Carnahan *et al.*, 2007; Kerem-Gunel *et al.*, 2009). There is still little practical experience in the use of these classifications in the pediatric population (Fergren and Lagergren, 1998; Simeonsson *et al.*, 2000, 2002; Beckung and Hagberg, 2002).

In our earlier study we studied impairments, activity limitations, and participation restrictions in domains of self-care, mobility, communication, social relations, learning and applying knowledge, as proposed in ICF. In addition, the relationship between ICF and functional independence was investigated in children with CP. We concluded that ICF could be an appropriate model for our country if a common language can be provided for the rehabilitation team of children with CP (Kerem-Gunel and Mutlu, 2007).

To understand the daily lives of children with CP and their families, we need to look outside the clinical setting as well as inside their 'activities' and 'social participation'. In addition, we should ask what is important to our patients and families, and use measures that are valid, reliable, and responsive. This approach will allow us to understand 'what they are really doing' in their daily life and whether our interventions have a meaningful impact (Bjornson, 2008).

Our purpose in this study was to evaluate the performance and capacity as defined by GMFCS and MACS from the 'activity limitation' perspective of ICF and to investigate the relationship between the two classification systems in different subtypes of CP.

Methods

Participants

The inclusion criteria for the study were children diagnosed with CP, children between 4 and 15 years of age, and parents accepting to participate in the study. The exclusion criteria for the study were children younger than 4 years of age, and parents not accepting to

participate in the study. Informed consent was obtained from the families after they were informed about the study. The study received Ethics Committee Approval from the Hacettepe University Medical Faculty Ethics Committee (*Registration Number: HEK 09/60*).

Procedure

This prospective cross-sectional study was performed on 448 children with CP referred to our physical therapy and rehabilitation unit by pediatric neurologists. Clinical type and extremity distribution were determined according to subtypes of CP and classified according to the Swedish Classification (Hagberg *et al.*, 1975). Swedish Classification divides CP into four subtypes as: spastic, dyskinetic, ataxic, and mixed type.

Activity limitations were studied with the GMFCS for gross motor function and MACS for manual ability (Palisano *et al.*, 1997; Eliasson *et al.*, 2006).

The GMFCS level (Palisano *et al.*, 1997) and the MACS level (Eliasson *et al.*, 2006) were classified by the same pediatric physiotherapist (A.M.) through observation, assessment, and questions asked to the parents and caregivers about the children.

Assessment tools

Gross Motor Function Classification System

The GMFCS is a common classification system and an evidence-based classification tool of five levels ranging from level I, which includes children with minimal or no disability with respect to community mobility, to level V which includes children who are totally dependent on external assistance for mobility (Palisano *et al.*, 1997; Morris and Bartlett, 2004a). The Turkish version of the expanded and revised GMFCS (translated by Kerem-Gunel *et al.*, 2009) was used in the study (Palisano *et al.*, 2008).

Manual Ability Classification System

MACS is based on self-initiated manual ability and provides a systematic method to classify how children with CP use their hands when handling objects in daily activities. Five levels are described. Level I includes children with CP with, at most, minor limitations compared with typically developing children, and where the limitations, if any, barely influence their performance of daily tasks (Eliasson *et al.*, 2006, 2007).

Statistical analysis

The statistical analysis was performed with SPSS for Windows version 15.0 (SPSS Inc., Chicago, Illinois, USA). *P* values less than 0.05 were considered to be statistically significant. The Spearman's correlation coefficient, contingency coefficient, and Cramer's V coefficient were used to assess the strength and significance of the association between GMFCS and MACS.

The overall agreement between the GMFCS and MACS was analyzed using the κ statistics. The κ value was interpreted as follows: < 0.20 poor, 0.21–0.40 fair, 0.41–0.60 moderate, 0.61–0.80 good, and > 0.80 very good agreement (Altman, 1997).

Results

The total material comprised all 448 children with CP ranging from 4 to 15 years of age, with a mean age of 7.04 ± 2.60 years. Two hundred and thirty-three (52%) of the children were girls and 215 (48%) were boys.

Table 1 reports the distribution of different clinical subtypes of CP and the level of limitation expressed by GMFCS and MACS.

Table 2 represents the distribution between the levels of GMFCS and MACS among children with CP. Totally, the highest number of participants were in level I and level II for both the GMFCS (29.01 and 25.66%) and MACS (31.47 and 33.48%) groups. There were no participants in both level V of GMFCS and level I of MACS. In addition, no participants were in both level V of MACS and level I–II and III of GMFCS. There was a significant positive association between GMFCS and MACS (Cramer $V = 0.398$, contingency coefficient = 0.623, $P < 0.001$).

Correlation of GMFCS and MACS levels in relation to CP subtypes were investigated and shown in Table 3

The overall agreement between GMFCS and MACS was found to be 41%. The agreement was 42% in spastic children, 40% in dyskinetic children, 50% in ataxic children, and 28% and in mixed type children. The overall κ value was $\kappa = 0.235$ ($P < 0.001$). The κ coefficient was 0.252 in spastic children, 0.245 in dyskinetic children, 0.318 in ataxic children, and 0.023 in mixed type children. All the κ coefficients except the value for the mixed type were found to be significant.

Discussion

The definition of CP focuses on activity limitations and problems in functional motor abilities. The World Health Organization’s International Classification of Functioning, Disability, and Health speaks of ‘activity’ as ‘...the

Table 2 Percentage distribution between levels of GMFCS and MACS among children with CP

GMFCS levels	MACS levels				
	1 (%)	2 (%)	3 (%)	4 (%)	5 (%)
1	19.19	8.03	1.33	0.44	0
2	7.58	11.16	3.34	3.57	0
3	3.57	7.58	2.45	0	0
4	1.11	5.35	5.35	4.46	0.22
5	0	1.33	4.91	5.13	3.79

Cramer $V = 0.398$ ($P < 0.001$), contingency coefficient = 0.623 ($P < 0.001$). CP, cerebral palsy; GMFCS, Gross Motor Function Classification System; MACS, Manual Ability Classification System.

execution of a task or action by an individual’, and identifies ‘activity limitation’ as ‘...difficulties an individual may have in executing activities’. This term amplifies the earlier concept of ‘disability’ to recognize changing international concepts and terminology (Bax *et al.*, 2005). The WHO’s ICF, along with several other recent publications, has sensitized health professionals to the importance of evaluating the functional consequences of different health states. The functional consequences of involvement of the upper and lower extremities should therefore be separately classified by using objective functional scales. GMFCS has been widely used internationally for the key function of ambulation to group individuals with CP into one of five levels based on functional mobility or activity limitation (Palisano *et al.*, 1997). MACS, a parallel classification scale, has been developed for assessing upper extremity function in CP, and has been shown to have good interrater reliability between parents and professionals (Eliasson *et al.*, 2006, 2007).

Although it is usually the impairments (muscle tonus, range of motion, hearing and visual problems) that are queried in CP, the ICF emphasizes that activity limitation is quite important and should also be assessed. We need to focus on inside the ‘activities’ and ‘social participation’ of children with CP to see what they are really doing in their lives as stressed by Bjornson (2008). This is why activity limitations and participation restrictions in social life as proposed in ICF requires more attention and research. We therefore focused on both gross motor and manual ability function measurements indicating activity limitations in this study. As the ICF becomes more popular within the clinical setting, an increase in relevant publications and information is noted.

Table 1 Activity limitations according to subtypes of CP (n=448)

Subtypes of CP	GMFCS (level)					MACS (level)				
	1 (n=130)	2 (n=115)	3 (n=61)	4 (n=74)	5 (n=68)	1 (n=141)	2 (n=150)	3 (n=78)	4 (n=61)	5 (n=18)
Spastic	109	82	53	61	51	116	121	62	42	15
Dyskinetic	9	14	4	3	5	12	6	4	11	2
Ataxic	6	3	0	3	6	5	5	4	3	1
Mixed	6	16	4	7	6	8	18	8	5	0

CP, cerebral palsy; GMFCS, Gross Motor Function Classification System; MACS, Manual Ability Classification system; n, number of participants.

Table 3 Correlation between GMFCS and MACS levels in relation to CP subtypes

CP	GMFCS–MACS agreement					
	<i>r</i> (Spearman's correlation coefficient)	Overall agreement (%)	κ coefficient	<i>z</i>	<i>P</i> value	95% CI for κ
Spastic CP	0.666	42	0.252	9.14	<0.001	0.204–2.269
Dyskinetic CP	0.528	40	0.245	3.09	0.001	0.183–0.296
Ataxic CP	0.729	50	0.318	3.10	0.001	0.127–0.429
Mixed CP	0.395	28	0.023	0.28	0.389	0–0.145
Total CP	0.639	41	0.235	9.56	<0.001	0.229–0.264

CI, confidence interval; CP, cerebral palsy; GMFCS, Gross Motor Function Classification System; MACS, Manual Ability Classification System; *P*, significance level; *r*, Spearman's correlation coefficient; *z*, differences between mean of the groups.

The highest percentage of children with CP was in level I of GMFCS and MACS with 19.29%. This result was parallel to the result of Kerem-Gunel *et al.* (2009) who found 21.1 of 185 spastic children in level I of both classifications.

Both classification systems are meant to discriminate and categorize rather than 'assess' (Damiano *et al.*, 2006). Studies have indicated that GMFCS and MACS are not only peer outcome measures, but may also complement each other for a total and complete classification of children with CP (Morris *et al.*, 2004b, 2006; Eliasson *et al.*, 2006; Kerem-Gunel *et al.*, 2009).

In this study, no children were in level V of MACS and level I–III of GMFCS or in level V of GMFCS and level I of MACS, similar to the study by Kerem-Gunel *et al.* (2009). This result was expected. Although each classification system measures different motor functions (gross and manual ability), both complement each other as outcome measures of children with CP and increasing the level from I to V indicates increasing severity of CP. This outcome is in corroboration with the content of the MACS and GMFCS, which are designed to determine the level of functional ability.

The structure of the MACS was purposely modeled on the GMFCS in that the distinctions among the levels are intended to be clinically meaningful (Palisano *et al.*, 1997). Similar to the GMFCS, the MACS will enable families, clinicians, policy makers, and researchers to communicate clearly with each other and will facilitate goal setting in clinical practice (Palisano *et al.*, 1997; Eliasson *et al.*, 2006). Researchers will be able to match children according to the MACS level, and to evaluate the various interventions designed to improve hand function (Eliasson *et al.*, 2006). MACS, in contrast, has its starting point in the upper limb function but is also influenced by environmental, personal, and contextual factors (Eliasson *et al.*, 2006). The focus of the MACS is on determining which level best represents the child's ability to handle objects and the need for assistance of adaptations to perform manual tasks in everyday settings such as at home, school and community settings and can be concluded including both 'capacity and performance' of activity (Eliasson *et al.*, 2006). The main difference between GMFCS and MACS is that MACS has a performance perspective in addition to capacity.

In another previous study, we used the Pediatric Functional Independence Measure, WeeFIM, as an outcome measure of functional independence and investigated the effects of both activity limitation and participation restriction on functional independence domains (Kerem-Gunel and Mutlu, 2007). We found statistically significant effects of activity limitation and participation restriction on functional independence. Battaglia *et al.* (2004) found a highly statistically significant correlation among Functional Independence Measure, Gross Motor Function Measure, and ICF activity and participation domains. Our study focused on outcome measures of activity limitations and did not focus on participation restriction, but the next step of this research may involve the participation restriction domain of children with CP.

The highest correlation was found in the ataxic and spastic types between GMFCS and MACS in our study. However, the ataxic type included 18 children and the low number of participants prevented us from generalizing these results for all children. In addition, the spastic type included 356 children. Kerem-Gunel *et al.* (2009) investigated the relationship between the GMFCS and MACS and indicated that there was a high correlation in 185 spastic children with CP ($r = 0.735$, $P < 0.01$). Our results corroborated with these results.

The degree of agreement was found to be statistically significant though not very high as different extremity distribution and different types exist in CP. This was expected as a child may have spastic CP and may have all four (quadriparetic) or two lower extremities (diparetic) affected. In a diparetic child, the gross motor level may be level III–V whereas manual ability is expected to be level I–II. These levels indicate that GMFCS and MACS may not have the same levels in the same children with CP.

Carnahan *et al.* (2007) examined the overall agreement among these classification systems by κ statistics and found a poor correlation between the MACS and GMFCS in 365 children with CP. Our study results were similar to those of the Carnahan study. Although we found a high correlation by the Spearman's rank correlation test, a poor relation was found by κ statistics. This may have occurred because of the characteristics of κ statistics that indicates the relation of the same scale in different studies (Altman, 1997). Hence, the correlation coefficient represents the

agreement between two different classifications. In the study by Beckung and Hagberg (2002), the correlation between GMFCS and Bimanual Fine Motor Function, a fine motor function measurement, was strong and showed that gross and fine motor functions were parallel, similar to our results.

One limitation of our study was that the cognitive level of a child might have affected the classification levels. A child's motivation and cognitive ability influence his/her ability to handle objects and thereby the classification systems and functional status. If the child's motivation to perform activities is low, or if he/she does not understand the task or continuously asks for help and support from adults, he/she should be classified according to the actual performance, even if thought to have higher capacity (Eliasson *et al.*, 2006). In our study we classified the children according to their 'actual performance' to prevent a possible classification error. Another limitation of the study might be the low number of participants in subtypes of CP except the spastic type.

Conclusion

The usage of two different classification systems, GMFCS and MACS, to describe the capacity and performance in children with CP as defined by the ICF provides an easy and quick classification tool for indicating 'activity limitations' of ICF in children with CP. The next step in research should be to highlight the other domains such as participation restrictions in these children.

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