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The association between motor capacity and motor performance in school-aged children with cerebral palsy: An observational study

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ABSTRACT

Background: This study aimed to investigate the association between motor capacity and motor performance in children with cerebral palsy (CP) aged 6–12 years with Gross Motor Function Classification System (GMFCS) levels I to III.

Methods: Forty-six children with CP (24 boys and 22 girls) classified as GMFCS levels I, II, or III were included. Motor capacity was measured by the Gross motor function measure (GMFM), Pediatric balance scale (PBS), Timed up and go (TUG), and 6-min walk test (6MWT). Motor performance was measured by triaxial accelerometers. Estimations of physical activity energy expenditure (PAEE) (kcal/kg/day), percentage of time spent on physical activity (% sedentary physical activity; %SPA; % light physical activity, %LPA; % moderate physical activity, %MPA; % vigorous physical activity %VPA; and moderate-to-vigorous physical activity, %MVPA), and activity counts (counts/minute) were obtained.

Results: Children with GMFCS level I showed a significantly higher motor capacity (GMFM-66, GMFM-88, D-dimension and E-dimension, PBS and 6MWT) than those with level II or III. Children with GMFCS level II and/or III had significantly lower physical activity (PAEE, % MPA, % VPA, %MVPA, and activity counts) than children with GMFCS level I. Multiple linear regression analysis (dependent variable, GMFM-66) showed that %MVPA was positively associated with GMFM-66 in the GMFCS level II & III children but not in GMFCS level I children.

Conclusions: These findings highlight the importance of increasing %MVPA in children with CP, especially GMFCS levels II and III.

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1. Introduction

Cerebral palsy (CP) refers to a group of disorders that affect movement and muscle tone or posture accompanied by disturbances of sensation, perception, cognition, communication, and behavior, by epilepsy, and by secondary musculoskeletal problems.¹ These impairments limit activities and restrict participation.^{1,2} According to the “International Classification of Functioning, Disability and Health: Children and Youth version” developed by

the World Health Organization (WHO), function and disability in children has two components as follows: 1) body functions and structures, and 2) activities and participation.³ Activity and participation construct capacity and performance; capacity indicates functioning in a standardized environment, while performance indicates functioning in the current environment.

Physical activity (PA) is represented by behaviors defined as “any bodily movement produced by skeletal muscles that requires energy expenditure.”⁴ The WHO defined the term habitual physical activity (HPA) as “bodily movement produced regularly by the contraction of skeletal muscles that results in a substantial increase over resting energy expenditure.”⁵ Participation in daily activities during childhood is important for maintaining the health of children, regardless of their disability. Children with CP participate in fewer PAs than their normally developed peers. In the past decades,

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Abbreviations

Cerebral palsy (CP)
 Gross Motor Function Classification System (GMFCS)
 Gross motor function measure (GMFM)
 Light physical activity (LPA)
 Minute walk test (MWT)
 Moderate physical activity (MPA)
 Moderate-to-vigorous physical activity (MVPA)
 Pediatric balance scale (PBS)
 Physical activity energy expenditure (PAEE)
 Sedentary physical activity (SPA)
 Timed up and go (TUG)
 Vigorous physical activity (VPA)

studies on the PA of children with CP have employed various questionnaires that can be perceived as subjective methods of assessment.⁶ Recently, accelerometers (StepWatch or ActiGraph) have been introduced to assess PAs in the pediatric populations.^{7–10} They can objectively register one or many dimensions of body-movement.

In the general population, the sedentary behavior present during childhood and adolescence persists throughout adulthood.¹¹ A systematic review suggested that early childhood (0–6 years) is a critical age when children with CP adopt either an active or a sedentary lifestyle.¹² Keawutan et al. reported that HPA in Gross Motor Functional Classification System (GMFCS) levels I and II remained stable with age.¹³ However, in children over 5 years old with GMFCS levels III–V, HPA significantly decreased with age.¹³ Children with CP are expected to achieve approximately 90% of their potential Gross Motor Function Measure (GMFM)-66 before the age of 6 years.¹⁴ Functional declines have been observed before 8 years of age in children at GMFCS levels III–V.¹⁵ Therefore, the PA level and its correlation with motor capacity in school-aged children with CP should be observed and adjusted accordingly to prepare them for the transition to healthy adolescence and adulthood. However, studies on PA in school-aged children with CP have not been reported as much as those in other age groups. Moreover, most of the studies on activities and participation in children with CP have been performed in the western countries. The results of these studies may not clearly represent the characteristics of children with CP in Asian countries, where there is a strong tradition of prioritizing education over participation in PA.

Researchers and clinicians have tried to provide objective data by studying “what children can do (capacity).” However, capacity measured in a clinical setting is different from “what children choose to do or actually do (performance).” The relation between what ambulatory children with CP are capable of doing in a clinical environment and their capability in daily life is mediated through their regular activity performance.¹⁶ Measuring HPA with accelerometers in children with CP is expected to provide objective information regarding real-life activity. Further, it could clearly explain the relationship between both motor capacity and performance representing the characteristics of children with CP.

This study aimed to investigate the association between motor capacity and HPA measured using accelerometers in children aged 6–12 years with CP and at GMFCS level of I–III.

2. Methods

2.1. Study design

This cross-sectional study was conducted in a tertiary care hospital located in Seoul. The Institutional Review Board of the Samsung Medical Center (2017–06–045) approved the protocol. Written informed consent was provided by the participants as well as their parents or guardians before the screening. The study was performed between August 2017 and December 2019.

2.2. Participants

Children satisfying the inclusion criteria were identified using the hospital database. In the case of children whose parents agreed to participation in the study on a telephone call, screening was conducted by the primary investigator.

Forty-seven children with CP participated in this study. Since one child declined to participate following screening, 46 children were included in the analysis (Table 1). The inclusion criteria were as follows: (a) diagnosis of CP, (b) classified at GMFCS level I, II, or III, (c) age between 6 and 12 years, and (d) body weight under 35 kg. In contrast, the exclusion criteria were as follows: (a) injection botulinum toxin in the last 3 months, (b) a selective dorsal rhizotomy or orthopedic surgery in the last 1 year, (c) poor visual acuity, (d) hearing impairment, (e) severe intellectual disability, (f) uncontrolled seizures, (g) hip dislocation, (h) Cobb angle > 30° in scoliosis, and (i) unhealed fracture.

2.2.1. Motor capacity

Two experienced pediatric physical therapists administered the GMFM, Pediatric Balance Scale (PBS), and Timed Up and Go test (TUG). Two experienced exercise researchers conducted the 6-min walk test (6MWT). All the participants were evaluated within 10 days.

2.2.2. GMFM

GMFM-88 is a formalized method for assessing gross motor function in children with CP.¹⁷ It constitutes 88 items in the following five dimensions: (a) lying and rolling, (b) sitting, (c) crawling and kneeling, (d) standing, and (e) walking, running, and jumping. The GMFM-66 scores were calculated from the GMFM-88 using the Gross Motor Ability Estimator. Rasch analysis of the 88-item GMFM helped us identify 66 of the original 88 GMFM items that met the assumptions of unidimensionality, and the test- and sample-free measurements. The reliability values of GMFM can be used very consistently in children with CP (intraclass correlation coefficient [ICC] = 0.99).¹⁷

2.2.3. PBS

The PBS is a common reference-based measure for evaluating the functional balance in life activities.¹⁸ This scale has been validated as a good test for children with CP.¹⁹

2.2.4. TUG

The TUG test is a simple test used in clinical practice to assess functional ambulatory mobility or dynamic balance. The TUG has demonstrated a test-retest reliability (ICC = 0.99) in children with CP.²⁰

Table 1
Descriptive characteristics of children with CP.

	GMFCS I (n = 21)	GMFCS II (n = 19)	GMFCS III (n = 6)	All (n = 46)
Boys/Girls (n)	12/9	9/10	3/3	24/22
Bilateral/unilateral (n)	5/16	16/3	6/0	27/19
Age (years)	7.48 ± 1.75	7.32 ± 1.45	8.00 ± 1.26	7.48 ± 1.56
Height (cm)	125.02 ± 10.91	122.53 ± 9.57	122.33 ± 10.23	123.64 ± 10.14
Weight (kg)	24.84 ± 4.92	24.19 ± 5.78	25.92 ± 6.26	24.71 ± 5.37

Values are expressed as mean ± SD.

GMFCS, Gross Motor Function Classification System.

2.2.5. 6MWT

6MWT is used to assess a child's aerobic capacity. Children were instructed to walk as fast as possible for 6 min on a flat hard-surfaced straight corridor. The total distance walked during 6MWT was recorded in meters. The 6MWT has demonstrated test-retest reliability for school-aged children with CP (ICC = 0.98).²¹

2.3. Motor performance

2.3.1. HPA

HPA was measured using an ActiGraph (GT3X) triaxial accelerometer monitor (model GT3X, ActiGraph, Pensacola, FL, USA). Epochs were set at 5 s to detect activity.

The children wore accelerometers during daily life, except while bathing and swimming. It was worn on the lower back continuously for a week.²² Their parents maintained an activity diary, which recorded the activity type and activity time during the wearing period. If the device was not worn, the reason for it was recorded.

Activity data were downloaded and analyzed using ActiLife software (version 6.9.0; ActiGraph LLC Pensacola, FL, USA). Data collected during the 5-h wear time on at least four consecutive days, were analyzed.²³ The data recorded in the ActiGraph was compared directly to the information in the activity diary to confirm the activity while wearing the accelerometer. The participants were expected to wear the device for a minimum of 4 days out of 7, with the monitoring period ranging within 600 min per day (10 a.m.–8 p.m.). We obtained an estimated physical activity energy expenditure (PAEE) in kcal/kg/day, percentage of time spent in each level of physical activity (% sedentary physical activity, % SPA; % light physical activity, % LPA; % moderate physical activity, % MPA; % vigorous physical activity % VPA; and % moderate-to-vigorous physical activity, % MVPA), and activity counts (counts/minute).

Estimated physical activity energy expenditure in kcal/kg/day was calculated with the Freedson model, inherently present in the ActiLife software.²⁴ To classify physical activity intensity, we used Evenson's cut-off points,²⁵ which are validated for use in children and adolescents with CP.²⁶ Consecutive epochs of zero counts or ≤100 counts per minute were registered as sedentary activity, 101–2295 counts per minute as light activity, 2296–4011 counts per minute as moderate activity, and ≥4012 counts per minute as vigorous activity. The output of the ActiGraph is vector magnitude/time called activity counts (counts per time), which have been reported to have excellent inter-instrument reliability in ambulant young people with CP (ICC = 0.981).²⁷

2.4. Statistical analysis

Analyses were performed with SPSS version 20.0 (IBM, Armonk, NY, USA), using a 0.05 level of significance for all statistical tests. All continuous variables were tested for normality using the Shapiro–Wilk descriptive test. Means and standard deviations are

presented for each continuous variable. Differences in motor capacity and motor performance between-GMFCS groups were investigated using a one-way ANOVA with a Bonferroni post hoc test. A bivariate Spearman correlation analysis was performed to assess the relationship between motor capacity (GMFM-66, GMFM-88, D-dimension, and E-dimension) and motor performance (PAEE, % SPA, % LPA, % MPA, % VPA, % MVPA, and activity counts). Univariate regression analyses were performed to examine the contribution of demographic factors (age and sex) and motor performance to motor capacity (GMFM-66). GMFM-66 was chosen as the dependent variable because it satisfies the assumption of one-dimensionality. Then, multiple stepwise regression analysis was performed to assess the relationship between motor capacity (dependent variable GMFM-66) and motor performance (PAEE, % SPA, and % MVPA).

3. Results

Children at GMFCS level I showed a significantly higher motor capacity (GMFM-66, GMFM-88, D-dimension and E-dimension, PBS and 6MWT) than those at level II or III (Table 2). Further, children at level II had a significantly higher motor capacity (GMFM-66, GMFM-88, D-dimension and E-dimension, PBS, TUG, and 6MWT) than those at GMFCS level III. TUG was significantly lower in children with GMFCS III than in children at levels I and II ($p < .001$).

Acquisition and analysis of HPA were performed for all children without adverse effects related to wearing accelerometers (7-day monitoring in 32 children, 6-day monitoring in 9 children, 5-day monitoring in 3 children, and 4-day monitoring in 2 children). The accelerometers of two children were exposed to underwater activity, and a data over only 4 days were analyzed for them. All the participants in this study wore accelerometers over the minimum required period (over 5 h on 4 consecutive days). The average wearing period was 6.54 (standard deviation = 0.80) days with no significant difference according to the GMFCS level.

% SPA and % LPA were not significantly different across the GMFCS levels. Children who were classified at GMFCS level III had significantly lower physical activity (PAEE, % MPA, % VPA, %MVPA, and activity counts) than children classified at GMFCS level I. Further activity counts at GMFCS III were significantly lower than those at GMFCS I and II (Table 3).

3.1. Relationship between motor capacity and motor performance

The % SPA was negatively associated with GMFM-66, GMFM-88, D-dimension, E-dimension, and PBS; conversely, it was positively associated with TUG (Supplementary Table 1). PAEE, % MPA, % VPA, % MVPA, and activity counts showed low to moderate correlation with GMFM-66, GMFM-88, D-dimension, E-dimension, PBS, and TUG. 6MWT showed a low correlation with PAEE, % VPA, and % MVPA.

Table 2
Motor capacity of children with CP according to GMFCS levels.

	GMFCS I (n = 21)	GMFCS II (n = 19)	GMFCS III (n = 6)	F	p
GMFM88 (%)	98.87	94.05	79.89	91.224	.000
	1.10	3.15 ^a	±6.24 ^{b,c}		
GMFM66 (%)	91.56	75.12	60.76	65.002	.000
	8.22	±4.51 ^a	± ^{b,c}		
Dimension D (%)	97.80	91.23	68.38	47.103	.000
	2.47	±5.08 ^a	±15.86 ^{b,c}		
Dimension E (%)	96.63	79.75	37.04	104.034	.000
	±3.47	±10.97 ^a	±14.48 ^{b,c}		
PBS (score)	54.57	50.05	28.83	43.612	.000
	±1.43	±4.91	± ^c		
TUG (s)	7.22	9.16	45.20	35.372	.000
	±1.16	±2.25 ^d	±35.66 ^e		
6MWT (m)	419.43	329.37	190.50	32.921	.000
	±48.49	±75.94 ^a	±67.24 ^{b,c}		

Values are expressed as mean ± SD.

GMFCS, Gross Motor Function Classification System; GMFM, gross motor function measure; 6MWT, 6-min walk test; TUG, timed up and go; PBS, pediatric balance scale; using one-way ANOVA with Bonferroni post-hoc test; ^a, I>II; ^b, I>III; ^c, II>III; ^d, I<III; ^e, II<III; p < .001.

Table 3
Motor performance of children with CP according to GMFCS levels.

	GMFCS I (n = 21)	GMFCS II (n = 19)	GMFCS III (n = 6)	F	p
Wear time (day)	6.47	6.73	6.16	1.284	.287
	±0.81	±0.65	±1.16		
PAEE (kcal/kg/day)	5.00	3.86	3.00	8.805	.001
	±1.29	±1.09 ^a	±.92 ^b		
% SPA	70.26	72.37	76.44	2.932	.064
	±5.69	±4.86	±7.44		
% LPA	23.59	23.14	20.66	.911	.410
	±4.59	±4.11	±6.80		
% MPA	3.91	3.16	2.14	6.553	.003
	±1.28	±.96	±.79 ^b		
% VPA	2.22	1.31	.74	7.874	.001
	±1.22	±.66 ^a	±.28 ^b		
% in MVPA	6.14	4.47	2.88	7.555	.002
	±2.43	±1.58 ^a	±1.02 ^b		
Activity counts (counts/min)	983.70	865.00	550.96	9.526	.000
	±232.70	±216.48	±108.29 ^{b,c}		

Values are expressed as mean ± SD.

GMFCS, Gross Motor Function Classification System; PAEE, physical activity energy expenditure; % SPA, percentage of sedentary physical activity time per wear time; % LPA, percentage of light physical activity time per wear time; % MPA, percentage of moderate physical activity time per wear time; % VPA, percentage of vigorous physical activity time per wear time; % MVPA, percentage of moderate to vigorous physical activity time per wear time; using one-way ANOVA with Bonferroni post-hoc test; a, I>II, p<.05; b, I>III, p<.01; c, II>III, p < .05.

Table 4
Results of multivariable regression analyses to examine the contribution of PAEE, %SPA, and %MVPA to GMFM66 in children with CP.

Subject	Independent variables	beta	SE	95% CI for beta	p-value	R ²
GMFCS I-III	PAEE	3.429	1.759	(-0.120, 6.978)	0.058	0.344
	%SPA	0.366	0.370	(-0.381, 1.114)	0.328	
	%MVPA	2.355	1.046	(0.243, 4.467)	0.030*	
GMFCS I	PAEE	1.001	2.319	(-3.892, 5.894)	0.672	0.064
	%SPA	-0.109	0.426	(-1.008, 0.790)	0.801	
	%MVPA	0.392	1.358	(-2.474, 3.258)	0.777	
GMFCS II & III	PAEE	2.784	1.813	(-0.987, 6.554)	0.140	0.307
	%SPA	0.678	0.400	(-0.154, 1.510)	0.105	
	%MVPA	3.237	1.040	(1.074, 5.401)	0.005**	

GMFM66, gross motor function measure-66; GMFCS, Gross Motor Function Classification System; PAEE, physical activity energy expenditure; % SPA, percentage of sedentary physical activity time per wear time; % MVPA, percentage of moderate to vigorous physical activity time per wear time; beta, regression coefficient, SE, standard error of beta; CI, confidence interval; multiple stepwise regression analysis was performed; *, p < .05; **, p < .01

3.2. Multiple linear regression analyses

Multiple stepwise regression analysis (dependent variables GMFM-66) showed that % MVPA was positively associated with GMFM-66 in children at GMFCS levels II and III, unlike children at GMFCS level I (Table 4). Results of the univariate regression analysis are provided in Supplementary Table 2.

4. Discussion

This study investigated the relationship between the motor capacity and motor performance in Asian school-aged children with CP and GMFCS I-III. We found that % MVPA is related to motor capacity in children with CP classified at GMFCS levels II and III. HPA measured by accelerometers can explain GMFM in children with CP.

Further, HPA measurement using accelerometers can be used in studies on activity and participation in children with CP.

Children with CP were reported to spend more than half of their active hours in a sedentary state. Keawutan et al. reported that children (5 years) classified at GMFCS levels I–II and GMFCS levels III–V spent 57.7% and 85.2% of their time in a sedentary state, respectively.¹³ In our study in children with CP (6–12 years), the value of %SPA was 70.26% at GMFCS level I, 72.37% at GMFCS level II, and 76.44% at GMFCS level III. These values were higher than those observed by Keawutan et al.¹³; however, our participants were older, and different cut-points (820 counts per minute in Keawutan study¹³ vs. 100 counts per minute in our study) were used for sedentary time. The results seem to reflect socio-cultural characteristics that emphasize education rather than participation in physical activity.

According to the “Physical Activity Guidelines for Americans, 2nd edition,” children and adolescents aged 6–17 years were recommended to engage in more than 60 min of MVPA daily.²⁸ Children with CP spent more than half of their active hours sedentarily, similar to our results (70–72%). In this study, the daily time spent for MVPA was 36.84 ± 14.58 min at GMFCS level I, 26.84 ± 9.50 min at GMFCS level II, and 17.34 ± 6.18 min at GMFCS level III. This shows that children with CP spend far less time on MVPA than the guideline recommendations. The children (9–18 years) performing VPA, especially when continuous, significantly predicted the VPA of their adulthood.²⁹ In this context, our results indicate that children with CP and at GMFCS levels I–III, aged between 6 and 12 years should increase their physical activity according to the guidelines. Although CP is a “non-progressive” condition, obesity and muscle loss due to habitual sedentary behavior can increase the severity of dysfunction in adulthood and can lead to cardiometabolic disease, fragility, and/or premature mortality.²⁹

So far, there has been no consensus on the cut-off points for classifying MVPAs in children and adolescents due to the lack of standardization when collecting and processing data. Until now, researchers have used different cut-off points to classify PA levels. In this study, we used Evenson’s cut-off points, which validated PA, including MVPA, in children with typical development (5–8 years). However, researchers should consider that the LPA and MVPA could be misclassified when applying the cut-off points published in accelerometer data collected from children with CP.³⁰

High HPA levels of children with CP were associated with a high level of motor capacity.³¹ GMFM, one of the representative factors of motor capacity, is strongly correlated with HPA in children with CP (2–17 years). One study reported significant associations between motor capacity (GMFM-66) and the Activity Scale for Kids (ASKp-30) in ambulatory children with CP (2–9 years).¹⁶ Another study reported that walking, running, and jumping activity (GMFM-E) were the best predictors regarding energy expenditure, step number, and sedentary time.³² In our study, motor capacity showed a mild to moderate correlation with HPA (PAEE, %MPA, %VPA, %MVPA, and activity counts), which was similar to those of previous studies. In this study, we aimed to elucidate the variables of HPA that predicts motor function (GMFM66). This was achieved since GMFM66 was predicted by %MVPA in children with GMFCS II and III.

The major limitation of this study was the small number of children at GMFCS levels III ($n = 6$). GMFCS levels II and III were subsequently grouped for multivariate analysis as suggested by the statisticians. Nevertheless, further studies are warranted to demonstrate the relationship between motor capacity and motor performance in children with other neuromotor disorders, as well as those at GMFCS levels IV and V.

Another limitation is that the exact value of %HPA could not be obtained due to motor impairment and abnormal muscle tone

caused by CP.³³ Other potential limitations include the inability to use the employed methods during water-based PA, such as swimming and the possibility of some light PAs, such as sitting when the trunk is immobile (when standing or riding a bicycle), being misinterpreted as sedentary. Further, it takes considerable effort and motivation for the child’s parents to ensure that the accelerometer is placed on the child for the requisite.³³

5. Conclusion

The present study investigated and analyzed the association between motor capacity and HPA in ambulatory children aged 6–12 years with CP (GMFCS level I, II, and III). Findings from this study demonstrate that MVPA is important to GMFCS level II and III.

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Author statement

Min-Hwa Suk: Data curation; Formal analysis; Investigation; Visualization; Roles/Writing - original draft, In-Kyeong Park: Data curation, Investigation, Soojin Yoo: Conceptualization; Methodology; Writing - review & editing, Jeong-Yi Kwon: Conceptualization; Funding acquisition; Investigation; Methodology; Supervision; Validation; Visualization; Roles/Writing - original draft; Writing - review & editing.

Declaration of competing interest

The Author declare that there is no conflict of interest.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.jesf.2021.07.002>.

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