Inter-relationships of functional status in cerebral palsy: analyzing gross motor function, manual ability, and communication function classification systems in children

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This article is commented on by Cans on page 679 of this issue.

PUBLICATION DATA
Accepted for publication 7th March 2012.
Published online 20th June 2012.

ABBREVIATIONS

AAC	Augmentative and alternative communication
CFCS	Communication Function Classification System
GMFCS	Gross Motor Function Classification System
ICF	International Classification of Functioning, Disability and Health
MACS	Manual Ability Classification System

AIM To investigate the relationships among the Gross Motor Function Classification System (GMFCS), Manual Ability Classification System (MACS), and Communication Function Classification System (CFCS) in children with cerebral palsy (CP).
METHOD Using questionnaires describing each scale, mothers reported GMFCS, MACS, and CFCS levels in 222 children with CP aged from 2 to17 years (94 females, 128 males; mean age 8y, SD 4). Children were referred from pediatric developmental/behavioral, physiatry, and child neurology clinics, in the USA, for a case–control study of the etiology of CP. Pairwise relationships among the three systems were assessed using Spearman's correlation coefficients (r_s), stratifying by age and CP topographical classifications.
RESULTS Correlations among the three functional assessments were strong or moderate. GMFCS levels were highly correlated with MACS levels (r_s=0.69) and somewhat less so with CFCS levels (r_s=0.47). MACS and CFCS were also moderately correlated (r_s=0.54). However, many combinations of functionality were found. Of the 125 possible combinations of the three five-point systems, 62 were found in these data.
INTERPRETATION Use of all three classification systems provides a more comprehensive picture of the child's function in daily life than use of any one alone. This resulting functional profile can inform both clinical and research purposes.

Many researchers and clinicians working in the field of cerebral palsy (CP) have adopted the framework of the International Classification of Functioning, Disability and Health (ICF) to inform choices of outcomes and measures.[1] Applying this framework, CP clinical practice and research go beyond describing the anatomy and physiology of individuals with CP to considering their ability to participate in daily activities. The potential interactions among these ICF components with environmental and personal contextual factors are receiving growing attention.[2–4]

This broader perspective has fostered development of classification tools to describe daily activities of mobility, handling objects, and communicating, which can be affected by CP.[5] The Gross Motor Function Classification System (GMFCS),[6] Manual Ability Classification System (MACS),[7] and Communication Function Classification System (CFCS)[8] classify mobility, handling objects, and communication respectively, at the activity/participation level of the ICF.

The purpose of this study was to describe and correlate GMFCS, MACS, and CFCS levels in a case series of children with CP. To our knowledge, this is the first study to describe a large series of children with CP who had all three classifications. We hypothesized that these classifications would not be strongly correlated. Mobility, handling objects, and communication are activities that are not functionally related, but the degree and locations of original brain injuries may overlap neural systems used in these activities. This could result in some correlations between the classifications. Understanding relationships among the three scales may be important in establishing functional profiles for children with CP.

METHOD

This case series is part of a continuing case–control study of CP etiology. Human participant approval was granted by institutional review boards at Michigan State University, Mary

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Free Bed Rehabilitation Hospital, Spectrum Health, and the University of Michigan. Informed consent was obtained from parents/legal guardians of the children.

Participants

Case eligibility was restricted to children diagnosed with CP, aged 2 to 17 years, born in Michigan, whose CP was the principal diagnosis and who had all three classifications provided by their mothers. A total of 222 children (94 females, 128 males; mean age 8y, SD 4) met the case definition. Case exclusions were for children with muscle tone abnormalities associated as an epiphenomenon to a principal diagnosis of a major malformation syndrome or genetic disorder.

This multicenter, clinic-based sample was recruited from child neurology, developmental/behavioral pediatric, or physiatry clinics in three cities in Michigan, USA: Ann Arbor, Grand Rapids, and Lansing. Physicians from these clinics treat many persons with CP from across the state. As such, any recruitment bias would tend toward enrolling more severely affected children.

Instruments

Each of the three classification systems – GMFCS,[6,9] MACS,[7,10] and CFCS[8] – has a series of five distinct but comprehensive levels described by 'word pictures', from level I (most able) to level V (least able), as shown in Table I. Reliability and content validity have been studied separately for each system. These systems were created to be used by professionals and carers without training beyond reading the classifications and their accompanying descriptions. (The classifications are available from http://www.canchild.ca/en/measures/gmfcs.asp, http://www.macs.nu, and http://www.cfcs.us respectively.)

Procedures

The following information was obtained for all children: prenatal and birth history, birth certificate, and maternal and infant hospital discharge abstracts. The child's physician, using his clinical expertise, noted CP topographical classification (regardless of motor type) as well as the presence of any associated comorbidities. These clinical diagnoses were recorded on a physician referral form. Mothers of participants provided (by mail or in person) levels of gross motor (GMFCS), manual (MACS), and communication (CFCS) function.

What this paper adds

- This is the first report of correlations among three systems for classifying everyday performance of children with CP at the activity/participation levels of the ICF.
- It identifies clusters of children with different combinations of levels of the GMFCS, MACS, and CFCS.
- It proposes clinical and research implications of functional profiles.

Analysis

Spearman's correlation coefficients (r_s) were calculated for GMFCS, MACS, and CFCS. Differences between chronological age groups and CP topographical classifications were assessed by Spearman's correlation coefficients among the three functional scales, and stratifying by three chronological age groups (up to age 5y, age 6–11y, and age 12y and older) and by CP topographical classifications (hemiplegia, diplegia, and quadriplegia).

Spearman's correlation coefficient strength was interpreted[11] as follows: $|r| \geq 0.8$ very strong relationship; $0.6 \leq |r| < 0.8$ strong relationship; $0.4 \leq |r| < 0.6$ moderate relationship; $0.2 \leq |r| < 0.4$ weak relationship; $|r| < 0.2$ very weak relationship. A probability level of $p < 0.05$ was considered statistically significant. Statistical analyses were conducted using SAS software, version 9.2 (SAS Inc., Cary. NC, USA).

RESULTS

Descriptive information from this case series, including GMFCS, MACS, and CFCS levels, is presented for 222 children with CP in Table II. At least one form of comorbidity, including seizures or impairment of cognition, hearing, speech, or vision, was reported in 146 (66%) of the children. Fifty-one (23%) of the children had two comorbidities

Table I: The five levels of GMFCS, MACS, and CFCS

	Classification systems		
Level	GMFCS	MACS	CFCS
I	Walks without limitations	Handles objects easily and successfully	Sends and receives information with familiar and unfamiliar partners effectively and efficiently
II	Walks with limitations	Handles most objects but with somewhat reduced quality and/or speed of achievement	Sends and receives information with familiar and unfamiliar partners but may need extra time
III	Walks using a hand-held mobility device	Handles objects with difficulty; needs help to prepare and/or modify activities	Sends and receives information with familiar partners effectively, but not with unfamiliar partners
IV	Self-mobility with limitations; may use powered mobility	Handles a limited selection of easily managed objects in adapted situations	Inconsistently sends and/or receives information even with familiar partners
V	Transported in a manual wheelchair	Does not handle objects and has severely limited ability to perform even simple actions	Seldom effectively sends and receives information even with familiar partners

GMFCS, Gross Motor Function Classification System; MACS, Manual Ability Classification System; CFCS, Communication Function Classification System. Adapted with permission from Hidecker et al.[8]

Table II: Characteristics of the children with cerebral palsy

Characteristics	All *n*=222 (100%)
Sex	
Female	94 (42)
Male	128 (58)
Plurality	
Singleton	182 (82)
Multiple	40 (18)
Gestational age	
≤32wks	108 (49)
>32wks	114 (51)
Age, y	
≤5	68 (31)
6–11	106 (48)
≥12	48 (21)
Comorbidities	
Cognitive impairment	57 (26)
Hearing impairment	6 (3)
Seizure	60 (27)
Speech impairment	82 (37)
Visual impairment	62 (28)

	All *n*=222 (100%)	Hemiplegia *n*=45 (20%)	Diplegia *n*=66 (30%)	Quadriplegia *n*=85 (38%)	Not given *n*=26 (12%)
GMFCS level					
I	59 (27)	20 (44)	25 (38)	8 (9)	6 (23)
II	62 (28)	14 (31)	25 (38)	16 (19)	7 (27)
III	26 (12)	4 (9)	10 (15)	9 (11)	3 (12)
IV	26 (12)	3 (7)	5 (8)	14 (17)	4 (15)
V	49 (22)	4 (9)	1 (2)	38 (45)	6 (23)
MACS level					
I	56 (25)	15 (33)	29 (44)	8 (9)	4 (15)
II	70 (32)	14 (31)	29 (44)	15 (18)	12 (46)
III	44 (20)	10 (22)	7 (11)	22 (26)	5 (19)
IV	32 (14)	4 (9)	0 (0)	23 (27)	5 (19)
V	20 (9)	2 (4)	1 (2)	17 (20)	0 (0)
CFCS level					
I	82 (37)	21 (47)	36 (55)	19 (23)	6 (23)
II	40 (18)	9 (20)	11 (17)	13 (15)	7 (27)
III	48 (22)	11 (24)	12 (18)	20 (24)	5 (19)
IV	36 (16)	4 (9)	6 (9)	19 (23)	7 (27)
V	16 (7)	0 (0)	1 (2)	14 (17)	1 (4)

Level I, most able; Level V, least able; GMFCS, Gross Motor Function Classification System; MACS, Manual Ability Classification System; CFCS, Communication Function Classification System.

reported, 20 (9%) had three comorbidities, and 10 (5%) had four comorbidities. As shown in Table II, the case series included children with functional performance from level I (most able) to level V (least able) for each scale

Spearman's correlations (r_s) were calculated between the overall classification levels and when stratified by topographical classifications of CP and child's age, as follows.

GMFCS–MACS correlations

GMFCS levels were strongly[11] correlated with MACS levels (r_s=0.69, p<0.001), and this correlation was similar across age groups. The GMFCS–MACS relationship was strongest in children with quadriplegia (r_s=0.70, p<0.001), weakest in children with diplegia (r_s=0.34, p=0.005), and moderate in strength in children with hemiplegia (r_s=0.51, p=0.004).

MACS–CFCS correlations

MACS levels were moderately[11] correlated with CFCS levels (r_s=0.54, p<0.001), and this correlation was similar across age groups. MACS levels were moderately correlated with CFCS levels in children with quadriplegia (r_s=0.58, p<0.001) and hemiplegia (r_s=0.50, p=0.005) but very weakly correlated in children with diplegia (r_s=0.19, p=0.12).

GMFCS–CFCS correlations

GMFCS levels were moderately[11] correlated with CFCS levels (r_s=0.47, p<0.001), and the correlation was weak in older children (r_s=0.31, p=0.029). A moderate GMFCS–CFCS correlation was found in children with quadriplegia (r_s=0.44, p<0.001), and a strong correlation was found in children with hemiplegia (r_s=0.69, p<0.001), but no association was seen in children with diplegia (r_s=0.07, p=0.58).

The GMFCS, MACS, and CFCS relationships do not form monotonic functions.

A nested cross-tabulation of the 125 possible combinations of the children's functional profile (i.e. GMFCS level by MACS level by CFCS level) is presented in Table III. In this case ser-

Table III: Cross-tabulations of MACS and CFCS levels within each level of GFMCS

		CFCS level					
GMFCS level I (n=59, r_s=NC)		I	II	III	IV	V	Row totals
MACS level	I	21[a]	5	5	0	0	31
	II	9	7	3	4	0	23
	III	4	1	0	0	0	5
	IV	0	0	0	0	0	0
	V	0	0	0	0	0	0
	Column totals	34	13	8	4	0	59
GMFCS level II (n=62, r_s=0.38)							
		CFCS level					
		I	II	III	IV	V	
MACS level	I	11	5	1	0	0	17
	II	11	5[a]	9	3	0	28
	III	3	3	2	3	1	12
	IV	2	0	1	1	0	4
	V	0	0	0	0	1	1
	Column totals	27	13	13	7	2	62
GMFCS level III (n=26, r_s=NC)							
		CFCS level					
		I	II	III	IV	V	
MACS level	I	4	0	0	3	0	7
	II	5	3	2	0	1	11
	III	3	1	2[a]	0	0	6
	IV	0	0	0	2	0	2
	V	0	0	0	0	0	0
	Column totals	12	4	4	5	1	26
GMFCS level IV (n=26, r_s=0.29)							
		CFCS level					
		I	II	III	IV	V	
MACS level	I	1	0	0	0	0	1
	II	1	0	3	0	0	4
	III	3	2	4	5	0	14
	IV	1	1	3	0[a]	0	5
	V	0	0	0	0	2	2
	Column totals	6	3	10	5	2	26
GMFCS level V (n=49, r_s=NC)							
		CFCS level					
		I	II	III	IV	V	
MACS level	I	0	0	0	0	0	0
	II	1	2	0	1	0	4
	III	1	2	2	2	0	7
	IV	1	2	8	7	3	21
	V	0	1	3	5	8[a]	17
	Column totals	3	7	13	15	11	49

[a]Absolute agreement counts. GMFCS, Gross Motor Function Classification System; MACS, Manual Ability Classification System; CFCS, Communication Function Classification System; NC, Spearman's correlations could not be calculated owing to row or column summing to zero.

ies, 62 (50%) of the 125 possible cells were represented by one or more children. Only 36 (16%) of the 222 children had the same classification level for all three scales, as indicated by the cells in Table III (10% were I,I,I; 2% were II,II,II; 1% were III,III,III; 0% were IV,IV,IV; and 3% were V,V,V).

In this series of 222 children, the functional profile of GMFCS level I, MACS level I, and CFCS level I was seen most often (n=21, 10%). The two profiles of GMFCS level II, MACS level I, and CFCS level I, and of GMFCS level II, MACS level II, and CFCS level I, occurred in 11 (5%) of the children. No other cell contained 5% or more of the children

DISCUSSION

The GMFCS is a well-established system to classify gross motor function, providing an easy-to-understand tool for clinicians and researchers that has a high level of interrater reliability[6] and which has been used extensively to describe study populations. However, the GMFCS does not predict functionality in domains other than mobility.[12] Complementing the GMFCS is the MACS for upper extremity function. The CFCS has been developed to describe communication skills. Taken together, these three classifications provide a view of overall functioning in a child with CP.

The GMFCS,[6] MACS,[7] and CFCS[8] were independently created and validated without expecting concordance across levels. Concordance may be highest at levels I and levels V, which may be related to the nature of ordinal systems: level I in each system represents the most functional performance, and level V represents the least functional performance. Because of obvious differences in the activities of mobility, handling objects, and communicating, no effort was made to make equivalent levels IIs, IIIs, or IVs. Instead, the nominal group and Delphi survey participants were focused on what were the underlying components of 'mild, moderate, or severe' ratings in each of the functions.[5] For example, in the CFCS development,[8] participants noted that most people use multiple methods of communication with augmentative and alternative communication (AAC) being a possible method. The person using AAC may function as a level V, IV, III, II, or I communicator. The key difference in functional communication was not the method, but related to a person being able to send and receive messages with familiar and unfamiliar partners.[8] The issue of communication pace was raised as a distinction that affected successful communication, especially when the communication partner was unfamiliar with the person.[8] These concepts related to levels of independence were then used in creating the CFCS levels. Similar processes also occurred in development of the GMFCS and MACS.[5–7]

Results of this study showed that the three classifications provided complementary information. Only 16% of the 222 children had the same classification level for all three scales. The 'all I' profile was the largest of the 125 cells but still represented only 10% of the children. The next all-one-level profile, 'all V,' included only 3% of the 222 children in the case series. For example, only one out of six children with GMFCS level V fitted an 'all V' profile with MACS and CFCS levels also being V. Of individuals who performed at the IV and V levels for MACS and GMFCS, fewer than one out of three had their communication classified as a CFCS level V. Knowing a child's classification in one system seldom predicted the child's classification in the other two systems. A next step would be to look for cell distributions in population-based samples.

Although children in GMFCS level I in this case series were generally high functioning in all areas (i.e. the GMFCS I table had no children classified at lower functioning levels for MACS and CFCS levels IV and V), a consistent functioning pattern was not noted for children in GMFCS level V (less effective mobility function). Twenty-five percent of children

with significant mobility impairment (GMFCS levels IV or V) often used some form of communication to make themselves understood to unfamiliar communication partners (CFCS levels I or II). These findings illustrate that individuals in GMFCS level V should not be assumed to have severe limitations in communication.[13]

The correlations of the GMFCS, MACS, and CFCS also supported our premise that each of the classification systems independently contributes to a description of the functional performance of children with CP. None of the correlation coefficients demonstrated a very strong relationship (r_s>0.80). This finding supports the hypothesis that use of only one system would not suffice in describing the functioning level of children with CP.

Topographical classifications did not show very strong correlations with the GMFCS, MACS, and CFCS. Mobility (GMFCS) and hand function (MACS) correlated well in children with hemiplegia and quadriplegia but were less well correlated in those with diplegia. The higher correlation between MACS and GMFCS is consistent with quadriplegia, where more extensive brain damage is likely to affect the legs, trunk, and arms. This lower correlation between MACS and GMFCS is consistent with a definition of diplegia[14] that specifies good hand function but difficulties with leg use. The CFCS had poor correlations with other measures for children with diplegia, suggesting that this group in particular is not well described by a single functional measure (see also Gorter et al.[15]).

Topographical pattern as described by the terms quadriplegia, hemiplegia, and diplegia is a classification at the level of World Health Organization (WHO) body functions, in contrast to mobility, handling object, and communicating as described by GMFCS, MACS, and CFCS levels which are at the level of WHO activities/participation. Each classification focuses on very different areas of function, controlled by different areas of the brain. GMFCS and MACS behaviors primarily involve different areas of the sensorimotor cortex; CFCS adds additional information about the function of the auditory cortex and posterior language area, including Wernicke's area and Broca's area.[16,17] Correlations of the classification systems to each other may be due to overlapping locations and amounts of the original brain injury, but do not necessarily correlate to differing functioning.

Functional profiles may be defined by an individual's GMFCS, MACS, and CFCS levels. The use of such functional profiles could improve interactions among professionals as well as with the person with CP and their families. Such information may be useful for clinical practice to direct approaches to therapy[12,18] and for public health purposes to ensure availability of appropriate services for all individuals with CP. Consideration should be given to the potential usefulness of the 125 possible combinations of GMFCS, MACS, and CFCS levels and whether clustering some of the cells together may be relevant.

Individuals with the functional profile of GMFCS level I (walks without limitation), MACS level I (handles objects easily and successfully), and CFCS level I (communicates with familiar and unfamiliar partners) are likely to function quite well in most everyday situations of family, school, community, and employment. If children who are close to this 'all I' profile (i.e. children whose performances were classified in GMFCS levels I or II, MACS levels I or II, combined with CFCS level I) are clustered with it, they could generally move around by walking, handle most objects, and have good communication with strangers. If those in CFCS level II are added to this grouping, they form a relatively high-functioning cluster.

The 'all V' functional profile (see Table I for descriptions of level Vs) describes a functional pattern that is likely to hinder participation significantly in many activities of daily life owing to limited activity and reliance on others to help with most, if not all, activities. If children who are in or close to this 'all V' profile (i.e. those whose performances were classified in GMFCS levels IV or V, MACS levels IV or V, combined with CFCS levels IV or V) are clustered, they form a relatively low-functioning cluster.

Functional profiles should consider desired outcomes or participation. Classification levels from these three systems should be considered separately and together. For example, profiles may suggest new approaches to assessing eligibility for interventions. If participation in a classroom discussion requires relatively fast communication, then the CFCS would be the central measure around which to cluster the other scales, with a research question as to how, for example, MACS levels affected response to interventions especially accessing speech-generating AAC. Spasticity intervention could be graded more on participation outcomes, suggesting a greater role for MACS and CFCS levels combined with GMFCS level in choosing the proper treatment.

The use of the three classifications together in a profile may help clinical teams think about relationships between participation and the activities of mobility, handling objects, and communication. For example, a child whose usual performance is classified in GMFCS level III, MACS level I, and CFCS level II uses crutches to walk, can handle objects easily, and talks with a speech-generating AAC system. However, the child is not an effective communicator while walking because the AAC system is not easy to carry and use while using crutches. The child may be without his AAC system unless someone else carries it for him. Another child is classified as GMFCS level IV, MACS level I, and CFCS level II, using a powered wheelchair and a speech-generating AAC system. Because the AAC system can be mounted to the wheelchair, the child can have his or her AAC system more readily available to communicate.

Limitations of this research included (1) the child's participation in daily activity not being measured, (2) the lack of generalizability of results as this sample was from a clinical case series, not a population-based sample, and (3) the small sample size relative to 125 possible combinations of GMFCS, MACS, and CFCS levels. Data and tables showing distributions (including percentages) of children across the three scales were based on a case series and should be interpreted cautiously. The distribution of children across the three scales may differ in population-based and other case series studies. Additional

research is needed to understand parent–professional agreement on MACS and CFCS levels. Future research could relate these functional performance profiles to activities and participation measures. For example, the GMFCS might inform us about fitness and cardiac challenges as individuals with more functional GMFCS levels generally have fewer barriers to exercise; whereas individuals with less functional GMFCS levels often need adapted exercises or equipment to perform an exercise regimen.[19] The MACS level can suggest a person's ability to use a keyboard, which is a necessary component of many jobs and leisure activities. Employment may depend particularly on effective communicative function, especially in times when manual labor is being replaced by service-oriented jobs requiring rapid and accurate communication skills.

Population-based studies of these functional profiles would be useful for public health planning and for considering the long-term effects on health status and community involvement. The three systems may prove useful as predictors for success in various domains of activity and participation. So far, only the GMFCS has been validated as a predictive tool.[20] Both the MACS and CFCS are more recently developed classification tools, and their predictive validity has not yet been explored. With 125 possible combinations of GMFCS, MACS, and CFCS levels, larger sample sizes across age ranges would aid in understanding whether all of these combinations occur in the population with CP and whether more variations occur in certain age groups such as young children. Parent–professional agreement on functional levels needs further research to understand reasons for differing GMFCS, MACS, and/or CFCS judgments of parents and professionals. Future research should also compare individuals' functional profiles with their quality of life, desires, and participation.[13]

The WHO ICF framework being adopted by professionals working with individuals with CP has revolutionized research and clinical practices by expanding assessment beyond problems in anatomy and physiology to include daily activities, participation, and contextual factors. The three classification systems considered here provide information about functional performance that complements classical neurological and topographical descriptions of CP.[21] For example, body structure and function such as magnetic resonance imaging results could be correlated to each of the functional profiles. In addition, the effects of comorbidities such as cognitive ability, speech impairment, and seizure on functional profiles could be examined. The use of functional profiles of GMFCS, MACS, and CFCS levels could help professionals, family members, and persons with CP better consider the roles of mobility, handling objects, and communication within participation in meaningful life activities.

ACKNOWLEDGMENTS

This research was supported in part by National Institutes of Health NINDS grant R01NS055101 to NP. We acknowledge the work of Julie Wirth, Ariel Brovont, Alice Stanley, Zach Dyme, David van Dyke, Andrea Kuldanek, and Connie James in this research.

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