Title: Relationship between habitual physical activity, motor capacity, and capability in children with cerebral palsy aged 4-5 years across all functional abilities

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Disclosures

The authors declare no conflict of interest.

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Abstract

Background: Children with cerebral palsy (CP) have lower habitual physical activity (HPA) than their typically developing peers. There are limited studies of HPA in young children with CP under the age of 5 years.

Objective: To investigate the relationships between HPA, sedentary time, motor capacity and capability in children with CP aged 4-5 years.

Methods: Sixty-seven participants were classified using Gross Motor Function Classification System (GMFCS), assessed for motor capacity using Gross Motor Function Measure (GMFM) and wore accelerometers for three days to measure HPA and sedentary time. Motor capability was assessed using parent-reported Pediatric Evaluation of Disability Inventory (PEDI) functional skills of mobility domain. Mixed-effects regression models were used for analyses.

Results: GMFM was positively associated with HPA (mean difference (MD)=19.6 counts/min; 95%CI=16.6 to 22.7, $p<0.001$) and negatively associated with sedentary time (MD=-0.6%; 95%CI=-0.7 to -0.5, $p<0.001$). The PEDI was also positively associated with HPA (MD=16.0 counts/min; 95%CI=13.1 to 18.8, $p<0.001$) and negatively associated with sedentary time (MD=-0.5%; 95%CI=-0.6 to -0.4, $p<0.001$). After stratification for ambulatory status, GMFM and PEDI were associated with HPA and sedentary time in ambulant participants but not in non-ambulant participants.

Conclusions: Gross motor capacity and motor capability are related to HPA and sedentary time in ambulant children with CP aged 4-5 years.

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Introduction

According to the International Classification of Functioning, Disability and Health: Children and Youth version (ICF-CY), activity and participation components contain two constructs, capacity and performance (1). Capacity is defined as what a person can do in a standardized, controlled environment (2). Performance refers to what a person actually does do in his/her environment (2). As environmental factors are one of contextual factors that impact on activity and participation (1), capability could be another structure that can impact on a person's ability. Capability is defined as what a person can do in his/her environment (2).

Habitual physical activity (HPA) is one of the performance behaviors that has many potential health benefits such as improved bone health, cardiorespiratory and muscular fitness (3, 4). Habitual physical activity (HPA) refers to any bodily movement in daily life which results in energy expenditure (5). Sedentary behavior is a major global health problem associated with a number of conditions including cardiovascular disease and diabetes (4). Sedentary behavior is defined as any activity using energy expenditure ≤ 1.5 metabolic equivalents such as lying, sitting and reclining (6).

Previous studies tracking physical activity and sedentary behavior in the general population reported that HPA behavior in childhood and adolescence can remain stable until adulthood (7, 8). A systematic review suggested that early childhood (0-6 years) is a critical period for carry-over of an active or sedentary lifestyle (9). Consequently, it is important to understand the level of physical activity in young children, including children with disabilities, in order to modify behavior at an early age to prevent detrimental outcomes including cardiovascular and metabolic diseases in adulthood.

Cerebral palsy (CP) is a group of disorders of movement and posture causing activity limitations (10). Gross motor function of children with CP can be classified by the Gross

Motor Function Classification System (GMFCS) into five levels from level I; walking without restriction, to children classified as level V whom are dependent or utilized powered wheeled-mobility (11). Body function and structure impairments, activity limitations, and participation restrictions in children with CP can impact on their HPA (12-16). A systematic review reported that children with CP aged 5-18 years had 13-53% less HPA than their peers and twice the maximum recommended sedentary time (13). Previous studies found that ambulant children with CP (GMFCS I-III) aged 6-10 years had less HPA and more sedentary time than children with typical development (14, 15). In addition, ambulant youth with CP aged 8-17 years have been reported to spend more time sedentary than their peers (16). Reduced levels of HPA and increased sedentary time were associated with elevated blood pressure in children with CP aged 6-17 years (17) and increased risk of developing cardiometabolic disease in adults with CP aged 18-62 years (18). Almost all previous studies were conducted in ambulant children with CP (GMFCS I-III) at school-age children and in adolescents (6-18 years). There are limited studies regarding HPA in young children with CP. A previous study has been conducted in toddlers with CP age 1.5-3 years in which it was reported that HPA and sedentary time in toddlers with CP classified as GMFCS I-II were not different from toddlers with typical development (19). Active and sedentary time was found to differ between toddlers without CP and toddlers with CP classified as GMFCS III-V.[19] Another study in children with CP aged 4-5 years found that ambulant children with CP (GMFCS I-II) had significantly higher HPA and lower sedentary time than non-ambulant children with CP (GMFCS IV-V) (20). A longitudinal study in young children with CP reported that HPA levels started to decline from 4 years of age and sedentary time significantly increased at the age of 4 years (21).

Regarding the relationship between motor capacity and HPA, previous studies in ambulant children with CP found that the Gross Motor Function Measure (GMFM) correlated

with HPA (22) and the GMFM dimension E (walking, running and jumping) were important predictors of HPA in adolescents and young adults with CP (23). A systematic review confirmed that motor capacity was directly related with HPA in children with CP but there are limited studies using objective measure of HPA in non-ambulant children with CP at age less than 5 years (24).

A previous study examined the relationship between motor capacity, capability, and performance in children with CP aged 2.5 years (2). The study reported that although there were high correlations between motor capacity, capability and performance, motor performance is partly reflected by motor capacity and motor capability (2). Motor capability in children with CP can be measured using parent-reported questionnaire, the Pediatric Evaluation of Disability Inventory (PEDI) functional skills of mobility domain (2). The PEDI consists of three separate sets of measurement scales including functional skills, caregiver assistance, and modifications (25). Each scale contains three domains which are self-care, mobility and social function (25). It is important to obtain an objective measure of HPA and sedentary performance in young children with CP and examine relationships to motor capacity and parent-reported motor capability. The aim of this study is to investigate the relationship between HPA, sedentary time, motor capacity, and capability in children with CP aged 4-5 years across all functional abilities.

Methods

This cross-sectional study was conducted in Queensland, Australia between October 2010 and December 2014. Data are derived from two population-based cohort studies, the Queensland CP Child Study of Motor Function and Brain Development (n=227) (26) and the Queensland CP Child Study of Growth, Nutrition and Physical Activity (n=175) (27).

Queensland children who were born in 2006-2009 and have a diagnosis of CP were eligible for the studies. Children with progressive disorders were excluded.

The CP Child Study of Motor Function and Brain Development assessed children every 6 months from 18 to 36 months corrected age, and then at 48 and 60 months corrected age. The Queensland CP Child Study of Growth, Nutrition, and Physical Activity assessed children at 3 time points depending on study entry, which was 17 to 25 months, 36 months and 60 months corrected age with additional assessment at 48 months for those who entered to the study after 25 months corrected age. This present study included participants from those two cohort studies who were assessed at 48 and 60 months of age. Ethics were approved by the University of Queensland Medical Research Ethics Committee (2008002260) and regional hospitals across Queensland, Australia. Informed consent was signed by all parents or legal guardians of participants.

Outcome measures and procedures

Participants were classified using the GMFCS into five levels: level I, independent walking without restriction; level II, independent walking with limitations on an uneven surface; level III, walking with an assistive device; level IV, limited self-mobility or use of power mobility; level V, were dependent or utilized powered wheeled-mobility (11).

Motor capacity

Participants were assessed for motor capacity (what a child can do in a structured environment) using the 66-item GMFM by a research physiotherapist. The GMFM is a standardized criterion-referenced measure which assesses motor capacity in children with CP over 5 dimensions (A: lying/rolling, B: sitting, C: crawling/kneeling, D: standing and E: walking/running/jumping). It contains 66 items; each item is scored in 4-point ordinal scales from 0 (does not initiate) to 3 (completed activity) (28).

Motor capability

Parents of participants completed the 59-item Pediatric Evaluation of Disability Inventory (PEDI) functional skills of mobility domain to determine motor capability (activities the child can do in a natural environment). The PEDI was scored either capable to do (1) or unable to do (0) for each item. The raw score was converted to scaled score from 0-100 (25).

Motor performance

This study measured motor performance (what a child actually does do in his/her environment) (2) using an objective measure of HPA. Participants wore the ActiGraph® accelerometer centered at their lower back (L2) for all waking activities except water-based activities for at least three days (two weekdays and one weekend) (29). Reasons for wearing the monitor at the lower back were to avoid limitations of participants' movement and to minimize the influence of asymmetrical gait movement in some participants (30, 31). Wearing an accelerometer at lower back and hip are not significantly different for detecting activity counts (31). Corresponding activity diaries which were completed by parents of participants contained the time when the child woke up, when the monitor was put on/taken off, reasons for taking off the monitor, when the child was being carried or pushed in pram, and sleep time. This study used the ActiGraph® triaxial accelerometer (GT3X and GT3X+) which detected acceleration of the body in three planes, vertical (X), anteroposterior (Y) and mediolateral (Z). Habitual physical activity was indicated by activity counts (count per epoch of time) which were calculated from vector magnitude ($VM=\sqrt{X^2+Y^2+Z^2}$). The monitor was set at 5 second-epoch to detect short bursts of physical activity in children with CP. Activity data were downloaded via ActiLife software® (Actigraph, FL, USA). Wear time periods were checked with activity diaries and non-wear time periods were deleted from analyses. The non-wear time was only when the ActiGraph® was not attached to a child. The period that a child was carried or transported in car was not deleted. This period was recorded as

sedentary time. Any ambiguous data were clarified with the parents. Each day was manually filtered for non-were time. Wear time period (hours), activity counts (counts per minute) and sedentary time as a percentage of wear time of each participant were calculated using MATLAB (The MathWorks Inc., version R2012b). Time spent sedentary was determined using the cut-point for sedentary time of 820 counts per minute (32) which was validated in children with CP aged 4-5 years in a previous study (33).

Accelerometer cut-points for sedentary time in children with CP aged 4-5 years across all functional abilities have been validated against direct observation, a criterion measure (33). The cut-points for each GMFCS level were derived using Receiver Operating Characteristic (ROC) curves. The cut-points that derived from each GMFCS level and the previously established cut-point from children with typical development (32) were applied in an independent sample of children with CP for cross-validation. Bland-Altman agreement statistics were calculated to compare predictive validity. Results support the use of the previously established cut-point for sedentary time of 820 counts per minute (32) in a group basis for all GMFCS levels (33).

Statistical analysis

Based on prior knowledge we expected our sample size of 67 individuals to complete approximately 80 assessments. We calculated we would be able to detect a difference of 150 counts per minute or greater between GMFCS levels with 80% power and alpha=0.05 (G*Power Version 3.1.9.2).

Characteristics of participants who were included and excluded from this study were compared by independent t-test (continuous variables) and Fisher's exact test (categorical variables). Mixed-effects regression models, with child included as a random effect were used to investigate differences of physical activity data between GMFCS levels (GMFCS level I as a reference group) and relationships between HPA, sedentary time, motor capacity

and capability. The GMFCS level, GMFM and PEDI score were independent variables while activity counts and sedentary time were dependent variables. All statistical analyses were performed using Stata® v13.0 (StataCorp, College Station, TX, USA). Statistical significance was set at alpha=0.05.

Results

Participants

Two hundred and ten assessments were conducted in 158 children with CP aged 4-5 years across Queensland, Australia. Ninety-one children were excluded because of incomplete activity data (2-day monitoring in 13 children, 1-day monitoring in 3 children and 0-day monitoring in 75 children). Reasons for not wearing the activity monitor were rejection from participants and inability of parents to attach the monitor to their child. Total participants with sufficient data were 67 children with 84 assessments, mean age 4.9 years. Characteristics of included and excluded participants were not significantly different in age, sex, and GMFM score. Characteristics of included participants were 43 (64%) boys; unilateral spasticity, n=30 (45%); bilateral spasticity, n=30 (45%); dystonia, n=5 (7%); ataxia, n=1 (1%); and hypotonia, n=1 (1%).

Motor performance

Physical activity data in each GMFCS level are shown in Table 1. Wear time of the activity monitor were not significantly different between GMFCS levels. Activity counts in children with CP classified as GMFCS II-V were significantly lower than GMFCS I. Sedentary time as a percentage of wear time in children with CP classified as GMFCS I and II were not significant difference while children with CP classified as GMFCS III-V had significantly higher sedentary time than GMFCS I.

Relationships between motor performance, motor capacity, and capability

Separate regression analyses in all participants showed that both the GMFM and PEDI were associated with activity counts and sedentary time (Table 2). Regression analyses according to ambulatory status found that the relationships in children with GMFCS I-III were the same as in all participants. In children with GMFCS IV-V, neither the GMFM nor the PEDI was associated with the physical activity data (Table 2).

Discussion

Activity counts significantly decreased and sedentary time significantly increased when GMFCS levels increase, except for the sedentary time between GMFCS level I and II. High motor capacity (GMFM) and capability (PEDI) are associated with high HPA levels and low sedentary time in children with CP aged 4-5 years. Both motor capacity and motor capability contribute to HPA and sedentary behavior in ambulant children with CP (GMFCS I-III). Although motor capacity and capability are associated with activity performance, a previous longitudinal study suggested that “change in motor capacity does not automatically translate to change in motor capability and change in motor capability does not automatically translate to change in motor performance” (34). In addition, there are many factors to consider including access to physical activity opportunities, environmental barriers and child and family motivation to engage in physical activity. In non-ambulant children with CP (GMFCS IV-V), motor capacity and capability did not contribute to HPA and sedentary time. Some physical activities that require energy expenditure in non-ambulant children with CP such as rolling or moving the upper extremities may not be able to be measured accurately as HPA using a body worn accelerometer. These findings suggest that using an accelerometer to measure HPA and sedentary time may not cover all physical activities in non-ambulant children with CP.

Facilitators and barriers for participating in physical activity for children and adolescent with CP have been identified. Various personal and environmental factors impact their ability to participate in physical activity such as experiences, enjoyment, parental awareness of benefits of physical activity, pain, fatigue, lack of opportunities for sport and physical activity (35). A previous study in preschool children with typical development found that parental participation in physical activity is a mediator of their children's physical activity participation (36). An active family of children with CP may promote their children to be active as well. Fatigue has been identified as a personal barrier to participation in physical activity (35). Previous studies reported that fatigue was associated with lower levels of physical activity in children, adolescents (37), and adults with CP (38, 39). Increases in physical activity have been recommended for people with CP to prevent and reduce fatigue (37-39).

Physical activity data are rarely available for children with CP classified as GMFCS IV-V. A strength of this study is that it has provided HPA and sedentary time in non-ambulant children with CP using an objective measurement. Nevertheless, the interpretation of the results for those who are non-ambulant may be incomplete. As the HPA monitor may be unable to detect or may miss classify some physical activities of non-ambulant children with CP. Additional placement and validation of HPA monitors may be required to detect activity of the upper limbs and movements with in the base of support for non-ambulant children with CP.

It is a challenge to attach an activity monitor to young children with CP. A potential limitation of this study was a small number of participants in the non-ambulant group. Also, there were a large amount of missing data which suggest that the ActiGraph® may not be appropriate as a surveillance measure. Measurement of physical activity using an accelerometer requires considerable effort and motivation by the child's parent to ensure it is

worn. In addition, accelerometers may be appropriate for use as an outcome measure following intervention.

The ActiGraph® has some specific limitations in that it cannot be worn to measure water-based activities and some light activities may be miss-classified as sedentary activities where the trunk is not moving for example when bike riding and standing to perform activities at a table. The position of the monitor (at centered lower back of the participant adjacent to the bodies centre of mass) may be less accurate for measuring HPA in non-ambulant children with CP as some physical activity such as moving the upper extremities during sitting would be classified as sedentary time.

Conclusion

Gross motor capacity and motor capability are associated with HPA and sedentary behavior in ambulant children with CP (GMFCS I-III) aged 4-5 years, however are not associated in non-ambulant children with CP (GMFCS IV-V).

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Table 2 Mixed-effects regression models of gross motor function (GMFM) and motor capability (PEDI) on activity counts and sedentary time as a percentage of wear time

	Independent variables	**Activity counts (counts/minute)**		**Sedentary time (% of wear time)**	
		MD (95% CI)	*p*-value	MD (95% CI)	*p*-value
All participants (n=84)	GMFM	19.6 (16.6, 22.7)	<0.001	-0.6 (-0.7, -0.5)	<0.001
	PEDI	16.0 (13.1, 18.8)	<0.001	-0.5 (-0.6, -0.4)	<0.001
GMFCS I-III (n=66)	GMFM	17.4 (10.4, 24.4)	<0.001	-0.4 (-0.6, -0.2)	<0.001
	PEDI	11.8 (6.1, 17.6)	<0.001	-0.3 (-0.5, -0.2)	<0.001
GMFCS IV-V (n=18)	GMFM	8.4 (-1.1, 17.9)	0.083	-0.2 (-0.6, 0.1)	0.199
	PEDI	2.7 (-3.5, 8.9)	0.391	-0.02 (-0.2, 0.2)	0.836

Key: GMFCS, gross motor function classification system; MD, mean difference; PEDI, pediatric evaluation of disability inventory

Table 1 Physical activity data in children with CP according to gross motor function classification system (GMFCS) level

GMFCS	N (%)	Wear time (hour)			Activity counts (counts/min)			Sedentary time (% of wear time)		
		Mean (SD)	MD (95%CI)	*p*-value	Mean (SD)	MD (95%CI)	*p*-value	Mean (SD)	MD (95%CI)	*p*-value
I	48 (57)	10.6 (1.4)	Reference group		1388 (367)	Reference group		56.1 (8.7)	Reference group	
II	9 (11)	10.9 (1.3)	0.2 (-0.8, 1.3)	0.69	1017 (186)	-274 (-488, -59)	0.012	64.3 (6.6)	4.9 (-0.5, 10.2)	0.08
III	9 (11)	10.9 (1.4)	0.3 (-0.9, 1.5)	0.61	838 (422)	-573 (-819, -327)	<0.001	72.9 (11.9)	17.2 (11.0, 23.5)	<0.001
IV	4 (5)	10.1 (0.9)	-0.7 (-2.4, 1.0)	0.41	469 (172)	-933 (-1290, -576)	<0.001	85.4 (5.2)	29.5 (20.4, 38.6)	<0.001
V	14 (17)	10.4 (2.3)	-0.3 (-1.3, 0.7)	0.52	154 (144)	-1216 (-1421, -1011)	<0.001	94.5 (5.4)	37.6 (32.4, 42.8)	<0.001

Key: MD, mean difference; SD, standard deviation; Mixed-effects regression models