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Reliably Measuring Ambulatory Activity Levels of Children and Adolescents With Cerebral Palsy

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Abstract

Objective—To identify sources of variance in step counts and to examine the minimum number of days required to obtain a stable measure of habitual ambulatory activity in the cerebral palsy (CP) population.

Design—Cross-sectional.

Setting—Free-living environments.

Participants—Children and adolescents with CP (N = 209; mean age $\pm$ SD, 8y, 4mo $\pm$ 3y, 4mo; n = 118 boys; Gross Motor Function Classification System [GMFCS] levels I–III) were recruited through 3 regional pediatric specialty care hospitals.

Interventions—Daily walking activity was measured with a 2-dimensional accelerometer over 7 consecutive days. An individual information-centered approach was applied to days with <100 steps, and participants with 3 days of missing values were excluded from the study. Participants were categorized into 6 groups according to age and functional level. Generalizability theory was used to analyze the data.

Main Outcome Measures—Mean step counts, relative magnitude of variance components in total step activity, and generalizability coefficients (G coefficients) of various combinations of days of the week.

Results—Variance in step counts attributable to participants ranged from 33.6% to 65.4%. For youth ages 2 to 5 years, a minimum of 8, 6, and 2 days were required to reach acceptable G coefficient (reliability) of .80 for GMFCS levels I, II, and III, respectively. For those ages 6 to 14 years, a minimum of 6, 5, and 4 days were required to reach stable measures of step activity for GMFCS levels I, II, and III, respectively.

Conclusions—The findings of the study suggest that an activity-monitoring period should be determined based on the GMFCS levels to reliably measure ambulatory activity levels in youth with CP.

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Suppliers

Keywords

Cerebral palsy; Monitoring, ambulatory; Motor activity; Rehabilitation; Reproducibility of results; Walking

Given the health benefits of regular physical activity (PA), becoming and staying physically active is critical regardless of physical limitations.¹ Yet, because of the restriction in functional mobility, children and adolescents with cerebral palsy (CP) experience challenges in engaging in physical activities² and face difficulty engaging in structured activity that is high enough in intensity to provide health benefits.¹ Moreover, because increasing severity of functional involvement is associated with increased energy cost of walking,³ PA level of youth with CP is often related to gross motor function classification⁴ and decreases with age.¹

As clinicians and researchers focus on the effectiveness of PA interventions among youth with CP, recent systematic review has suggested that structured exercise programs combined with online behavioral education programs may be effective in improving habitual PA in persons with CP.⁵ In studies that are summarized in this review, 2 have used objective measures to assess changes in participants' ambulatory activity levels. Such measures including pedometers and accelerometers have commonly been applied in clinical populations.⁶⁻⁹ Accelerometers have been typically worn over a 7-day period drawing from protocols that were empirically developed for able-bodied and disabled adult populations.^{1,4}

We believe that before a quantification of changes in PA levels of youth with CP can take place, it is critical to establish the reliability and validity of methods for assessing ambulatory activity. The generalizability theory (G theory) has been applied in different populations to determine the number of days that is sufficient to achieve an acceptable reliability coefficient.¹⁰⁻¹² The G theory is unique in that it allows the researchers to identify the relative contribution of different sources of variance to total variance in measurement.^{13,14} The findings of the studies by Ishikawa et al¹⁰ demonstrated that 2 days were necessary to reliably measure daily PA of adults with incomplete spinal cord injury. In addition, Wickel and Welk¹² revealed that 7 to 8 days are the minimum number of days of step activity monitoring required to reach stable measure of PA in healthy able-bodied youth. To our knowledge, the G theory has not been applied in the CP population. Against this milieu, the purpose of the study was to employ the G theory to identify the sources of variance in total step activity and to determine the minimum number of days of ambulatory activity monitoring necessary to obtain a reliable estimate of habitual PA level in children and adolescents with CP.

Methods

Participants

A total of 209 youths with CP (mean age $\pm$ SD, 8y, 4mo $\pm$ 3y, 4mo; n = 118 boys) were recruited for the study through 2 institutional review board approved studies at the Seattle Children's Research Institute, with data collected throughout the year between September 2004 and December 2011. An assent form was signed by each participant, and an informed consent form was signed by the parent or guardian of each participant. Participants were grouped into 6 separate groups according to their age and Gross Motor Function Classification System (GMFCS) level.

Age groups

Age categories were 2 to 5 years (including up to age of 5y, 11mo) and 6 to 14 years. These age ranges were chosen because children aged 2 to 5 years are typically preschoolers and are considered to be engaging in unstructured activities in a home-based environment, whereas those aged 6 to 14 years are school age youth who are more likely to engage in structured activities in a school environment. Furthermore, Stevens et al¹⁵ illustrated the difference in step counts between younger and older children, such that older children with CP took fewer steps than younger children with CP, suggesting the notion of age-specific analyses in the present study.

GMFCS levels

The GMFCS is a validated and reliable classification system that has been widely used by researchers and developmental clinicians to describe the gross motor and functional performance of persons with CP.^{16,17} There are 5 levels of motor function (table 1),^{16,18} among which levels I to III were included in the present study.

Ambulatory activity monitoring

The ambulatory activity level of each participant was measured by a StepWatch Activity Monitor (SAM).^a This is a lightweight, ankle-mounted motion sensor device that has been shown to have high accuracy (ie, within $\pm 3\%$) in quantifying continuous step activity among children,¹⁹ and in various activities including walking, running, and stair climbing.²⁰ Each participant wore a SAM on the left ankle, which was calibrated specific to their height (ie, estimated limb length) and gait patterns with calibration accuracy collected from 100 steps trial walk at a self-selected walking speed. In this study, the mean absolute percent error $\pm$ SD was determined to be $2.7\% \pm 1.9\%$ based on step counts obtained from manual counts (ie, criterion) and those recorded by the device (ie, observed) during calibration trials. The unique feature of SAM calibration is the adjustable sensitivity and cadence according to specific movement characteristics of the ankle where the device is placed.^{4,20,21} Hence, the SAM has been used in clinical settings as a valid instrument to measure stepping activity of persons (eg, youth with CP and muscular dystrophy) with atypical gait patterns.⁶

Each participant was provided with verbal and written instructions on how to wear the SAM on the left ankle^{4,20,21} and was asked to wear the device while engaging in normative activities during waking hours (except when bathing) over a period ranging from 3 to 16 consecutive days. On retrieval of devices, because the SAM captures step activity of a single leg, step counts were doubled in order to obtain the overall step activity for each day of the week.

Statistical analysis

In treating step counts for each participant, the first 7 days were used. Days where the number of steps was less than 100 were considered outliers and were treated as missing values.²² Where 2 days of the week were missing, an individual information-centered approach (ie, replacing missing values with the average of the remaining days of the participant)^{23,24} was applied to replace the missing values. Six percent of the total number of days analyzed were replaced by the approach. Eight participants with 3 days of missing values were excluded from the analysis, leaving the step activities of 201 participants for analysis. This exclusion criterion was applied based on the fact that large amounts of missing data reduce the accuracy of recovery. A recent study conducted by Kang et al²⁵

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showed that replacing missing step counts for a maximum of 2 days of missing data yields an absolute percent error <10% using 7-day pedometer data.

Overview of the G theory

The G theory was used to analyze the data. A generalizability study (G study) and a decision study (D study) are the 2 components of the G theory.²³ A total of 6 separate analyses were performed using generalized analysis of variance software.^{b, 26}

G study

The G study allows the quantifying of the relative magnitude of variance components to the total variance in step counts. The G study identifies the magnitude of potentially relevant sources of variance in a measurement. In the current study, a single-facet, random-effects, crossed design of the G study was employed, in which the potential sources of variance included participant term, day term, and the participant $\times$ day interaction terms. The participant term is the object of measurement that is considered as the true variance component that indicates the attribution of an individual's ability to engage in ambulatory activity to the total variance in step counts. The day term is a random facet or a source of systematic error that can potentially influence the total variance in step counts because of the variation in step counts across days. The participant $\times$ day term in this model is the unidentified source of variance in step count measurement that may impact the total variance in step counts. The relative magnitude of error in step counts for each term was computed by dividing the estimate of the single variance component by the total variance and multiplying by 100.¹⁰

D study

The G study was followed by the D study in order to obtain the minimum number of monitoring days required to reliably capture ambulatory activity of youth with CP. The reliability estimates (and generalizability coefficient [G coefficients]) were derived for various combinations of days of the week throughout a relative decision approach, which uses the estimated variance components of the participant term and participant $\times$ day term obtained from the G study. The G coefficient of .80 was considered an acceptable level of reliability in step activity monitoring¹³ and is interpreted in a comparable manner as the classical test theory (ie, intraclass correlation coefficient of .80).²⁷

Results

A total of 201 participants were analyzed. Mean step counts per day of the week for each group are presented in table 2. Mean step activity $\pm$ SD for age 2 to 5 years GMFCS levels I (n = 21), II (n = 19), and III (n = 16) were 11,580 $\pm$ 3229 steps, 9670 $\pm$ 3868 steps, and 3234 $\pm$ 2257 steps, respectively. Mean step activity for age 6 to 14 years GMFCS levels I (n = 54), II (n = 59), and III (n = 32) were 10,732 $\pm$ 4031 steps, 9002 $\pm$ 3564 steps, and 3149 $\pm$ 1992 steps, respectively. The coefficient of variation (CV) (ratio of SD to mean step counts) ranged from .28 to .70 among the groups.

Findings of the G study (table 3) depicted that relative magnitudes of the variance in step counts attributable to participants ranged from 33.6% to 65.4%, with age 2 to 5 years GMFCS level I having the lowest and age 2 to 5 years level III having the highest percentage. The relative magnitudes of variance in step counts accounted for by day term were relatively small for all 6 groups (ie, range, 0%–3.3%). The participant $\pm$ day term,

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which reflects unidentifiable sources of variation in step counts accounted for 31.4% to 65.1% of the total variation, with age 2 to 5 years GMFCS level I having the highest and age 2 to 5 years level III having the lowest value.

As shown in table 4, for age 2 to 5 years, a minimum of 8, 6, and 2 days of the week were required to reach acceptable reliability coefficient (ie, G coefficient .80) for GMFCS levels I, II, and III, respectively. For age 6 to 14 years, a minimum of 6, 5, and 4 days of the week were required to reach stable measures of step activity for GMFCS levels I, II, and III, respectively. Figure 1 illustrates the pattern of level III (ie, most physical limitations) requiring fewer days and level I (ie, least severe disability) requiring more days to reach a desirable reliability level. When comparison of step counts was made relative with age, 2 to 5 year-old children in levels I and II required longer monitoring periods to obtain a stable measure of step activity than 6 to 14 year-old adolescents, but the opposite result was observed for level III (ie, age 2–5 years required a shorter monitoring period than the 6–14 year old group).

Discussion

We aimed to apply the G theory to identify the relative contribution of variance components (ie, participant, day, and participant $\times$ day terms) and to determine the minimum number of days of step activity monitoring necessary to achieve a reliable estimate of ambulatory activity in youth aged 2 to 14 years with CP.

As revealed in table 2, for a given age group (ie, 2–5y and 6–14y), GMFCS level I had the greatest grand mean step count (ie, $11,580 \pm 3229$ steps for age 2–5y and $10,732 \pm 4031$ steps for age 6–14y), while level III had the lowest grand mean step count (ie, 3234 ± 2257 for age 2–5y and 3149 ± 1992 for age 6–14y). Consistent with previously reported studies,^{1,4} daily walking activity decreased as functional walking level (GMFCS level) decreased. It has been postulated that the differences in mean step counts across levels are because of the higher energy cost associated with increased level.³ In addition, when grand mean step count values were compared between age groups, the older group had fewer overall step counts throughout a week-long monitoring period. While we acknowledge that a decline in step counts may solely be because of a function of age, as illustrated in typically developing youth,¹⁹ we speculate that stepping activity in this particular population is influenced by both age and GMFCS level, supporting the inverse relationships of step counts to gross motor function¹ and age,¹⁵ as previously documented.

Our G study results displayed a relatively large amount of variance in step activity attributable to the participant term, ranging from 33.6% to 65.4% of the total observed variance (see table 3). We found that the greater the severity of disability, the larger the relative magnitude of variance in step counts accounted for by the participant term. GMFCS level III children had a higher CV for mean step counts across individuals compared with level I children in both age groups, meaning that, particularly among level III children, the majority of variance in step counts was possibly related to perceived fitness and participation limitations,²⁸ as well as demographic influences on PA of children with disabilities.²⁹ This was also explained by a relatively high CV in level III compared with level I in both age groups. Typically, greater variation is seen with greater grand mean step counts; yet, discrepancy was examined in the CP population. The SD, which indicates variation in step counts across individuals, was high in level III in relation with the grand mean step counts, leading to greater CV in level III than in level I, which had a smaller SD in relation with the grand mean step counts. The G theory applied in adults with incomplete spinal cord injury¹⁰ showed similar findings, in that the participant term accounted for approximately 70% of the total variance in daily step activity.

In regards to the day term, a very small amount (ie, 0%–3.3%) of variance in total step activity was accounted for by the variation in step counts across days. This finding was also similar to that reported by Ishikawa,¹⁰ Wickel,¹² and colleagues who noted a day term accounting for 1.3% and 2.7% of the total variance in daily ambulatory activity in adults with incomplete spinal cord injury¹⁰ and youth,¹² respectively. As suggested by Wickel and Welk,¹² the current study denoted a relative stability in mean step counts across the 7-day monitoring period, meaning that increasing the number of step activity monitoring days could be predicted to have a minimal impact on variance in ambulatory activity¹⁰ among youth with CP.

The relative magnitudes of variance in step counts accounted for by the participant $\times$ day term, which reflects unidentifiable sources of variance in step activity, ranged from 31.4% to 65.1%. In contrast to the participant term, the higher the functional walking level, the higher the relative magnitude attributable to participant $\times$ day term requiring a longer monitoring period to assure a reliable measure of locomotor activity in CP. Potential sources of variance in this term may be the variation in pain levels across days, as well as varying day-to-day demands in youth with CP,³⁰ which may fluctuate the daily step counts across days. Furthermore, barriers and facilitators to PA in this population may also play a significant role in determining their habitual ambulatory activity levels.² Therefore, future research should focus on investigating those factors that possibly impact PA levels among physically challenged individuals.

As presented in table 4 and figure 1, the results from the D study show that fewer monitoring days were required to obtain a stable measure of habitual activity level in GMFCS level III for both age groups. Because high reliability is achieved when there is a greater interindividual variation in step counts relative to between-day variation,¹⁰ the current analyses support that, when level III is compared with level I, there is a relatively greater variation in daily step activity across individuals (as revealed by the large percentage in the participant term) and a smaller amount of variation across days. Viewed collectively, fewer days are necessary to obtain a reliable estimate of habitual PA in level III compared with level I. This finding can be applied to other populations with disabilities, given that PA levels are considerably influenced by specific diagnostic categories within each disability type (ie, sensory and physical disabilities) among youth.²⁸

Study strengths

The strength of the study was the large sample size of 201 youths with CP, age ranging from 2 to 14 years and GMFCS levels from I to III. The sample size of the current study allowed 6 separate analyses in order to examine the differences in step activity counts by age and functional status and to determine the minimum number of days of locomotor activity monitoring required to reliably estimate daily PA levels, specifically for different age groups and GMFCS Levels.

Study limitations

As with any research, there are some limitations to this study. The step activity measure of each participant was taken over a period of less than 2 weeks out of a whole year, a relatively short period of time to determine the habitual PA level of an individual.³¹ Therefore, the step counts used in the analysis might not have portrayed the actual profile of ambulatory patterns of youth with CP. While we acknowledge the possibility of monthly and seasonal effects on variation in step activity,^{12,32} a minimal bias because of age and GMFCS level was suspected, provided that the majority of data collection was spread apart throughout the year, regardless of age and GMFCS level.

Conclusions

The findings of the study demonstrate that, in order to reliably measure the stepping activity of children and adolescents with CP, a minimum of 8, 6, and 2 days is required for GMFCS levels I, II, and III of age 2 to 5 years; and 6, 5, and 4 days for levels I, II, and III of age 6 to 14 years. Though caution may be warranted, because potential confounders of PA are yet to be fully identified, a shorter monitoring period is necessary to reliably estimate the ambulatory activity level of participants with more functional limitations compared with less functionally challenged youth with CP ages 2 to 14 years. From a practical point of view, the present study suggests that the monitoring period should be chosen according to age and GMFCS level in youth with CP in order to reliably capture changes in their habitual PA level. A stable measure of step activities will help to appropriately evaluate the effectiveness of future research aimed to enhance PA of youth with CP.

List of abbreviations

CP	cerebral palsy
CV	coefficient of variation
D study	decision study
GMFCS	Gross Motor Function Classification System
G coefficient	generalizability coefficient
G study	generalizability study
G theory	generalizability theory
PA	physical activity
SAM	StepWatch Activity Monitor

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Fig 1.

Relations between the days of step activity monitoring and the G coefficient: (A) ages 2–5 years and (B) ages 6–14 years. The dotted line indicates a desirable reliability coefficient of .80. The D study revealed a minimum number of days required to achieve a G coefficient .80: ages 2–5 years, GMFCS level I = 8 days, GMFCS level II = 6 days, and GMFCS level III = 2 days; ages 6–14 years, GMFCS level I = 6 days, GMFCS level II = 5 days, and GMFCS level III = 4 days.

Table 1Summarized descriptions of functional limitations by levels of GMFCS^{4,16,18}

GMFCS Level	Definition
I	Walks without restrictions; able to climb stairs without upper-extremity assistance; limitations with more advanced gross motor skills (ie, speed, balance, and coordination); may participate in physical activities and sports depending on personal choices and environmental factors.
II	Walks without assistive devices; requires upper-extremity assistance to climb stairs; limitations walking outdoors and in the community; environmental factors and personal preference influence mobility choices.
III	Walks only with assistive mobility devices; unable to climb stairs; may require a seat belt for pelvic alignment and balance when seated. Sit-to-stand and floor-to-stand transfers require physical assistance; difficulty traveling long distances.
IV	Self-mobility with limitations; uses power mobility outdoors and in the community; requires adaptive seating for trunk and pelvic control; physical assistance is required for most transfers.
V	Self-mobility is severely limited to the ability to maintain antigravity head and trunk postures; transfers require complete assistance of an adult.

Table 2

Mean step counts for each day of the week by age and GMFCS

Day	Aged 2–5y			Aged 6–14y		
	GMFCS Level I (n = 21)	GMFCS Level II (n = 19)	GMFCS Level III (n = 16)	GMFCS Level I (n = 54)	GMFCS Level II (n = 59)	GMFCS Level III (n = 32)
Monday	11,457±4787	9858±5497	3372±3079	10,744±5711	9184±4329	2761±1842
Tuesday	10,448±4319	8941±5778	3464±3172	11,359±5149	8923±4178	3653±2666
Wednesday	10,928±4667	9429±5296	2832±2456	11,243±6326	9286±4719	3350±2846
Thursday	11,827±4405	9194±4731	3465±2924	10,996±5441	10,013±4823	3357±2262
Friday	10,565±5427	10,031±5780	2177±1172	11,314±5892	9240±5518	3485±2533
Saturday	12,935±6022	9832±5052	4169±3119	10,123±6130	8402±5471	2576±3118
Sunday	12,902±4390	10,405±5325	3159±2048	9344±4868	7969±3956	2862±2699
Grand mean	11,580±3229	9670±3868	3234±2257	10,732±4031	9002±3564	3149±1992
CV	0.28	0.40	0.70	0.38	0.40	0.63

NOTE. Values are mean ± SD.

Table 3
Variance component estimate and relative magnitude of variation for participant, day, and participant $\times$ day terms

Term	Aged 2–5y			Aged 6–14y		
	GMFCS Level I (n = 21)	GMFCS Level II (n = 19)	GMFCS Level III (n = 16)	GMFCS Level I (n = 54)	GMFCS Level II (n = 59)	GMFCS Level III (n = 32)
	Variance	Variance	Variance	Variance	Variance	Variance
P	8,168,100 (33.64)	12,659,300 (44.02)	4,768,887 (65.36)	13,610,440 (42.11)	11,065,080 (48.57)	3,506,635 (51.55)
D	313,923 (1.29)	0 (0.00)	237,716 (3.26)	217,607 (0.67)	243,431 (1.07)	67,764 (1.00)
P $\times$ D	15,798,480 (65.07)	16,099,470 (55.98)	2,290,245 (31.39)	18,496,030 (57.22)	11,471,420 (50.36)	3,228,334 (47.45)
Total	24,280,502	28,758,770	7,296,847	32,324,077	22,779,932	6,802,733

NOTE. Values in parentheses are percentages.
Abbreviations: D, day; P, participant; P $\times$ D, participant $\times$ day.

Table 4

Minimum number of days to achieve desirable level of reliability (G coefficient .80)

Age Group	<u>GMFCS Level</u>		
	I	II	III
2–5y	8	6	2
6–14y	6	5	4