

Investigation of Differences Between the Representatives of the High-to Moderate Functional Status According to Upper Limb Functions and Participation in Children with Congenital Hemiplegic Cerebral Palsy: A Cross-Sectional Study

Konjenital Hemiplejik Serebral Palsili Çocuklarda Üst Ekstremitte Fonksiyonlarına ve Katılıma Göre Yüksek-Orta Fonksiyonel Durumun Temsilcileri Arasındaki Farkların İncelenmesi: Kesitsel Bir Çalışma

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ÖZ

Bu çalışmanın amacı, sınıflandırma sistemleri bağlamında Serebral Palsili (SP) bireylerde el becerisi ve katılım performansı kazanımlarını araştırmaktır. Başka bir anlatımla, SP'li bireylerde fonksiyonel durumun üç temsilcisinin el becerisi ve katılım sonuçlarına göre farklılık gösterip göstermediğini görmek amaçlandı. Çalışmaya SP tanısı konulmuş 71 kişi (ortalama 12.4 SS 2.3 yıl; 38 erkek ve 33 kız) alındı. Çalışma katılımcılarının fonksiyonel durumları El Becerileri Sınıflandırma Sistemi (EBSS), Kaba Motor Fonksiyon Sınıflandırma Sistemi-Genişletilmiş&Düzenlenmiş (KMFSS-G&D) ve İletişim Becerileri Sınıflandırma Sistemi (İFSS) kullanılarak belirlendi. El becerisi ve katılım sonuçları, sırasıyla ABILHAND-Kids anketi ve Çocuk ve Adölesan Katılım Ölçeği (CASP) kullanılarak belgelendi. Post-hoc testleri, yüksek EBSS ve KMFSS-G&D seviyelerine sahip bireylerin, düşük MACS ve GMFCS-E&R seviyelerine sahip kişilere kıyasla ABILHAND-Kids ve CASP'ta daha yüksek puanlar aldıklarını göstermiştir (I>II>III, P<0.0001). Benzer şekilde, İFSS I'deki bireyler hem ABILHAND-Kids hem de CASP'ta İFSS III' tekilere kıyasla daha iyi puan aldılar (I>III, P<0.001). Aksine, İFSS I-II veya İFSS II-III olarak sınıflandırılan bireylerde ABILHAND-Kids ve CASP skorları benzerdi (P>0.05). Son olarak, çoklu regresyon analizleri EBSS ve KMFSS-G&D'nin el becerisini (EBSS: %65; KMFSS-G&D: %23) ve katılım sonuçlarını (EBSS: %46-%62; KMFSS-G&D: %57-%68) güçlü bir şekilde öngördüğünü ortaya koydu. EBSS ve KMFSS-G&D'den elde edilen bilgiler, SP'li bireylerde el becerisi ve katılım sonuçları hakkında fikir verebilir.

Anahtar Kelimeler: Serebral palsy, hemipleji, spastik, konjenital, fonksiyonel durum

ABSTRACT

This study aimed to explore the acquisition of manual ability and participation performance in individuals with cerebral palsy (CP) in the context of classification systems. In other words, the aim was to determine whether the three representatives of functional status in individuals with CP differed according to manual ability and participation outcomes. Seventy-one individuals (mean 12.4 SD 2.3 years; 38 boys and 33 girls) diagnosed with CP were enrolled. The functional status of the study participants was defined using the Manual Ability Classification System (MACS), Gross Motor Function Classification System Expanded & Revised (GMFCS-E&R), and Communication Function Classification System (CFCS). Manual ability and participation outcomes were documented using the ABILHAND-Kids and Child and Adolescent Scales of Participation (CASP), respectively. Post-hoc tests demonstrated that individuals with high MACS and GMFCS-E&R levels had higher scores on the ABILHAND-Kids and CASP than those with low MACS and GMFCS-E&R levels (I>II>III, P<0.0001). Likewise, individuals in CFCS I scored better on both the ABILHAND-Kids and CASP than those in CFCS III (I>III, P<0.001). In contrast, ABILHAND-Kids and CASP scores were similar in individuals classified as CFCS I-II or CFCS II-III (P>0.05). Finally, multiple regression analyses revealed that MACS and GMFCS-E&R strongly predicted manual ability (MACS:65%; GMFCS-E&R: 23%) and participation outcomes (MACS: 46%-62%; GMFCS-E&R: 57%-68%). Knowledge from MACS and GMFCS-E&R can provide insight into the manual ability and participation outcomes of individuals with CP.

Keywords: Cerebral palsy, hemiplegia, spastic, congenital, functional status

The study protocol was approved by the Ethical Board at Muş Alparslan University (approval number: 26.11.2021-30767)

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INTRODUCTION

Assessing individuals with unilateral or hemiplegic Cerebral Palsy (CP) is important in documenting available problems associated with body functions, activity, and participation in different life situations (1). Prior systematic reviews have demonstrated that there are a variety of measures or assessment tools to describe body structures, body functions, activity, and participation of individuals with hemiplegic CP (2). However, the use of these measures is commonly ineffective because of the amount of time needed for their application in clinical environments. Therefore, classification tools have been developed to succinctly describe the functional status of individuals with hemiplegic CP and (3), in turn, enhance communication between healthcare professionals and parents in clinical settings.

The Gross Motor Function Classification System (GMFCS), The Manual Ability Classification System (MACS), and the Communication Function Classification System (CFCS) are the commonly utilized classification tools in pediatric area (4,5). MACS has been created to characterize a child's capacity to manage daily life activities (6). GMFCS was created to differentiate individuals with CP in respect of gross motor function (7). Hence, using MACS and GMFCS simultaneously at particular time point ensures a broad range of knowledge regarding functional motor performance of individuals with CP (1). Subsequently, the CFCS concentrates on categorizing the communication performance of individuals with CP concerning their ability to communicate with both familiar and unfamiliar individuals in their daily lives (8). As a result, MACS, GMFCS, and CFCS are used to classify the manual ability, gross motor function, and communication skills of individuals with CP by providing an overview of their abilities in everyday life (9). A priori research has illustrated that these classification tools complement each other in depicting upper limb and gross motor functions, as well as communication performance (10). Associations between MACS, CFCS, and GMFCS have previously been demonstrated to be excellent to moderate in individuals with CP aged 2 to 7 years (11). Likewise, a more recent study by Compagnone et al. revealed a good association between GMFCS, MACS, and CFCS tools (10).

A common perspective on validation in classification tools is their alignment with an external reference (6). Thus, it is very important to establish what each level of the classification tool stands for in real-life situations by using outcome measures. That is, the validation of each level of classification tool within the context of actual performance in everyday life is fundamental for health professionals. Within this aim, some studies have demonstrated potential disparities between GMFCS and MACS levels in terms of mobility, self-care, and fine motor skills (6,12-13). Despite their valuable findings, these studies included children from various CP types, which limited a more in-depth exploration of the distinctions between high and moderate levels of classification tools. Therefore, it is essential to distinguish between high and moderate levels of classification tools in relation to manual ability and participation in daily activities. As a result, although there have been prior attempts to investigate variations among classification tool levels, the existing literature is inadequate, and no prior study has focused on the actual distinctions among three high-to-moderate functional status representatives in the context of daily life performance. This study aimed to identify the acquisition of manual ability and participation in individuals with hemiplegic CP in relation to functional classification systems.

MATERIAL VE METHODS

This research employs a cross-sectional design, involving data collection at a single point in time. A convenience sample of 71 individuals with hemiplegic cerebral palsy was determined through a threshold of 1.02 units of change, which exhibited statistical significance in the ABILHAND-Kids data from a prior study (14), using G* Power v.3.1. The participants in the study were between the ages of 9 and 17 years (mean age 12.4 ±2.3; 38 males, 33 females) and

had been diagnosed with hemiplegic CP born in the city of Muş. A flow diagram illustrating the recruitment process is shown in Figure 1.

Ethical Aspect of The Study

The study protocol was approved by the Ethical Board at Muş Alparslan University (approval number: 26.11.2021-30767). All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. Written permission was obtained from the parents after thorough information was provided. The functional status of the participants in relation to the classification tools was described by a physiotherapist with eight years of experience in pediatric rehabilitation. Individuals with low functional levels (IV-V) or a diagnosis of acquired brain injury were excluded.

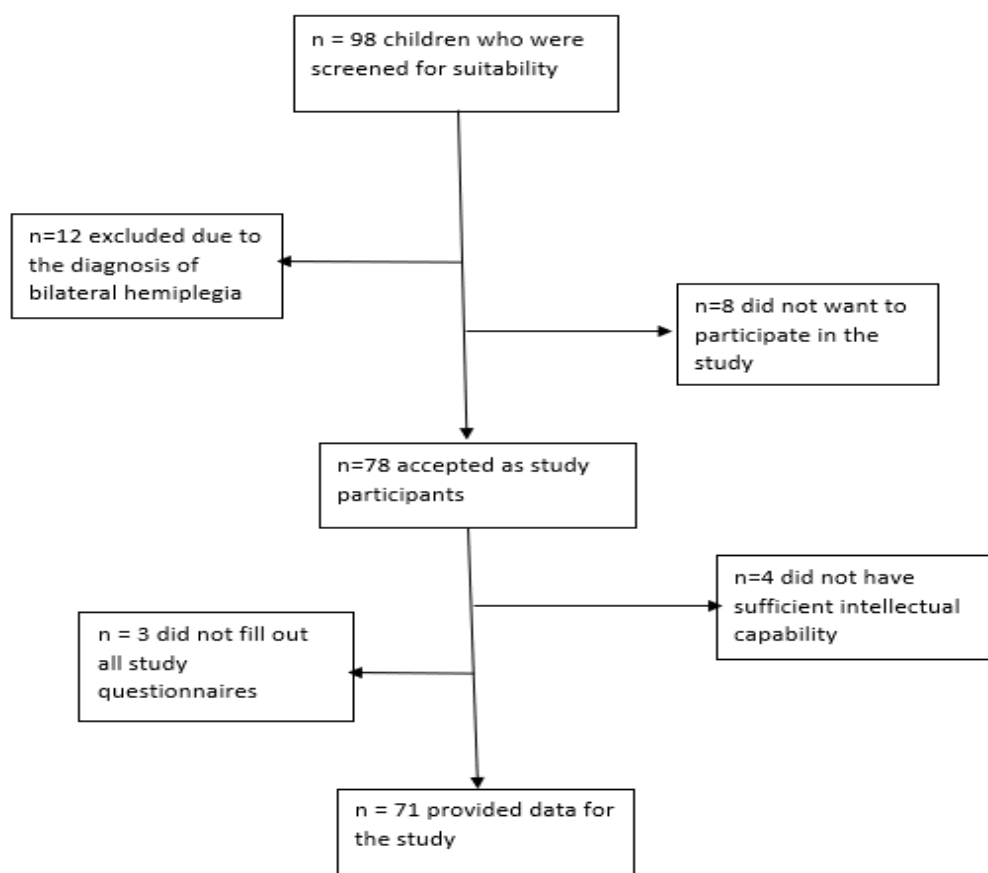


Figure 1. Flow diagram of enrolling participants in the study

The functional status of the study participants was defined using the MACS (test-retest reliability; inter-class correlation coefficient [ICC]=0.91–0.98) (15), GMFCS (intra-rater reliability; generalizability coefficient [G]= 0,79) (16), and CFCS (inter-rater reliability; weighted kappa= 0.82) (8). In this study, upper extremity functions were evaluated in the context of manual ability using the ABILHAND-Kids. The ABILHAND-Kids is a Rasch-based instrument developed to assess the perceived manual ability of individuals with CP aged 6 to 15 years (17). The ABILHAND-Kids comprises 21 items that inquire about a range of daily activities that necessitate the use of one or both hands (14). The possible superiority of this measure is its utility in clinical or research environments because it is quick to complete (17).

The mean score of ABILHAND-Kids is calculated online by transforming the raw score into a logit metric using the Rehab-Scales website (18). Eventually, ABILHAND-Kids was demonstrated to have good stability (ICC=0.98) in assessing the manual ability of Turkish individuals with CP (19). The Child and Adolescent Scale of Participation (CASP) was employed to assess the level of a child's participation in various life situations, encompassing their participation in activities at home, school, and within the community (21). The CASP comprises 20 ordinal-scaled items that inquire about various aspects of participation, including 6 items related to home participation, 4 items concerning community participation, 5 items related to school participation, and an additional 5 items related to home and community living activities. Its items are suitable for evaluating participation outcomes in school-aged children (five years or older) and are aligned with a wide range of daily life activities. The CASP has been validated in Turkish, and its psychometric properties have been demonstrated to be robust among Turkish children with various disabilities (test-retest reliability: ICC=0.95) (21). Statistical analyses were conducted using IBM SPSS version 24 (SPSS Statistics for Windows, Version 23.0. IBM Corporation Armonk, NY). The study data were analyzed using both visual methods (histograms and probability plots) and analytical techniques (such as the Kolmogorov-Smirnov test) to determine their distribution normality. Categorical variables were described using percentages or frequencies, whereas numerical data were represented as mean \pm standard deviation. "One-way ANOVA and Tukey's test were conducted to examine potential differences among MACS, GMFCS, and CFCS levels concerning manual ability and participation outcomes. A univariate linear regression model was used to independently investigate the extent to which the variables affected manual ability and participation outcomes. Subsequently, a multiple linear regression analysis was performed to explore the collective influence of MACS, GMFCS, and CFCS on manual ability and participation. Level III served as the reference category (coded as 0). Statistical significance was defined as a p-value less than 0.05.

Limitation

Although a homogeneous sample was one of the strengths of this study, it might also be a drawback. Because the functional profiles of participants changed between levels I and III, our data could not incorporate individuals at a low functional level (IV-V). This study was restricted to individuals diagnosed with hemiplegic CP however, it can be expanded to include various types of CP. Therefore, it is necessary to conduct additional research to examine the differences in manual ability and participation outcomes across all levels of classification systems.

RESULTS AND DISCUSSION

The demographic and clinical information of the individuals were summarized in Table I.

Table 1. Demographic Features and Descriptive Data of the Individuals

Variables	N=71
Gender n (%)	
Male	38 (53.5)
Female	33 (46.5)
Age (y)	
Mean \pm SD	12.4 \pm 2.3
Age range	9-17
Affected Side n (%)	
Right	33 (46.5)
Left	38 (53.5)

Comorbidity			n	%
Cognitive impairment	Yes		6	8.5
	No		65	91.5
Hearing impairment	Yes		0	0
	No		71	100
Speech impairment	Yes		3	4.2
	No		68	95.8
Visual impairment	Yes		2	2.8
	No		69	97.2
MACS Level n (%)				
I	24 (33.8)			
II	19 (26.8)			
III	28 (39.4)			
GMFCS Level n (%)				
I	26 (36.6)			
II	24 (33.8)			
III	21 (29.6)			
CFCS Level n (%)				
I	25 (35.2)			
II	22 (31)			
III	24 (33.8)			

X: mean, SD: Standard Deviation, CASP: Child and Adolescent Scale of Participation, MACS: Manual Ability Classification System, GMFCS: Gross Motor Function Classification System, CFCS: Communication Function Classification System.

One-way ANOVA results indicated significant statistical differences among MACS levels concerning both manual ability and participation outcomes. Subsequently, Tukey's post-hoc test revealed that individuals with higher MACS levels exhibited superior manual ability and participation outcomes compared to those with lower MACS levels (I>II>III; P=0.000) (Table II). Furthermore, individuals in MACS I exhibited a higher degree of variability in their mean score of manual ability than individuals in MACS II and III.

Table 2. Manual Ability and Participation Outcomes by MACS Levels

		MACS									
		I ^{a*} (n=24)			II ^{b*} (n=19)			III ^{c*} (n=28)			P
		X	SD	%95 CI	X	SD	%95 CI	X	SD	%95 CI	
CASP	Home Participation	92,7	10,0	88.8- 96.5	77,6	9,5	73.9- 81.2	60,0	12,0	56.2- 63.7	0,000
	Community Participation	87,9	13,6	82.6- 93.1	72,6	11,1	68.3- 76.8	58,2	14,7	53.5- 62.8	0,000
	School Participation	87,3	13,2	82.1- 92.4	71,2	8,9	67.8- 74.5	54,8	13,2	50.6- 58.9	0,000
	Home and Community Living Activities	75,9	14,5	70.2- 81.5	63,4	13,3	58.3- 68.4	45,2	12,5	41.2- 49.1	0,000
	ABILHAND-Kids (logits)	3,9	1,1	3.4- 4.3	2,2	0,5	2.0- 2.3	1,2	0,8	0.9- 1.4	0,000

CASP: Child and Adolescent Scale of Participation, MACS: Manual Ability Classification System, X: Mean, SD: Standard Deviation, *: Post-hoc, a>b>c; n, no

The manual abilities and participation outcomes of the children in GMFCS I, II, and III showed statistically significant differences ($p=0,000$) (Table III). Furthermore, Tukey’s post-hoc test demonstrated that individuals with a high GMFCS level achieved significantly better outcomes in terms of both manual ability and participation than those with a low GMFCS level (I>II>III).

Table 3. Manual Ability and Participation Outcomes by GMFCS Levels

		GMFCS									P
		I ^{a*} (n=26)			II ^{b*} (n=24)			III ^{c*} (n=21)			
		X	SD	%95 CI	X	SD	%95 CI	X	SD	%95 CI	
CASP	Home Participation	82,6	15,7	78.3-86.8	67,6	12,2	63.5-71.6	45,8	7,5	37.9-53.6	0,000
	Community Participation	81,1	14,4	77.1-85.0	61,5	12,2	57.4-65.5	39,6	9,4	29.7-49.4	0,000
	School Participation	78,2	15,6	73.9-82.4	60,9	11,6	57.0-64.7	35,8	7,4	28.0-43.5	0,000
	Home and Community Living Activities	67,9	17,6	63.0-72.7	51,6	12,5	47.4-55.7	31,7	6,1	25.2-38.1	0,000
	ABILHAND-Kids (logits)	2,8	1,5	2.3-3.2	1,8	0,8	1.5-2.0	0,3	0,2	0.0-0.5	0,000

CASP: Child and Adolescent Scale of Participation, GMFCS: Gross Motor Function Classification System, X: Mean, SD: Standard Deviation, *: Post hoc: a>b>c; n, number of participants

Table IV summarizes the manual ability and participation outcomes of the study participants according to CFCS levels. Using Tukey's test for paired comparisons, the analysis revealed a statistically significant difference between CFCS levels I and III ($P=0.000$), whereas no significant differences were observed between CFCS levels I and II ($P>0.05$) or between CFCS levels II and III ($P>0.05$) (I>III, I=II, II=III).

Table 4. Manual Ability and Participation Outcomes by CFCS Levels

		CFCS									P
		I ^{a*} (n=25)			II ^{b*} (n=22)			III ^{c*} (n=24)			
		X	SD	%95 CI	X	SD	%95 CI	X	SD	%95 CI	
CASP	Home Participation	80,7	16,0	76.3-85.0	72,1	13,9	66.8-77.3	55,6	14,5	47.2-63.9	0,000
	Community Participation	79,4	14,6	75.4-83.3	65,6	15,2	59.8-71.3	49,0	13,6	41.14-56.8	0,000
	School Participation	76,1	15,8	71.8-80.3	65,5	15,4	59.6-71.3	47,9	13,4	40.1-55.6	0,000
	Home and Community Living Activities	65,8	18,2	60.8-70.7	56,4	14,6	50.8-61.9	40,4	11,8	33.5-47.2	0,000
ABILHAND-Kids (logits)		2,5	1,5	2.0-2.9	2,2	1,1	1.7-2.6	1,2	0,9	0.6-1.7	0,007

CASP: Child and Adolescent Scale of Participation, CFCS: Communication Function Classification System, X: Mean, SD: Standard Deviation, Post-hoc: *: a>c, a=b, b=c, X: mean, n: number of participants

Table V presents the results of the regression analysis. Specifically, MACS, GMFCS, and CFCS explained 65%, 23%, and 8% of the total ABILHAND-Kids score, respectively. Furthermore, MACS emerged as the most influential factor in determining participation in different life contexts, such as family, school, and community, explaining 62%, 56%, and 48% of the variability in the respective subtest scores. Moreover, MACS was identified as an equivalent determinant of GMFCS for participation in the community environment, particularly where maintaining an adequate level of mobility is crucial (MACS: 68% GMFCS of variance; 68% of variance).

Table 5. Regression Analyses on manual ability, Participation in Different Situations, MACS, GMFCS, and CFCS.

		Univariate Analysis						Multivariate analysis		
		R	R ²	df	F	t	p-value	B	Adjusted R ²	P-value
ABILHAND-Kids	MACS	0.8	0.65	1	179.505	13.398	0.000	1.363	0.65	0.000
	GMFCS	0.48	0.23	1	28.998	5.385	0.000	1.105		
	CFCS	0.29	0.08	1	9.186	3.031	0.003	0.56		
CASP-Home	MACS	0.78	0.62	1	158.113	12.574	0.000	16.425	0.71	0.000
	GMFCS	0.59	0.34	1	51.375	7.168	0.000	16.693		
	CFCS	0.49	0.24	1	30.258	5.501	0.000	11.611		
CASP-Community	MACS	0.68	0.46	1	83.048	9.113	0.000	14.827	0.68	0.000
	GMFCS	0.68	0.46	1	83.64	9.145	0.000	20.171		
	CFCS	0.6	0.36	1	54.035	7.351s	0.000	14.883		
CASP-School Participation	MACS	0.74	0.56	1	122.846	11.084	0.000	16.292	0.84	0.000
	GMFCS	0.65	0.42	1	70.619	8.404	0.000	19.223		
	CFCS	0.53	0.28	1	38.786	6.228	0.000	13.288		
CASP-Home and Community Living Activities	MACS	0.69	0.48	1	90.725	9.525	0.000	15.505	0.58	0.000
	GMFCS	0.57	0.32	1	46.319	6.806	0.000	17.231		
	CFCS	0.47	0.22	1	27.437	5.238	0.000	11.946		

MACS: Manual Ability Classification System, GMFCS: Gross Motor Function Classification System, CFCS: Communication Function Classification system, R: Correlation Coefficient, R²: Explained Variance (%), df: degrees of freedom.

We found statistically significant differences in manual ability and participation outcomes between MACS levels I, II, and III, as well as between GMFCS levels I, II, and III. Individuals who were classified as having high functioning levels on MACS and GMFCS were more likely to be able to manage daily activities and achieve better participation outcomes. Regarding CFCS, only individuals classified as levels I and III showed a statistically significant difference with respect to manual ability and participation outcomes. In contrast, individuals with similar CFCS levels, that is, classified as CFCS I, II, II, and III, did not differ in manual ability and

participation outcomes. Finally, the study results showed that manual ability, as described by MACS, was the most important factor influencing manual ability and participation outcomes.

MACS, GMFCS, and CFCS tools were established to classify upper limb functions/manual ability, mobility level, and communication skills, respectively, at a particular time point (10,22-23). The differences between GMFCS, MACS, and CFCS levels with respect to external references (e.g., activity and participation) have been examined in various studies (6,12,23). However, all earlier studies included heterogeneous samples; therefore, they did not focus on the differences among the three levels representing high to moderate functional status in the context of manual ability and participation outcomes.

Our study demonstrated statistically significant differences among MACS and GMFCS levels I-III as to manual ability and participation outcomes between MACS and GMFCS levels I-III, apart from CFCS. From this aspect, our work is separate from previous studies (6,12) by concentrating on examining differences among three levels representing high to moderate functional status with respect to manual ability and participation outcomes. Our findings have therefore provided initial data regarding the differences between the three functional status representatives in terms of manual ability and participation outcomes. As a result, the evident differences between the MACS and GMFCS levels suggest that they are beneficial in predicting the actual performance of children with hemiplegic CP in daily life. As for CFCS, the results showed that manual ability and participation scores vary only between levels I and III. However, these results show that improved communication ability is necessary for daily independence.

MACS emerged as the most robust predictor of manual and participation outcomes. Additionally, the variance in participation in the community score was shown to be equally explained by the MACS and GMFCS tools, and the GMFCS was revealed to be a second significant predictor of the remaining outcomes. As a result, MACS and GMFCS tools worked together to predict manual ability and participation outcomes. As a result, classification systems can be used by researchers, parents, and healthcare professionals as concise languages to assess manual ability and participation outcomes in individuals with CP.

CONCLUSION AND SUGGESTIONS

This study is the first to focus on differences among three representatives of functional status, using ABILHAND-Kids and the CASP as external references. The study results showed meaningful differences between all three MACS and GMFCS levels with respect to upper manual ability and participation outcomes. Although a significant difference was found between CFCS I and III scores, no statistically significant differences were found between scores I, II, II, and III. In addition, MACS was the strongest determinant of manual ability and participation outcomes. Thus, MACS, GMFCS, and CFCS can be effectively utilized in both clinical and research settings to describe the functional capabilities of children with hemiplegic CP and present a concise picture of their current functional status.

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