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Physical activity in non-ambulatory toddlers with cerebral palsy

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ABSTRACT

Background: Children with cerebral palsy are less likely to be physically active than their peers, however there is limited evidence regarding self-initiated physical activity in toddlers who are not able, or who may never be able, to walk.

Aims: The aim of this study was to measure self-initiated physical activity and its relationship to gross motor function and participation in non-ambulatory toddlers with cerebral palsy.

Methods and procedures: Participants were between the ages of 1–3 years. Physical activity during independent floor-play at home was recorded using a wearable tri-axial accelerometer worn on the child's thigh. The Gross Motor Function Measure-66 and the Child Engagement in Daily Life, a parent-reported questionnaire of participation, were administered.

Outcomes and results: Data were analyzed from the twenty participants who recorded at least 90 min of floor-play (mean: 229 min), resulting in 4598 total floor-play minutes. The relationship between physical activity and gross motor function was not statistically significant ($r = 0.20$; $p = 0.39$), nor were the relationships between physical activity and participation ($r = 0.05 - 0.09$; $p = 0.71 - 0.84$).

Conclusions and implications: The results suggest physical activity during floor-play is not related to gross motor function or participation in non-ambulatory toddlers with cerebral palsy. Clinicians and researchers should independently measure physical activity, gross motor function, and participation.

What this paper adds?

This paper adds to the current literature by reporting the magnitude of self-initiated physical activity during floor-play time in non-ambulatory toddlers with cerebral palsy. To date, physical activity has been measured only from full-day recordings in this patient population. By measuring physical activity during independent floor-play time, this study presents an alternative method which may isolate self-initiated movement, and be less influenced by caregiver and family activity. Additionally, this paper presents evidence that physical activity, gross motor function, and participation are not related constructs in non-ambulatory toddlers with cerebral palsy.

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1. Introduction

Cerebral palsy (CP) is the most common physical disability in children (Pakula, Van Naarden Braun, & Yeargin-Allsopp, 2009). Half of all children with cerebral palsy additionally have an intellectual disability; one in four have a language disorder, and one in four have a behavioral disorder (Novak, Hines, Goldsmith, & Barclay, 2012). Despite advances in pre- and perinatal care, the prevalence of CP has remained stable at 2–3.5 per 1000 births over recent decades (Colver, Fairhurst, & Pharoah, 2014; Kirby et al., 2011). CP is characterized by impairment in the development of movement and posture resulting from early brain injury (Rosenbaum et al., 2007). The degree of physical disability varies widely from limitations in balance and coordination to full dependence on others for care (Palisano et al., 1997). Regardless of the severity of physical limitation, physical activity is an important contributor to health for all people (Cooper et al., 1999; Johnson, 2009). Low levels of physical activity in individuals with CP may lead to poor aerobic and anaerobic fitness which could increase the risk of chronic diseases over time (Maltais, Wiart, Fowler, Verschuren, & Damiano, 2014).

Children with developmental disabilities are less likely to be physically active than their peers with typical development, with those with CP demonstrating 13–53% less habitual physical activity (Carlon, Taylor, Dodd, & Shields, 2013; Maltais et al., 2014; Ryan, Forde, Hussey, & Gormley, 2015). Much of the physical activity research in individuals with CP has examined activity in ambulatory school-age children (Bania, 2014; Bjornson, Belza, Kartin, Logsdon, & McLaughlin, 2007; Bjornson, Zhou, Stevenson, & Christakis, 2014; Ryan et al., 2015). Bjornson et al. in two studies, reported that children with CP ages 10–14 and 2–9 years old who have greater functional limitations have lower levels of physical activity (Bjornson et al., 2007, 2014). Similarly, Keawutan et al. concluded in a systematic review that motor capacity is related to habitual physical activity in children with CP ages 2–17 years (Keawutan, Bell, Davies, & Boyd, 2014).

There is little evidence reporting the physical activity of young children with developmental disabilities who are not yet able, or may never have the ability, to walk. In typically-developing children ages 0–5 years, physical activity has been favorably associated with motor development, fitness, and bone health (Carson et al., 2017; Fisher et al., 2005), but most typically-developing toddlers are able to walk by the age of 1 year or soon after, and most evidence reports stepping behavior data from this population as well (Avery, Russell, Raina, Walter, & Rosenbaum, 2003; Bjornson et al., 2007; Keawutan et al., 2014). One challenge in non-ambulatory populations is that the most common tools to measure the total magnitude of physical activity, wearable step counters, are not designed to measure non-stepping activity.

More sophisticated tools and analyses are needed to measure or estimate physical activity from varied movements that do not involve stepping. Van Cauwenbergh, Gubbels, De Bourdeaudhuij, & Cardon (2011) report the feasibility of using accelerometry to collect physical activity data in non-ambulatory toddlers. Triaxial, but not uniaxial, accelerometry data were also able to distinguish active time from sedentary time in non-ambulatory toddlers with CP (Oftedal, Bell, Davies, Ware, & Boyd, 2014; Ryan et al., 2015). Similarly, Smith and colleagues have used triaxial accelerometer technology to measure spontaneous lower extremity movements in infants with and without developmental disabilities (Smith, Trujillo-Priego, Lane, Finley, & Horak, 2015; Smith, Vanderbilt, Applequist, & Kyvelidou, 2017). Tri-axial accelerometers have also been used to measure the amount and types of upper limb movements in infants (Trujillo-Priego et al., 2017) and adults who have had a stroke (Lang, Waddell, Klaesner, & Bland, 2017; Trujillo-Priego et al., 2017).

In addition to being limited, the existing evidence on physical activity in non-ambulatory toddlers with CP has only been reported from full-day recordings (Keawutan et al., 2017; Oftedal, Bell, Davies, Ware, & Boyd, 2015). While full-day recordings importantly allow the ratio of active and sedentary times to be measured over one or more days, they are influenced by family and environmental factors, and may not accurately reflect the child's self-initiated movements. For example, passive movements like being carried or assisted by an adult, car transport, or placement in a positioning device or stroller are recorded as activity by accelerometers. Floor-play time provides the greatest opportunity for self-initiated physical activity and motor exploration for non-ambulatory toddlers with CP and is less influenced by the daily activities than full-day recordings. To our knowledge, no research has examined the amount of physical activity during floor-play in non-ambulatory toddlers with CP. The purpose of this study was to measure self-initiated physical activity and its relationship to gross motor function and participation in non-ambulatory toddlers with CP. We hypothesized that self-initiated physical activity would have a positive relationship with gross motor function but would not be significantly related to participation.

2. Materials and methods

2.1. Participants

Children with CP were recruited from an ongoing rehabilitation clinical trial investigating two types of rehabilitation approaches and their influence on motor development (ClinicalTrials.gov Identifier: NCT02340026). Participants were eligible for the trial if they met the following criteria: 12–36 months of age, diagnosis of CP or suspected CP (suspected CP was identified as a motor percentile rank less than the 10th percentile on the Bayley Scales of Infant Development (Bayley, 2006; Lee et al., 2013), and a neurological sign associated with CP such as spasticity), the ability to initiate pulling to stand at a surface as indicated by a score of 1 on the Gross Motor Function Measure (GMFM) item 52 (Russell, Rosenbaum, Avery, & Lane, 2002), and the cognitive ability to follow one-step commands. Participants were not eligible for the trial if they demonstrated any of the following: secondary orthopedic, neuromuscular or cardiovascular condition unrelated to CP, general muscle hypotonia without other neurological signs associated with CP, independent walking ability as indicated by a score of 3 on GMFM item 69, or history of surgery or injury to the lower extremities in



Fig. 1. Participant with wearable inertial sensor donned.

the past 6 months. All procedures were approved by the Institutional Review Board at The Children's Hospital of Philadelphia.

2.2. Data acquisition

Parents/caregivers of participants were provided with a wearable inertial sensor (Sapphire sensor, APDM, Inc., Portland, OR) with a hook and loop fastening strap (see Fig. 1) to record physical activity during self-initiated play time at home. A tri-axial accelerometer in the sensor recorded data at 128 Hz and stored raw data locally on the sensors until they were returned. The sensor was placed by the caregiver on the dominant leg on the lateral aspect of the thigh so that it did not interfere with either sitting or movement in prone. A member of the research team demonstrated how to use the device and where to place the device on the child's thigh. Caregivers were instructed that the sensor should be worn during free play time on the floor without being moved by another individual. Caregivers were provided with a log to record several bouts of floor-play time over a one-week period. On the log, they recorded the date, the start and the stop times of free floor-play time, and activities completed during floor-play. An example was provided with the log. See Fig. 1.

Gross motor capacity was assessed using the GMFM by the same reliable evaluator [32]. The GMFM was developed specifically for children with cerebral palsy and generally all items on the test can be completed by a child of typical development by the age of 5 years (Russell et al., 2002). The GMFM-66 score is a Rasch-analyzed interval measure of gross motor function with strong validity, reliability, and responsiveness to change (Avery et al., 2003; Russell et al., 2000, 2002). Dimensions or subscales of the GMFM are grouped into different gross motor activities and can be scored individually. Participant scores for Dimensions B (Sitting), C (Crawling & Kneeling), D (Standing), and E (Walking, Running & Jumping) were included in the analysis.

To measure participation, caregivers completed the Child Engagement in Daily Life (CEDL) measure, a caregiver-proxy report of children's participation in family and recreational activities and in self-care skills (Chiarello et al., 2014). It has been studied in children with CP ages 18–60 months within the full-range of severity levels (Gross Motor Function Classification System, GMFCS, I–V), and its validity and reliability have been established (Chiarello et al., 2016; Palisano, Chiarello, McCoy, Bartlett, & An, 2014). The CEDL has two parts. Part one assesses participation in family and recreational activities including both the frequency of the child's participation and the perceived enjoyment by the child. The activities vary from sedentary to highly active and include independent play and social play. Part two assesses the child's self-care skills and amount of assistance required to complete activities such as feeding, dressing, bathing, and toileting. Children completed floor-play within their natural home environment, and the CEDL provides greater insight from the parent/caregiver perspective regarding the child's participation and self-care abilities in their natural environment.

2.3. Data analysis

To determine the relevant segments of the sensor data, the floor-play log recorded by the parents and the raw accelerometer data were reviewed. The sensor recorded data whenever it was removed from the charger, which allowed data files to range in length from 20 to 30 minutes if the sensor was returned to the charger after use, or up to 17 h if the sensor remained unplugged until the battery charge was depleted. Raw data files were viewed in Motion Studio (APDM, Inc., Portland, OR). The floor-play start and stop times in the raw data were determined from the parent/caregiver logs, reconciled with visual inspection. The raw data were visually examined during the parent recorded start/stop intervals. When accelerations were, in fact detected during the interval, the data segment was trimmed by approximately 10 s at the beginning and at the end to account for donning/doffing of the sensor or

placement of the child on the floor. There were occasional instances when the raw data had an unusual pattern or large amplitude spike beyond 10 s that appeared different from the remaining raw data in the same interval. This was presumed to be caregiver interference at the beginning or end of floor-play time, and these files were trimmed accordingly, greater than 10 s. The reconciled start and stop times, representing free movement of the child during floor-play time, were used to crop the raw data for analysis. Raw data were excluded from analysis if the parent log did not contain any recorded floor-play time, if the recorded floor-play time on the log occurred at a time when no acceleration was recorded by the sensor (including if raw data reflected flat, stable acceleration due to gravity only, as if the sensor were lying untouched in place), or if the activities listed during floor-play time on the log included activities during which the child's movements were being assisted (such as "physical therapist working with the child," or "child was in a bounce"). Data files were trimmed if the parent log reported a diaper change occurring at a specific time.

Raw motion data files were imported into a custom MATLAB script for further processing. The triaxial accelerometer recorded acceleration in three planes of motion, oriented to its own frame of reference, which was converted to the world frame of reference with the custom MATLAB script so that acceleration components due to gravity could be accurately removed from each plane individually. The gravity vector was subtracted from the rectified acceleration signal to obtain the user acceleration. We then calculated the total magnitude of user acceleration over the floor time interval by taking the integral of the user-acceleration vector function to measure the area under the curve. This gave us the final rectified acceleration, which acts as a proxy measure of total physical activity (Hayward et al., 2015; Trujillo-Priego et al., 2017) during floor-play time. User-acceleration was then time-normalized to the duration of floor-play time for comparison across floor-play bouts and across children.

The toddlers' physical activity data were included in the analysis if at least 90 min of usable floor-play were identified. Ninety minutes of floor-play time was selected as a sufficient representation of independent physical activity and an achievable amount for families to record. Data were included from either the first (baseline) or second testing session (6 weeks later) of the ongoing clinical trial. If the total floor-play duration was greater than 90 min for both the first and second testing sessions, the first testing session was used for analysis.

To account for non-normal distributions in the measures, Spearman's rho correlation coefficients were used to quantify the relationships between the child's physical activity during floor-play time, GMFM scores, and CEDL subsection scores. The level of statistical significance was set at $p < .05$.

3. Results

Data from thirty toddlers enrolled in the rehabilitation clinical trial (14 female, 16 male; mean age of 23.9 months; Gross Motor

Table 1

Description of participants including age (mo), sex, and Gross Motor Function Classification System (GMFCS) levels, and explanation of excluded data.

Participant	Age	Gender	GMFCS	Included in analysis?
1	29.0	M	IV	Yes
2	35.8	F	III	No: Incomplete log
3	29.6	F	IV	Yes
4	28.8	F	III	Yes
5	31.4	M	III	Yes
6	35.4	F	IV	Yes
7	25.3	M	II	No: Incomplete log
8	29.7	F	III	Yes
9	33.1	F	III	Yes
10	17.6	M	IV	Yes
11	18.4	F	IV	Yes
12	25.1	M	III	Yes
13	29.3	M	II	No: Parent recording error
14	34.6	M	IV	Yes
15	29.6	M	II	Yes
16	21.7	M	I	No: No log complete
17	27.7	F	II	Yes
18	19.3	F	III	No: No corresponding data files
19	15.9	M	I	No: No log complete
20	17.4	F	IV	No: No log complete
21	21.5	M	II	No: No corresponding data files
22	16.8	M	II	Yes
23	21.2	M	II	Yes
24	20.9	F	II	Yes
25	12.2	F	II	Yes
26	21.9	F	III	Yes
27	26.0	F	IV	Yes
28	13.1	M	II	No: Withdrew from study
29	15.5	M	I	No: Toddler removing sensor
30	13.6	M	III	Yes

Table 2
Data files excluded from analysis.

Reasons Data Files Not Included	Number of Data Files
Total number of logged parent/caregiver recordings	123
No corresponding data file	9
No apparent movement	3
In equipment (bouncer/swing) or during PT	2
Timestamp error	1
Very small file size; file errors	1
Start or stop time was not documented on log	2
Sensor slid off toddler or was taken off by toddler	3
Number of parent/caregiver recordings not included	21
Number included	102

Function Classification System (GMFCS) Level I: 10%, Level 2: 30%, Level III: 33%, Level IV: 27%) were included. See Table 1 for participant demographics and an explanation of excluded data.

Of these, 20 toddlers had at least 90 min (mean: 229 min) of usable free floor-play. Out of the 6,129 min of floor-play recorded by parents/caregivers, a total of 4,598 min were included in the analysis. See Table 2 for a description of data files that were unable to be included in the analysis. See Table 3 for summary of results.

There were no statistically significant relationships between physical activity during floor-play and gross motor function as measured by GMFM-66 score ($r = 0.20$; $p = 0.394$). There was also no significant relationship between the physical activity and the GMFM dimension (subscale) scores (B: $r = 0.18$; C: $r = 0.19$; D: $r = -0.02$; E: $r = 0.11$; $p = 0.426 - 0.946$).

Physical activity was not related to the frequency of participation in home and recreational activities ($r = 0.09$, $p = 0.710$), perceived enjoyment of activities ($r = 0.05$, $p = 0.835$), or participation in self-care skills (CEDL Part 2; $r = 0.07$, $p = 0.769$). See Table 4 for the relationships between physical activity, motor function, and participation.

4. Discussion

The aim of the present study was to measure the self-initiated physical activity of toddlers with CP and to examine the relationship of self-initiated physical activity with participation and gross motor function. The results suggest that self-initiated physical activity, gross motor function, and participation are not related in non-ambulatory toddlers with CP.

Accelerometers have been used in multiple ways to measure physical activity in children with CP (Jiang, Lane, Perkins, Schiesel, & Smith, 2017; Keawutan et al., 2017; Ryan, Walsh, & Gormley, 2014, 2015; Smith & Bagley, 2010; Smith et al., 2015, 2017; Trujillo-Priego & Smith, 2017; Trujillo-Priego et al., 2017; Van Cauwenberghhe et al., 2011), but no prior study has reported the amount of physical activity during free floor-play when the toddler's movements are self-initiated and unassisted. A non-ambulatory toddler with CP will often spend time in various types of supportive equipment throughout the day, such as a stroller, car-seat, or high-chair

Table 3
Participant self-initiated physical activity (mean acceleration (g)/minute) during floor-play time, gross motor function, and participation.

Participant Number	Physical Activity	GMFM-66 Score	CEDL		
			Part 1. A	Part 1. B	Part 2
1	34.4	28.0	48.4	3.8	40.0
3	11.3	24.0	46.3	2.9	0.0
4	29.9	41.8	44.3	4.3	34.0
5	28.6	43.0	73.2	4.9	57.1
6	30.9	34.8	52.8	4.7	47.0
8	46.2	44.8	29.7	1.3	36.9
9	17.7	39.5	56.6	4.3	54.5
10	36.5	33.4	51.7	3.5	12.8
11	15.8	28.0	50.6	3.7	0.0
12	53.7	37.1	41.1	3.5	26.5
14	47.9	30.5	48.4	3.4	51.7
15	59.5	50.3	59.4	4.4	54.5
17	35.8	52.1	n/a	4.4	44.5
22	35.5	43.6	43.5	3.1	0.0
23	29.0	44.0	39.9	3.9	36.9
24	51.2	39.7	60.9	5.0	62.0
25	38.5	35.7	62.5	4.1	10.3
26	51.1	28.0	60.9	5.0	28.1
27	15.3	36.4	50.6	4.8	59.5
30	37.8	30.0	47.4	4.1	30.6
Median	35.7	36.8	50.6	4.1	36.9

Table 4

Correlations between self-initiated physical activity, gross motor function, and participation.

	GMFM-66	GMFM B	GMFM C	GMFM D	GMFM E	CEDL Part 1 A	CEDL Part 1 B	CEDL Part 2
Physical Activity	0.202 (0.394)	0.177 (0.455)	0.188 (0.426)	−0.016 (0.946)	0.113 (0.634)	0.091 (0.710)	0.050 (0.835)	0.070 (0.769)
GMFM-66		0.971* (0.000)	0.914* (0.000)	0.664* (0.001)	0.535* (0.015)	−0.132 (0.590)	0.152 (0.521)	0.386 (0.092)
GMFM B			0.900* (0.000)	0.665* (0.001)	0.422 (0.064)	−0.005 (0.983)	0.243 (0.301)	0.341 (0.142)
GMFM C				0.664* (0.001)	0.438 (0.053)	−0.086 (0.727)	0.269 (0.252)	0.321 (0.167)
GMFM D					0.052* (0.024)	0.211 (0.387)	0.310 (0.183)	0.488* (0.029)
GMFM E						−0.127 (0.604)	0.100 (0.674)	0.467* (0.038)
CEDL Part 1 A							0.717* (0.001)	0.359 (0.131)
CEDL Part 1 B								0.622* (0.003)

Values are Spearman's rho (* denotes significance < 0.05); p-values are in parentheses (2-tailed).

limiting opportunity for active play time. Family's preferences, daily activities, and culture also impact the opportunity for active play. The advantage of examining physical activity during free floor-play only, versus full day recordings, is that it provides an opportunity to capture the child's self-initiated movements while decreasing the influence of external factors. In a longitudinal study of physical activity using full-day recordings, Keawutan et al concluded that habitual physical activity declines after the age of 4 years old in children with CP (Keawutan et al., 2017). Understanding the natural trajectory of self-initiated physical activity from infancy through childhood would fill a gap in our current knowledge.

Our findings did not support our hypothesis that self-initiated physical activity would be related to gross motor function. A relationship between physical activity and gross motor function in children with CP has been reported in other studies (Bjornson et al., 2007; Keawutan et al., 2014). Our contrasting observations could be a result of our extraction of only self-initiated activity during floor-play time, our younger sample, or our non-ambulatory sample. In fact, using full day accelerometer recordings, Keawutan et al reported a relationship between gross motor capacity and habitual physical activity in ambulant 4–5 year old children with CP, but similar to our findings, did not observe this relationship in non-ambulant children of the same age (Keawutan et al., 2018b). Further study comparing activity during self-initiated floor-play and the child's activity during a full day would improve our understanding of physical activity in non-ambulatory children.

While participation is related to physical activity in ambulatory children with CP (Bjornson et al., 2014), our findings supported our hypothesis that in non-ambulatory toddlers, physical activity would not be related to participation. Chiarello et al reported that multiple determinants predict frequency of participation in children with CP, including adaptive behavior, family ecology, and available community recreational activities (Chiarello et al., 2016). This may support the results of the present study, indicating that toddlers' participation in family and social activities are due to factors such as adaptive behavior and availability of community recreational activities and not physical activity levels. Our findings underscore the need for clinicians and researchers to independently measure physical activity, gross motor function, and participation in young children with CP.

There were several limitations of our research study. The participants in our clinical trial were required to be able to initiate pulling to stand, but not walk independently, which limited the variability of motor function in our sample compared to the population of children with CP. The data in our analysis were collected from children evenly distributed between GMFCS levels II-IV, but did not include children with the least (I) or most (V) motor impairment, refer to Table 1 for participant demographics and an explanation of excluded data.

Greater heterogeneity may have strengthened the relationships we observed. Another limitation was the adherence of caregivers to remember to use the sensor during floor-play, complete the parent log, and charge the sensor. There were several raw data files that were not able to be analyzed due to timestamp errors after the battery had been depleted. Advances in battery technology and data storage capabilities may help prevent these errors. Finally, we chose to place the accelerometer on the toddler's thigh in an attempt to capture movement of the legs and trunk during gross motor activities, but this approach does not record physical activity of the upper extremities and may have underestimated total body activity in children who are more active with the upper extremities (for example, children who "commando" crawl with their abdomen in contact with the floor). We chose a single-sensor system for ease of recording by families, but a multiple sensor system would more accurately capture whole-body activity.

Similar to how widely used, commercial physical activity devices measure activity by reporting the total number of steps during an interval of time, we calculated total acceleration magnitude as a proxy measure of physical activity. This measure is an estimate of total activity during a period of time, but does not measure the intensity or distribution of activity during the relevant time. Higher total acceleration can be a result of either higher accelerations during individual movements or by a greater number of movements, and the measure we report here does not distinguish between these. Future work should analyze other measures of physical activity such as temporal characteristics, including sedentary vs. active time, or combinations of temporal and magnitude characteristics, such as high and low intensity bouts that would offer insight on the distribution of physical activity during floor-play time (2018b,

Keawutan et al., 2018a, 2017; Oftedal et al., 2015).

In summary, we did not observe a relationship between self-initiated physical activity and gross motor function or participation in non-ambulatory toddlers with CP. When considering measures and interventions for physical activity, gross motor function, and participation, clinicians and researchers should be careful to measure each construct independently. Future research is needed in larger samples, particularly in samples with wider heterogeneity in gross motor function and other developmental disability populations. Future research should also examine the intensity of self-initiated physical activity in toddlers with developmental disabilities and consider multi-sensor systems to measure whole-body movement.

Author contributions

Laura Prosser PT PhD conceived, designed and obtained funding for the scientific research study and assisted in writing the paper. Sam Pierce PT PhD NCS collected the gross motor function and participation data, performed the statistical analyses, and assisted with writing the paper. Julie Skorup PT PCS and Megan Bochnak PT PCS trained caregivers in the use of the sensors and collected the parent logs. Mayumi Mohan MS wrote the data processing script in consultation with Laura Prosser PT PhD and Julie Orlando PT PCS. Athylia Paremski BA and Julie Orlando PT PCS analyzed the physical activity data and parent logs, and Julie Orlando PT PCS wrote the paper. All authors reviewed, edited, and approved the final paper.

Conflicts of interest

The authors declare no conflict of interest. The funding sponsors had no role in the design of the study; in the collection, analyses, or interpretation of data; in the writing of the manuscript, or in the decision to publish the results.

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