International Journal of Pediatrics

Fitness and Physical Activity in Children and Youth with Disabilities

Guest Editors: Maria A. Fragala-Pinkham, Margaret E. O'Neil, Kristie F. Bjornson, and Roslyn N. Boyd

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Editorial Board

Contents

Fitness and Physical Activity in Children and Youth with Disabilities, Maria A. Fragala-Pinkham, Margaret E. O'Neil, Kristie F. Bjornson, and Roslyn N. Boyd
Volume 2012, Article ID 162648, 2 pages

A Systematic Review of the Clinimetric Properties of Habitual Physical Activity Measures in Young Children with a Motor Disability, Stina Oftedal, Kristie L. Bell, Louise E. Mitchell, Peter S. W. Davies, Robert S. Ware, and Roslyn N. Boyd
Volume 2012, Article ID 976425, 12 pages

Play and Be Happy? Leisure Participation and Quality of Life in School-Aged Children with Cerebral Palsy, Keiko Shikako-Thomas, Noémi Dahan-Oliel, Michael Shevell, Mary Law, Rena Birnbaum, Peter Rosenbaum, Chantal Poulin, and Annette Majnemer
Volume 2012, Article ID 387280, 7 pages

Accelerometry: A Feasible Method to Quantify Physical Activity in Ambulatory and Nonambulatory Adolescents with Cerebral Palsy, Jan Willem Gorter, Stephen G. Noorduyn, Joyce Obeid, and Brian W. Timmons
Volume 2012, Article ID 329284, 6 pages

A Review of the Potential for Cardiometabolic Dysfunction in Youth with Spina Bifida and the Role for Physical Activity and Structured Exercise, Kevin R. Short and Dominic Frimberger
Volume 2012, Article ID 541363, 11 pages

Arterial Structure and Function in Ambulatory Adolescents with Cerebral Palsy Are Not Different from Healthy Controls, Audra A. Martin, Lisa M. Cotie, Brian W. Timmons, Jan Willem Gorter, and Maureen J. MacDonald
Volume 2012, Article ID 168209, 8 pages

Evaluation of Functional Electrical Stimulation to Assist Cycling in Four Adolescents with Spastic Cerebral Palsy, Ann Tokay Harrington, Calum G. A. McRae, and Samuel C. K. Lee
Volume 2012, Article ID 504387, 11 pages

Factors Influencing Physical Activity in Children and Youth with Special Health Care Needs: A Pilot Study, Katie Feehan, Margaret E. O'Neil, Diana Abdalla, Maria Fragala-Pinkham, Monica Kondrad, Zekarias Berhane, and Renee Turchi
Volume 2012, Article ID 583249, 11 pages

Participation in Physical Activity for Children with Neurodevelopmental Disorders, Rubab G. Arim, Leanne C. Findlay, and Dafna E. Kohen
Volume 2012, Article ID 460384, 9 pages

Hindawi Publishing Corporation
International Journal of Pediatrics
Volume 2012, Article ID 162648, 2 pages
doi:10.1155/2012/162648

Editorial

Fitness and Physical Activity in Children and Youth with Disabilities

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Received 10 December 2012; Accepted 10 December 2012

Over the last decade, there has been a major shift in pediatric rehabilitation from an impairment-based model to activity-based interventions focusing on improving fitness, physical activity, and participation for children and youth with disabilities [1]. A catalyst for this shift is related to the emphasis on health promotion and disease prevention in the health care arena. There is growing evidence on the importance of daily physical activity for health in children and youth as well as evidence on positive health outcomes from programs that promote physical activity and fitness for children and youth; however, less information is available on these topics for children and youth with disabilities [2–6].

As rehabilitation interventions incorporate more strategies to increase fitness, physical activity, and participation in children and youth with disabilities, it is critical that outcome measures are appropriate to examine intervention effectiveness. Over the past few years, there has been an increase in measurement methodology research to ensure the feasibility, reliability, validity, and responsiveness of fitness and physical activity measures for children and youth with disabilities. It is important that pediatric rehabilitation researchers design sound intervention and measurement protocols to identify the effectiveness of activity-based interventions to improve fitness and physical activity in children and youth with disabilities. It is important that these measures are accessible and feasible for researchers and clinicians. Further, it is important that researchers articulate clear operational definitions of the fitness components (strength, endurance, flexibility, and body composition) and physical activity dimensions (frequency, duration, and intensity) that are being examined in the research. The aim of this special issue is to expand the level of knowledge about fitness interventions, physical activity participation, and measurement protocols for children and youth with disabilities.

This special issue represents an international forum of physical activity and fitness research. The articles reflect the components of the World Health Organization's International Classification of Functioning, Disability, and Health (the ICF Model) and they target a variety of disabilities and conditions of childhood including cerebral palsy, spina bifida, motor disability, and neurodevelopmental disorders.

The ICF Model includes three personal dimensions; body structure and body function impairments, activity limitations, and participation restrictions. In this special issue, two articles focused on body structure and function impairments with one examining the potential for cardiometabolic dysfunction in youth with spina bifida and the role of physical activity and exercise while the other examined arterial structure and function in cerebral palsy. Four articles examined different aspects of activity. Two articles examined measures of physical activity; one was a systematic review of clinimetric properties of habitual physical activity measures in motor disability and one examined the feasibility of accelerometry to measure physical activity in cerebral palsy. Another article focused on functional electrical stimulation as an intervention strategy to assist in cycling activity and one examined child, family, and environmental factors that influence physical activity levels in children with special health care needs. Two articles examined participation; one in the context of play in children with cerebral palsy and one in the context

of overall physical activity participation in children with neurodevelopmental disorders.

Each study illustrates the multiple aspects of physical activity and fitness that must be considered when designing an intervention program and measurement protocol to examine outcome effectiveness. Further, these papers also represent the variety of issues to consider when conducting research with children and youth with disabilities. There is a multitude of physiological, medical, functional, and environmental factors that contribute to the effectiveness and sustainability of interventions and outcomes. Also, when one conducts pediatric research, there are multiple maturational and developmental factors to consider. The articles in this special issue reflect the breadth of childhood as different researchers focused on different age ranges of childhood and adolescence.

This special issue provides a good resource to inform and facilitate future research on fitness and physical activity in children and youth with disabilities. This collection of work represents a substantial contribution to the burgeoning field of activity-based research and fitness and physical activity outcomes for children and youth with disabilities.

Maria A. Fragala-Pinkham
Margaret E. O'Neil
Kristie F. Bjornson
Roslyn N. Boyd

References

[1] E. G. Fowler, T. H. A. Kolobe, D. L. Damiano et al., "Promotion of physical fitness and prevention of secondary conditions for children with cerebral palsy: section on pediatrics research summit proceedings," *Physical Therapy*, vol. 87, no. 11, pp. 1495–1510, 2007.

[2] C. A. Maher, M. T. Williams, T. Olds, and A. E. Lane, "Physical and sedentary activity in adolescents with cerebral palsy," *Developmental Medicine and Child Neurology*, vol. 49, no. 6, pp. 450–457, 2007.

[3] G. C. Frey, H. I. Stanish, and V. A. Temple, "Physical activity of youth with intellectual disability: review and research agenda," *Adapted Physical Activity Quarterly*, vol. 25, no. 2, pp. 95–117, 2008.

[4] P. J. Morris, "Physical activity recommendations for children and adolescents with chronic disease," *Current Sports Medicine Reports*, vol. 7, no. 6, pp. 353–358, 2008.

[5] J. A. Rimmer and J. L. Rowland, "Physical activity for youth with disabilities: a critical need in an underserved population," *Developmental Neurorehabilitation*, vol. 11, no. 2, pp. 141–148, 2008.

[6] C. C. Johnson, "The benefits of physical activity for youth with developmental disabilities: a systematic review," *American Journal of Health Promotion*, vol. 23, no. 3, pp. 157–167, 2009.

Hindawi Publishing Corporation
International Journal of Pediatrics
Volume 2012, Article ID 976425, 12 pages
doi:10.1155/2012/976425

Review Article

A Systematic Review of the Clinimetric Properties of Habitual Physical Activity Measures in Young Children with a Motor Disability

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Received 24 January 2012; Revised 4 April 2012; Accepted 23 April 2012

Academic Editor: Kristie F. Bjornson

Aim. To identify and systematically review the clinimetric properties of habitual physical activity (HPA) measures in young children with a motor disability. *Method*. Five databases were searched for measures of HPA including: children aged <6.0 years with a neuromuscular disorder, physical activity defined as "bodily movement produced by skeletal muscles causing caloric expenditure", reported HPA as duration, frequency, intensity, mode or energy expenditure, and evaluated clinimetric properties. The quality of papers was assessed using the COSMIN-checklist. A targeted search of identified measures found additional studies of typically developing young children (TDC). *Results*. Seven papers assessing four activity monitors met inclusion criteria. Four studies were of good methodological quality. The Minimod had good ability to measure continuous walking but the demonstrated poor ability to measure steps during free-living activities. The Intelligent Device for Energy Expenditure and Activity and Ambulatory Monitoring Pod showed poor ability to measure activity during both continuous walking and free-living activities. The StepWatch showed good ability to measure steps during continuous walking in TDC. *Interpretation*. Studies assessing the clinimetric properties of measures of HPA in this population are urgently needed to allow assessment of the relationship between HPA and health outcomes in this group.

1. Introduction

Habitual physical activity (HPA) is an established determinant of health in children and is required for healthy development, including the growth of bone and muscle mass, improved balance and motor skills, maintaining a healthy weight and improved psychological wellbeing [1, 2]. Limited evidence suggests young children with motor disorders are less physically active than their typically developing peers [3]. Consequently they may be at risk of suboptimal growth and development in addition to the development of secondary conditions such as chronic pain, fatigue, and low bone density which can lead to diminished bone health [4, 5]. Australian physical activity guidelines state children aged from one to five years should be physically active for three hours throughout the day and should not be sedentary, restrained, or kept inactive, for more than one hour at a time, with the exception of sleeping [2]. Studies investigating the link between HPA and health outcomes for children with motor impairments have not been conducted. These studies are urgently needed to (i) determine the HPA patterns and intensities of children with motor disabilities, (ii) support the importance of physical activity promotion and inactivity prevention, (iii) determine the dose-response relationship

between physical activity and health outcomes, and (iv) allow assessment of the effectiveness of interventions aimed at increasing HPA.

Physical activity is defined as "any bodily movements using skeletal muscles that results in energy expenditure" [6]. The International Classification of Functioning, Disability and Health (ICF) further divides physical activity into the concepts of "performance" and "capacity" [7]. Performance is defined as the execution of an activity in the natural environment. This is distinct from capacity which refers to a child's maximal ability to perform an activity in an ideal or standardised environment. Performance and capacity to complete a task (e.g., jumping) can be determined by observing the child undertake that specific task within a limited time frame in their natural and ideal environment, respectively. HPA, on the other hand, describes a child's typical daily activity pattern and hence includes their performance of a multitude of activities which necessitates measurement across multiple days [8]. The variables of frequency (how often a child does an activity), intensity (how hard a child works to do the activity), duration (how long a child does an activity), and mode (what the child is doing) are used to characterise HPA patterns, while activity-related energy expenditure (AEE) is used as a summary variable of all the other indicators [9, 10].

Direct observation is considered the gold standard for physical activity classification, while doubly labelled water in combination with resting energy expenditure is considered the gold standards for calculating AEE [10]. Measures of HPA include both objective and subjective methods [11]. Objective measures include heart rate monitors, accelerometers, and pedometers. Subjective methods include self- or proxy reports, interviews, and diaries. The measurement of HPA in typically developing children (TDC) has received a great deal of attention in the last decade, primarily due to a concern over the increasing rate of overweight and obesity in childhood [12]. A systematic review of pedometers and accelerometers has identified moderate-to-good evidence of validity and strong evidence of reliability for the ActiGraph accelerometer in preschool-aged TDC [13]. Children with motor impairments may not be walking but instead cruising, crawling, bottom shuffling, rolling, use walking aids or a wheelchair as means of locomotion, and those who are walking may have different gait patterns and speed of movement than TDC [14]. This variation in movement patterns necessitates validation of motion sensors specifically for this population.

Systematic reviews of measures of physical activity in children and adolescents with cerebral palsy (CP) have been conducted [15, 16]. Capio et al. [15] included children with CP from birth to 18 years and concluded that the subjective measures of the Activities Scale for Kids-Performance Version (ASKp) and the Children's Assessment of Participation and Enjoyment (CAPE) are the only measures with established validity and reliability in children with physical disabilities from the age of six years. The review by Clanchy et al. [16] included children with CP aged 10–18 years and identified the CAPE as the measure with the strongest evidence of reliability and validity. It was also noted that no measures have been assessed in children with more severe disability who are wheelchair dependent. This paper did not include the ASKp as it did not meet inclusion criteria of 60% of items relating to a domain of physical activity performance. This was done to ensure the included measures assess physical activity as traditionally defined by Caspersen et al. [6] and not "participation" which does not take into consideration the sedentary or active nature of the activity. Both reviews note the lack of HPA measures which can assess activity intensity, which limits their ability to provide meaningful comparison with physical activity guidelines, as children from the age of six years are advised to accumulate 60 minutes of moderate-to-vigorous physical activity every day [17]. This current paper aims to systematically review the clinimetric properties (validity, reproducibility, and clinical utility) of measures of HPA in children less than 6 years of age with a motor disability [6].

2. Method

2.1. Search Strategy. A systematic literature search was performed by one reviewer (SO) of the electronic bibliographic databases PubMed, CINAHL, EMBASE, SPORTDiscus, and Web of Science from their inception to September, 2011. Databases were searched using medical subject headings (MeSH) terms and text words for physical activity and disability, limiting the search to the age group <6.0 years.

2.2. Inclusion and Exclusion Criteria. Measures of HPA were included which met the following criteria: (i) included children less than six years of age with a motor disability caused by neuromuscular disorders; (ii) defined physical activity as any bodily movement produced by skeletal muscles that results in caloric expenditure [6]; (iii) reported physical activity in terms of duration, frequency, intensity, or energy expenditure; (iv) questionnaires where at least 60% of items related to a domain of HPA; (v) evaluated clinimetric properties for the measurement of HPA. Studies were excluded if they were (i) not published in English, due to lack of translation services; (ii) primarily measured capacity or participation.

The title and abstract of all retrieved references were screened by the first author to exclude any papers which did not include children in the target age group or were not on the topic of physical activity measures. A second screening of abstracts was performed by two authors (SO, LM) independently to exclude those that did not include habitual physical activity measures but rather assessed capacity or participation measures. For the remaining abstracts, full papers were retrieved and screened by two authors (SO, LM). Publications which did not assess the clinimetric properties of measures of HPA in children with motor disabilities were excluded. Upon disagreement between the two reviewers, a third reviewer (KB) reviewed the publication in question. The reference lists of included papers and relevant reviews were screened, and electronic author and citation tracking were performed when available, to identify relevant publications not identified by the initial search strategy. For measures where clinimetric properties were

found in children less than six years of age with a motor disability, a further search identified any evidence in typically developing children in the same age group.

2.3. Data Extraction and Quality Review. The COSMIN (COnsensus-based Standards for the selection of health Measurement INstruments) checklist [18] was used to rate the methodological quality of the study designs used to evaluate the clinimetric properties validity and reproducibility for measures of HPA. The COSMIN was developed through an international Delphi study to assess the methodological quality of studies on measurement properties of health-related patient-reported outcomes (HR-PROs) [18]; however the measurement properties are also relevant to other health-related measurement instruments. The scoring system was developed based on expert discussion and testing on 46 articles identified by a systematic review and uses a "worst score counts" algorithm [19]. Each item within a specific measurement property is rated individually as "excellent" (+++), "good" (++), "fair" (+), or "poor" (−) and an overall score is given by taking the lowest score of any of the items within the assessment of the measurement property. Measurement properties included in this paper were measurement error (COSMIN box C), hypothesis testing (COSMIN box F), and criterion validity (COSMIN box H). The methodological quality rating of the papers was separated into study design and statistical analysis.

2.4. Rating of Study Design. The study design for the assessment of measurement properties was rated as having "excellent" quality if all relevant items within a given checklist scored "excellent." Study design was rated as "good" if information for some items was not complete and therefore could not be scored "excellent," though it could be assumed these were of "good" quality. A "fair" quality rating was given if there were minor flaws in the design. If the results were not to be trusted because of major flaws in the design, a study is rated as "poor" [19]. To assess if there were any important flaws in the design or methods of the study (COSMIN Box H, item five), items from the systematic review of activity monitors in TDC were used [13] as a guide in addition to whether or not the testing protocol included free-living activities.

One modification was made to the original COSMIN checklist. Proposed sample size requirements [20] were applied in the clinimetric properties section instead of the study design section. The decision to separate study design and sample size requirements was made as sample size does not determine study design quality as such but does affect the statistical power available to detect a significant result.

2.5. Statistical Methods and Clinimetric Properties of HPA Measures. For the assessment of criterion validity, the COSMIN checklist accepts correlations or the area under the receiver operator characteristic (AUC-ROC) curve as "excellent" statistical methods for continuous measures [19]. For dichotomous scales sensitivity and specificity calculations are considered "excellent" measures [19]. For the assessment of concurrent validity (hypothesis testing) it is up to the reviewer to assess if the method is "appropriate" and therefore scores "excellent." For the assessment of measurement error (agreement), Standard Error of Measurement (SEM), Smallest Detectable Change (SDC), and Limits of Agreement (LoA) calculated using the Bland-Altman method are accepted as "excellent." According to the COSMIN a sample size of less than 30 participants is considered "poor," 30–49 participants is considered "fair," 50–99 participants is considered "good," and 100 or more participants is considered "excellent" [19]. Authors were contacted to see if they had used any power calculations when determining their sample size.

Quality criteria for the clinimetric properties of measures were proposed to give a framework to help distinguish between the quality of the studies and the performance of the measurement tools [20]. Agreement, criterion validity, and construct validity were therefore scored as good (+), indeterminate (unable to assess: u/a), or poor (−) [20]. Criterion validity was considered "good" if correlation with gold standard was ≥ 0.70, sensitivity and specificity ≥ 0.70, and area under receiver operating characteristic curve ≥ 0.70 [20]. Construct validity was considered "good" if 75% of the results were in accordance with the hypothesis [20]. Measurement error is considered "good" if the SDC or the LoA are smaller than the minimal important change or if "authors provide convincing arguments that the agreement is acceptable" in a sample size of at least 50 participants [20]. Studies that used a "poor" statistical method scored "indeterminate" for performance of clinimetric properties.

The assessment of clinimetric properties of HR-PRO questionnaires might be of a different nature to those assessing activity measures due to the availability of gold standards, something which is not readily available for HR-PROs as these are usually questionnaires regarding patient outcomes such as pain and quality of life [21]. The statistical methods and performance of clinimetric properties were therefore scored according to the COSMIN but were also discussed further, and a different score was given if authors presented a convincing argument that the measure was acceptable.

2.6. Clinical Utility. The clinical utility of measures of HPA was assessed in terms of the need for individual calibration of the equipment, the interpretability of the data output, and the need for software analysis, cost of equipment, and any required software. The feasibility of the equipment for using children aged less than six years with a motor disability in free living situations was also considered (size, weight, number of sensors, place worn, ease of correct placement, and battery life). The richness of data was also assessed in terms of what aspects of HPA (frequency, duration, intensity, mode, and energy expenditure) it was able to assess in children with a motor disability.

2.7. Overall Level of Evidence. The overall level of evidence for the clinimetric properties of measures of HPA in young children with motor disabilities was determined as a whole by assessing the methodological quality of study design and statistical methods used in the assessment of

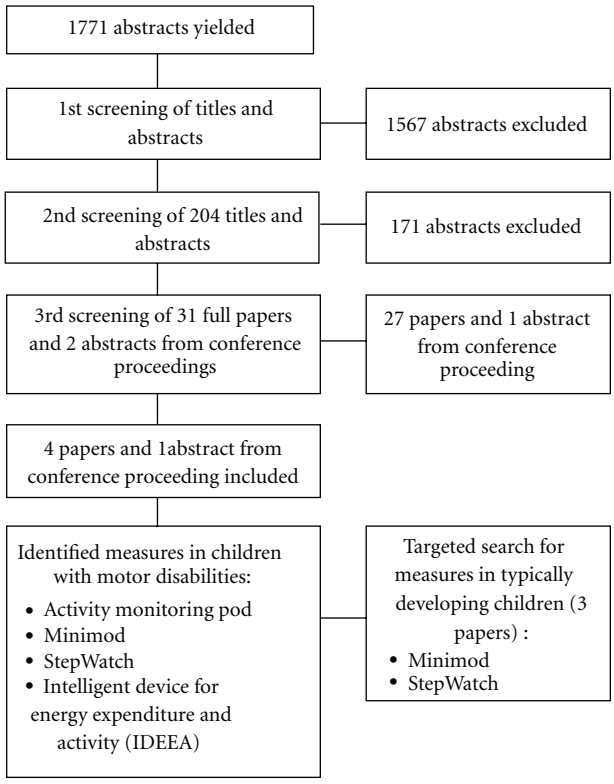

Figure 1: Flow diagram for search strategy.

HPA measures using the "worst score counts" algorithm, agreement between studies on the quality of clinimetric properties of these HPA measures, and the number of studies available for each HPA measure.

3. Results

Description of the results of the search strategy can be seen in Figure 1. The initial search yielded 1771 titles, and abstracts after duplicates were deleted. The initial screening by titles and abstracts excluded 1567 titles, and the second screening excluded a further 171 papers. Thirty-one full papers and two abstracts from conference proceedings were further reviewed. Four papers and one abstract from conference proceedings on the clinimetric properties of four measures of HPA in children with motor disabilities were identified which included children aged less than six years in their sample. None of the studies focused exclusively on children aged less than six years, which led to the inclusion of studies with a wide age range (4–18 years).

The characteristics of the studies are detailed in Table 1. Evidence of clinimetric properties was found for three accelerometer-based activity monitors [22–26] and one pedometer using inertial sensors [23]. A paper by Kuo et al. [23] and an abstract from conference proceedings by Brandes et al. [22] were identified for the Minimod pedometer (DynaPort McRoberts, Hague, Netherlands). The Activity Monitoring Pod-331 pedometer which uses inertial sensors (AMP; Dynastream Innovations, Alberta, Canada) was also assessed in the paper by Kuo et al. [23]. Papers by Stevens et al. [24] and McDonald et al. [25] were identified for the StepWatch pedometer (Orthocare Innovations, WA, USA). A paper by Aviram et al. [26] assessed the Intelligent Device for Energy Expenditure and Activity (IDEEA; Minisun, CA, USA). Only McDonald et al. [25] assessed children with Duchenne muscular dystrophy (DMD); others assessed children with CP who were able to walk with or without aids (Gross Motor Function Classification System levels I–III) [22–24, 26]. None of the study samples included children who did not walk as their main means of locomotion.

All of the studies of children with motor disabilities included TDC as a reference group [22–26]. Sample sizes ranged from 17 to 27 children with motor disabilities and 7–27 in the TDC reference group. All authors were contacted to clarify the proportion of children in their study who were under the age of six. The study by Brandes et al. [22] on the use of the Minimod included one child (5%) with CP aged five years old and four children (20%) in the TDC sample aged three to five years. The study by Aviram et al. [26] included nine children (43%) aged four to five years with three children in each GMFCS category (I–III). The authors did not supply the number of TDC aged five years, but the youngest of the seven children in the TDC group was 5 years and 8 months old [26]. In the study by Kuo et al. [23] it was estimated that a maximum of two children in the CP (11%) and TDC (10%) group were less than six years old, although authors were not able to readily access information to confirm this. It is not known how many children were under the age of six in the study by Stevens et al. [24] and McDonald et al. [25]. The targeted search for studies reporting on the clinimetric properties of the identified measures in young TDC identified a further three studies assessing the Minimod [27] and the StepWatch [28, 29]. The sample size in the three studies ranged from 20 to 162 children, and ages ranged from 2 to 16 years. The StepWatch study by Bjornson et al. [28] reported results for two-to-three-year olds ($n = 60$) and four-to-five-year olds ($n = 62$) TDC separately. The StepWatch study by Song et al. [29] used two age bands (5–7 years and 9–11 years) with ten children in each group, but the number of five year old children in the youngest group was not specified by the authors upon request. In the paper by Brandes et al. [27], authors report on the same sample of TDC ($n = 20$, four children $\leq$6.0 years) as in the previously mentioned abstract from conference proceedings. The full paper and abstract from conference proceedings will be rated as one paper.

3.1. Rating of Study Designs. Description of the study design of included papers is outlined in Table 1. None of the authors using manual step count as a criterion method reported on the accuracy of the manual step count (intra- or interrater agreement). Although it can be assumed that it is a gold standard, no evidence has been provided, and therefore the maximum attainable score for all studies according to the COSMIN checklist was "good" [19]. The study by Brandes et al. [22, 27] assessed the criterion validity of the Minimod in children with CP and TDC compared to manually counted steps and meters walked was rated "good" for study design. Authors report complete information in regarding test

Table 1: Characteristics of studies describing measures of habitual physical activity.

Brand	Population						Protocol		
	n	Boys	Mean age (range)	Children aged ≤5 years (n)	Motor disorder	Reference	Setting (measurement units)	TDC reference group	Study
Minimod[a]	17	—	10 yr 6 mo (4–16 yr)	~2	CP[1]	DO	Continuous walking, structured activity lap, and stair climbing (steps and meters walked)	n = 19, 4–16 yr	Kuo et al. [23]
	20	—	5–16 years (−)	1	CP[2]		Self-paced walking (steps and meters walked)	n = 20, 3–16 yr	Brandes et al. [22, 27]
AMP[b]	20	13	10 yr 6 mo (4–16 yr)	~2	CP[1]	DO	Continuous walking, structured activity lap, and stair climbing (steps and meters walked)	n = 20, 4–16 yr	Kuo et al. [23]
StepWatch[c]	16	16	9 yr 2 mo (5–13 yr)	—	DMD	DO	Slow/fast self-paced walking (steps) + 3-day HR and pedometer (steps and HR)	n = 21, 5–13 yr	McDonald et al. [25]
	27	22	−(4–18 yr)	—	CP[3]	DO	Treadmill walking at different speeds (steps)	n = 27, age matched	Stevens et al. [24]
	60	30	−(2-3 yr)	60	TDC	DO	Self-paced walking (steps)	n/a	Bjornson et al. [28]
	62	31	−(4-5 yr)	62	TDC	DO	Self-paced walking (steps)	n/a	
IDEEA[d]	20	10	−(5–11 yr)	—	TDC	DO	Self-paced walking and running (steps)	n/a	Song et al. [29]
	21	—	−(4 yr–10 yr 1 mo)	9	CP[1]	IC + HR	Series of everyday activities, walking on treadmill, climb staircase (energy expenditure kcal/min)	n = 7, 5 yr 8 mo–8 yr 6 mo	Aviram et al. [26]

[a]Minimod, Dynaport, McRoberts BV, Hague, Netherlands; [b]AMP: Activity Monitoring Pod 331, Dynastream Innovations, Alberta, Canada; [c]StepWatch, Orthocare Innovations, WA, USA, [d]IDEEA: Intelligent Device for Energy Expenditure and Activity, Minisun, CA, USA; CP: cerebral palsy; GMFCS: Gross Motor Function Classification System; DMD: Duchenne muscular dystrophy; TDC: typically developing children; DO: direct observation; IC: indirect calorimetry; HR: heart rate monitor; [1]GMFCS I–III; [2]GMFCS not specified, able to walk; [3]GMFCS I–II; (—): not reported.

protocol (monitor settings, placement, data output, average steps needed for length walked, and range of steps across group) but only tests of continuous walking that may not adequately reflect free-living walking activity. The study by Kuo et al. [23] on the criterion validity of the Minimod and AMP compared to manually counted steps and measured meters walked was rated "good" on this construct. Authors report complete information on test protocol and use of a variety of structured activities: continuous walking; activity lap walking which includes walking, stopping, completing a simple task, and walking again; stair ascent and descent.

The studies by Stevens et al. in children with CP [24], McDonald et al. in children with DMD [30], and Song et al. in TDC [29] all assessed the criterion validity of the StepWatch against manual step count and scored "poor" for study design due to providing no information on parts of their study protocol (number of steps, range of steps needed, and length walked). This does not necessarily mean the design was genuinely poor, but the lack of reporting does not enable assessment on applicability in measuring habitual walking activity and has implications for the interpretation of any reported statistics. The study by Bjornson et al. [28] assessed the StepWatch for criterion validity in TDC against manual step counts and was rated "good" for study design as they report a complete test protocol, though only assessed performance during continuous walking.

The study by Aviram et al. [26] assessed the criterion validity of energy expenditure (EE) measured by the IDEEA compared to the gold standard of indirect calorimetry using the Cosmed (K4b2, Rome, Italy) which measured oxygen consumption (VO_2) in five-second epochs. Authors reported complete study protocol information and assessed a range of everyday activities in addition to comfortable and fast treadmill walking and stair climbing. The study scored "good" for study design due to not providing any evidence of the Cosmed being a gold standard measurement although it can be assumed adequate [19]. Authors discuss possible limitations of the use of the Cosmed, which include a poor fitting mask, which may lead to inaccurate measurement. This study also assessed the test-retest reliability of the IDEEA and received an "excellent" rating for study design in the assessment of this construct.

The StepWatch study in children with DMD [25] used a heart rate monitor to assess its concurrent validity over four days of wear. This study was rated "fair" for study design in the assessment of this construct as an *a priori* hypothesis of the relationship between the StepWatch and heart rate was not stated, but it was possible to deduce what was expected [19].

3.2. Rating of Statistical Methods and Clinimetric Performance of Measures. The clinimetric properties measured for all the studies reviewed are outlined in Table 2. All studies apart from the study by Bjornson et al. [28] had a sample size of less than 30 children which constitutes a "poor" score on the COSMIN checklist, while the sample size of 60–62 children in each age group found in the Bjornson et al. study [28] receives "good" rating on this item. Three of the four authors who replied had not used a power calculation to determine sample size [22, 23, 26, 27], and one author stated in their reply that the recruitment of 60 children in each group (30 boys and 30 girls) was chosen as it was expected this would "increase the likelihood of approximating a normal distribution" [28]. It is not known if Stevens et al. [24] and McDonald et al. [25] used any power calculations to determine their sample size. As the COSMIN works on a "worst score counts" algorithm, the highest score possible for all but one study is therefore "poor," but as several studies report significant results, further discussion about the clinimetric performance of measures is warranted.

Percent agreement was used to assess the criterion validity of the Minimod in children with CP and TDC by Brandes et al. [22, 27], and the use of this method leads to a "poor" rating for statistical methods according to the COSMIN as it comes under "any other statistical method" [19]. Percent agreement is a relative measure and therefore depends on reporting of the absolute values for meaningful interpretation. Authors present rich information such as the average steps needed to walk the 20-meter track (CP children) and 160-meter track (TDC) and range of steps taken [22, 27]. It is therefore possible to see that the measurement error is small. Children with CP ($n = 19$) walked an average of 79.8 steps (min: 57; max: 126), and on average the Minimod over- or underestimated by one step (agreement = 98.7%; range: 94.1 to 101.8%) [22, 27]. TDC ($n = 20$) walked an average of 273.7 steps (min: 207; max: 377), and on average the Minimod over or underestimated by one step (agreement = 99.6%; range: 98.5 to 101.5%) [22, 27]. Children with CP walked only one-third of the steps TDC walked, and a higher likelihood that agreement occurred by chance exists. Due to good reporting of study protocol and results, percent agreement was considered an "excellent" statistical method. The Minimod showed "good" accuracy and precision for the measurement of continuous walking. A limitation of this study was the assessment of continuous walking only, and therefore the criterion validity for measuring HPA is "indeterminate."

The issue of use of percent agreement arises in two other studies assessing the StepWatch in TDC. Percent agreement is used by Song et al. [29] who compare steps measured and manual step count. They found a measurement error of $3 \pm 1\%$ [29] but did not report how long children walked or how many steps they took which does not allow the assessment of absolute measurement error or the likelihood that the error was low by chance. For this same study, a Pearson correlation coefficient has been reported in a separate paper [31] which by COSMIN standards was an "excellent" statistical method, but the lack of information about the study protocol still applies as correlation could have been estimated based on a small number of steps. The use of percent agreement and Pearson correlation coefficient in this study receives a "poor" rating, and evidence of criterion validity based on this study was "indeterminate." Percent agreement was also used in the study by Bjornson et al. [28] but total steps taken are reported as ≥ 100 steps, and therefore it is possible to assess the absolute value of agreement in step count for two-three-year olds ($99.2 \pm 4.6\%$) and 4-5-year olds ($100.0 \pm 4.4\%$). In this study, the use of percent agreement therefore

rates as "excellent," and the evidence of criterion validity for continuous walking is "good" which is further strengthened by the large sample size. Criterion validity for measurement of HPA cannot be determined based on this study. A further two studies assess the use of the StepWatch in children with CP [24] and DMD [25] but do not report any statistical methods or results which was rated "poor," and criterion validity for these populations was therefore "indeterminate."

Bland-Altman plots [32] and percent of activity laps which were detected were used to assess criterion validity in the combined study of the Minimod and AMP by Kuo et al. [23] and due to this would have scored "poor" on the COSMIN checklist [19]. The lack of a specific index to summarise the degree of agreement is a limitation of the Bland-Altman technique, and inferences about the estimate cannot be performed [33]. A strength of the Bland-Altman technique is that it produces a meaningful graph and computes the confidence limits from the paired differences between the criterion method (manual step count or meters walked) and the same variables measured by the Minimod [33]. This provides us with the ability to assess both accuracy and precision of measures and allows a comparison between groups and therefore still allows a thorough assessment of the agreement between methods and can be rated as an "excellent" statistical method. The Minimod performed well in continuous walking trials compared to measured length and direct observation of steps (mean difference = −0.4 m/−0.4 steps, 95% limits of agreement = −4.7 to 4 m/−4.1 to 3.3 steps) but showed a larger random error for activity lap walking (mean diff. = −2.3 m/−0.4 steps, 95% LoA = −27.9 to 23.3 m/−87.8 to 10.4 steps) [23]. The Minimod only detected walking activity in 19–37% of stair walking trials (ascent/descent) [23]. In TDC, the Minimod performed well for continuous walking trials and poor in structured activity laps (see Table 2); however it performed better for detecting stair walking (84%) [23]. The AMP showed greater underestimation and random error in continuous walking trials (mean diff. = −4.8 m/−3.5 steps, 95% LoA = −20.1 to 10.5 m/−16.9 to 10 steps) which increased with increasing distance walked [23]. The AMP performed better than the Minimod in the structured activity lap walking trials, but still showed considerable bias difference and large random error in this trial (mean diff. = −3.6 m/−11.2 steps, 95% LoA = −19.2 to 12.0 m/−40 to 17.7 steps), however detected more of the stair climbing trials (85%) [23]. Results were similar for TDC (see Table 2) [23]. Due to the lack of an index, a specific cut-off for what classifies as a "good" result cannot be set; however the results indicate that the Minimod has "good" accuracy and precision for continuous walking. When it comes to more free-living type activities such as the structured walking trial and stair walking, the performance of the Minimod is "poor." The AMP showed "poor" precision and accuracy in both walking trials, however detected more stair trials. As the focus of this paper is the ability to measure free-living activities, both monitors score "poor" for evidence of criterion validity for measuring HPA.

A Pearson correlation coefficient was used to assess agreement between the EE measured by the IDEEA and the criterion method of indirect calorimetry using the Cosmed [26]. A Pearson correlation is considered an "excellent" statistical method by the COSMIN [19]. For the daily activities trial, total energy expenditure was used as the outcome measure. For comfortable and fast treadmill walking and stair climbing trials, energy expenditure rate (kcal/minute) was used. For children with CP and TDC the correlation between measurement outputs was "good" by COSMIN standards during all activities (CP: $r = 0.70$–0.88; TDC: $r = 0.74$–0.97, $P \leq 0.05$) [26]. A limitation of the Pearson correlation coefficient is that it only measures precision not accuracy and is therefore not a true measure of agreement [33]. This is demonstrated by the authors of this paper as the IDEEA significantly overestimated energy expenditure during the series of everyday activities and during comfortable treadmill walking both in children with CP and TDC, and during fast treadmill walking in TDC (paired t-test, $P < 0.01$) [26]. During fast treadmill walking in children with CP and during stair walking in both groups, measured energy expenditure did not differ significantly between methods (paired t-test, $P > 0.01$) [26]. A limitation of the t-test is that it will only show a significant difference if there is a systematic constant difference between two values, not if there is a systematic proportional difference as the paired difference will end up close to zero [34]. A Bland-Altman plot or a regression analysis would provide a better identification of the nature of any systematic differences in the EE estimation of the IDEEA. The present energy expenditure calculations used in the IDEEA software do not accurately assess energy expenditure, and the clinimetric properties of the IDEEA are therefore considered "poor." Authors suggest the good correlation between the IDEEA and the Cosmed indicates it can be used as a clinical follow-up tool for quantitative evaluation of efficacy of treatment interventions. As the IDEEA appears to perform better at higher intensities, systematic differences may exist which may lead to inaccurate conclusions. The reliability of EE measurement by the IDEEA was assessed in children with CP ($n = 12$) [26] using a Pearson's correlation coefficient and a paired t-test. Authors aimed to assess "agreement" between repeated measures as discussed in de Vet et al. [35]. This study therefore scored "poor" for choice of statistical method as measures can significantly correlate despite being significantly different, and the inability of a t-test to detect systematic proportional differences as discussed previously still applies [33, 34]. Agreement between repeated measures of EE using the IDEEA is therefore "indeterminate" based on this study.

Authors of the StepWatch study in children with DMD [25] used a heart rate monitor to assess its concurrent validity and received a "good" rating for statistical method as they used a Pearson correlation coefficient but did not report standard deviation. Evidence of concurrent validity was rated poor as $r < 0.70$ (CP: $r = 0.295$; TDC: $r = 0.477$; $P < 0.05$).

3.3. Clinical Utility. Clinical utility was assessed for the four identified measures and is summarised in Table 2. The Minimod and StepWatch require individual calibration and software for analysis. The StepWatch is expensive (cost not available for Minimod). The rich data collected by the

Table 2: Evidence of criterion or construct validity, reliability and utility.

Measure	Utility	Study	n, age (years), disability	Criterion validity	Construct validity	Reliability
Minimod	Cost not available	Kuo et al. [23]	$n = 17$, 4–16, CP	Mean difference ± 2SD: (−3.3 ± 2.2)–(8.9 ± 2.5) m; (−38.7 ± 49.1)–(−1.0 ± 1.7) steps; rate of activity detection: 19–97%	n/a	n/a
	Calibration and analysis software		$n = 19$, 4–16, TDC	Mean difference ± 2SD (−0.6 ± 5.2)–(7.5 ± 2.8) m; (−57.4 ± 67)–(−1.0 ± 2.0) steps; rate of activity detection for stair ascent and descent 84–100%		
	Rich data collection	Brandes et al. [22, 27]	$n = 19$, 5–16, CP	$\text{Agreement}_{\text{steps}} = 98.9\%$ (range: 94.1–101.8%) $\text{Agreement}_{\text{distance}} = 101\%$ (range not reported)		
	Feasible HPA measure		$n = 20$, 3–16, TDC	$\text{Agreement}_{\text{steps}} = 99.6 \pm 0.6\%$ (range: 98.5–101.5%) $\text{Agreement}_{\text{distance}} = 100.6 \pm 3.3\%$ (range: 93–106.7%) $\text{Agreement}_{\text{time}} = 101.3 \pm 2.8\%$ (range: 94.5–106.6%)	n/a	n/a
AMP	Cost not available No calibration Analysis software	Kuo et al. [23]	$n = 20$, 4–16, CP	Mean difference ± 2SD (−4.8 ± 15.3)–(1.3 ± 2.5) m; (−11.2 ± 28.8)–(−3.5–13.4) steps; rate of activity detection 85–95%	n/a	n/a
	Only total steps and meters walked Feasible HPA measure		$n = 20$, 4–16, TDC	Mean difference ± 2SD (−2.5 ± 7.1)–(0.7 ± 1.0) m; (−4.4 ± 14.5)–(−1.3 ± 1.6) steps; rate of activity detection: 92–100%		
StepWatch	Expensive (unit: \$500, software: \$1500)	McDonald et al. [25]	$n = 16$, 5–13, DMD	Authors state "no difference between observed and measured"; no statistical measure reported	$r = 0.295$, $P < 0.05$	
	Calibration and analysis software		$n = 20$, 5–13, TDC		$r = 0.477$, $P < 0.05$	n/a
	Rich data collection	Stevens et al. [24]	$n = 27$, 4–18, CP $n = 27$, 4–18,TDC	Authors state "readjusted until all valid step activity recorded"; no statistical measure reported	n/a	n/a
		Bjornson et al. [28]	$n = 162$, 2–5, TDC	Agreement = 99.2 ± 4.6% (2-3yr); Agreement = 100.0 ± 4.4% (4-5yr)	n/a	n/a
	Feasible HPA measure	Song et al. [29]	$n = 20$, 5–11, TDC	Walking: $r = 0.97$; Running: $r = 0.96$; ($P < 0.05$); measurement error 3 ± 1%	n/a	n/a
IDEEA	Cost not available Poor battery life (60 hrs) Poor utility in HPA research	Aviram et al. [26]	$n = 21$, 4–10, CP	$r = 0.72$ ($P < 0.001$); difference means (t-test): $P = 0.000$	n/a	Test-retest $r \geq 0.995$; Difference mean: t-test: $P = 0.33$
			$n = 7$, 5–8, TDC	$r = 0.88$ ($P < 0.05$); difference means (t-test): $P = 0.002$		

CP: cerebral palsy; DMD: Duchenne muscular dystrophy; TDC: typically developing children; AMP: ambulatory monitoring Pod; IDEEA: Intelligent Device for Energy Expenditure and Activity; n/a: not assessed.

Table 3: Evidence of criterion validity.

Measure	Study	Study design	Statistical method	Criterion validity for HPA
Minimod	Kuo	++	+++	−
	Brandes	++	+++	u/a
AMP	Kuo	++	+++	−
IDEEA	Aviram	++	+++	−
StepWatch	Stevens	−	u/a	u/a
	McDonald	−	u/a	u/a
	Song (TDC only)	−	−	u/a
	Bjornson (TDC only)	++	+++	u/a

IDEEA: Intelligent Device for Energy Expenditure and Activity; TDC: typically developing children; (+++): excellent; (++): good; (+): fair, (−): poor, (u/a): unable to assess/indeterminate.

StepWatch and Minimod allows measurement of intensity, frequency, and duration of walking activity. The units are both small and unobtrusive and have battery lives of seven days for the Minimod and up to six weeks for the StepWatch, depending on settings. This makes them both feasible tools for the measurement of habitual walking activity. In the event that data collection is delayed once the device is provided to the child's parents, having a battery life of more than seven days, and therefore not requiring recharging is a strength of the StepWatch as there is less likelihood of loss of data. The Minimod is worn around the waist centred at the lower lumbar spine, while the StepWatch is worn on the ankle in a small cuff. Consistency of placement for repeated and all-day wear may be easier to achieve with an ankle placement which would reduce measurement error due to inconsistent placement. Both pedometers were considered to have good clinical utility. The AMP is small, worn around the ankle, does not require calibration or software analysis, and has a good battery life; however it only measures total step count or meters walked per wear period. The AMP was considered to have good clinical utility. The IDEEA consists of a data recorder worn on the waist with 5 individual sensors attached to the chest, thighs, and under each foot connected by wires to allow measurement of postures and energy expenditure, and it only has a battery life of 60 hours. Both of these factors limit the IDEEAs utility in the measurement of HPA in young children.

3.4. Level of Evidence. A summary of the quality of evidence for criterion validity is provided in Table 3. Two studies assess the Minimod in children with CP and TDC in samples which include children under the age of six. Both studies were rated "good" for study design and "excellent" for statistical method. They provided "good" evidence of the ability of the Minimod to accurately measure continuous walking. One of these studies [23] also used free-living type activities, and the Minimod displayed "poor" accuracy and precision in this setting. The AMP was assessed in one study of "good" study design quality which used an "excellent" statistical method, though displayed "poor" accuracy and precision in both continuous walking and free-living type activities.

Four studies assess the StepWatch in samples which included children under the age of six. Reporting of study protocol and statistical method was "poor" in three of these studies. The fourth study was of "good" quality and used an "excellent" statistical method. It provided "good" evidence of the ability of the StepWatch to accurately measure continuous walking in TDC. The ability of the StepWatch to accurately measure free-living activities cannot be determined based on this study. The ability of the IDEEA to measure EE was assessed in one study of "good" study design quality which used an "excellent" statistical method; however the IDEEA displayed "poor" accuracy and precision in both free-living type activities and continuous walking trials.

4. Discussion

This paper has systematically reviewed the clinimetric properties (validity, reliability, and clinical utility) of four measures of HPA in children with a motor disability which included children aged less than six years. A precise and accurate measurement of the daily physical activity levels in this population will allow researchers to investigate the dose-response relationship between HPA and health outcomes. It will also allow the assessment of the efficacy of interventions aimed at increasing HPA in terms of establishing the dose and distribution of treatment necessary to achieve worthwhile results in the long term. For clinicians, an accessible, precise, and accurate measurement tool would allow the identification of children with low levels of HPA and in turn the assessment of the effectiveness of prescribed interventions.

Promising measures of HPA have been assessed in young children with motor disabilities, and the methodological quality of the papers was good to poor for study design and excellent to poor for statistical methods. A limitation of the findings of this paper is that only two studies of overall "good" methodological quality assessed children while undertaking free-living type activities. Under these conditions, both activity monitors displayed poor accuracy and precision [23, 26]. Two activity monitors displayed "good" accuracy during continuous walking [23, 28]. This relates more to the ICF definition of walking "capacity" rather than habitual walking activity (i.e., how many steps does a child need to walk a set distance in an ideal environment compared to how many steps do they take during the day in a variety of settings and intensities). Another limitation is that studies included in this paper assessed

children across a wide age range (4–18 years), with no studies of children with motor disabilities exclusively focusing on the under six-year age group which limits this review's ability to make specific recommendations for this age group. The proportion of children aged less than six in the study samples ranged from 5 to 43% for the studies which provided a breakdown of their sample [22, 23, 26, 27]. The use of the COSMIN checklist for the assessment of methodological quality together with quality rating criteria of measurement properties is also a possible limitation. The COSMIN checklist was developed for assessing HR-PROs and does not have established psychometric properties in assessing objective physical activity measurement tools, and the quality rating criteria were not developed based on consensus. This issue was minimised by being guided by a systematic review of activity monitors in TDC for the assessment of the somewhat ambiguous "flaws in design or methods" item used in the COSMIN (Box H, item 5) [19] and by providing an in-depth appraisal of the statistical methods.

The current Australian physical activity guidelines recommend children aged from 1 to 5 years engage in active play for at least three hours per day without specific intensity recommendations [2]. This recognises the sporadic and intermittent nature of young children's activity patterns and places demands on the measurement tools used to assess HPA in this population. Firstly they have to be able to record activity as a measure of time, and secondly they need to accurately recognise "active play" behaviours. The accelerometer-based pedometers StepWatch and Minimod have the ability to collect the most complete information on habitual walking activity as they record steps within an epoch of time which can be downloaded onto a computer to visualise an activity pattern. They allow the assessment of intensity (e.g., steps/min), frequency (e.g., number of walking bouts), and duration (e.g., length of walking bouts). The validation protocols of the included studies primarily used structured walking activities which may not accurately represent the way young children move in active play, and therefore it cannot be assumed activity monitors that do well in continuous walking trials would have done equally well had the children's steps been counted during free-living play. This was demonstrated in the assessment of the Minimod by Kuo et al. [23]. In typically developing children, activity monitors are usually validated by direct observation of free play to circumvent this issue, and validation studies typically use a narrower age range to control for differences in motor skills [36, 37].

Children with a motor disability may not be able to walk but instead use a range of other methods of ambulation such as crawling, cruising, rolling, bottom shuffling, walking aids, or wheelchairs. Those who are able to walk may have different gait patterns than typically developing children [14]. This could explain why the accelerometer Minimod, which relies on recognising gait patterns to count steps, had a lower rate of activity detection in children with CP (19–97%) than in typically developing children (84–100%) [23]. Other measurement tools such as accelerometers, which report raw activity counts per epoch of time, bypass this limitation but are yet to be validated in young children with a motor disability. A systematic review of measures of HPA in TDC preschoolers identified the ActiGraph (Shalimar, FL, USA) accelerometer as having the best clinimetric properties in this population. Similarly, the systematic review of HPA measurements in adolescents with CP by Clanchy et al. [16] identified accelerometers as the most comprehensive measure of HPA patterns despite the limited evidence available, and the ActiGraph has since been validated in adolescents with CP [38, 39]. Studies of doubly labelled water [40, 41], the Compendium for Physical Activity Questionnaires [3] and ActiGraph in infants at risk of neurodevelopmental delay [42], did not meet inclusion criteria as their clinimetric properties for the measurement of HPA in children with motor disabilities aged less than six years had not been assessed.

5. Conclusion

This systematic review identified four measures of HPA with evidence of clinimetric properties in study samples which included children aged less than six with motor disabilities. Only a very small number of studies assessing activity monitors in this population are available, and none of the studies focus exclusively on children aged less than six years. The pedometers StepWatch and Minimod are the most comprehensive measures of habitual walking activity utilised in the current literature. While both demonstrate good accuracy for step count during continuous walking, only the Minimod has been tested during conditions which included walking trials other than continuous walking, and it performed poorly during these conditions. It is possible the ankle placement of the StepWatch would allow a more accurate assessment of free-living walking activities in children with motor disabilities but this is yet to be demonstrated. Pedometers are only suitable as an estimate of HPA for children with high functional capacity as children's HPA patterns are likely to consist of a progressively smaller proportion of walking as the severity of their impairment increases. In the most severely impaired children walking activity is completely nonexistent. Further research is needed to ascertain the clinimetric properties of activity monitors available for measuring HPA in young children with motor disabilities, and testing protocols should include a range of activities and ideally direct observation of free play. This will enable an understanding of the HPA patterns of children with motor disabilities across the spectrum of functional capacity for clinicians and researchers alike.

Conflict of Interests

No conflict of interests such as financial gain exists between the authors of this paper and the companies producing the equipment which is discussed in the paper.

Acknowledgments

The authors would like to thank the National Health and Medical Research Council (NHMRC) for Project Grant

(465128) "Qld CP Child-Queensland prospective cohort study of children with cerebral palsy" and Project Grant (569605) "Qld CP Child-Growth, Nutrition and Physical Activity"; the Queensland Children's Medical Research Institute PhD Scholarship (SO); the Australian Postgraduate Award (APA) Scholarship (SO, LM) and the Smart State Fellowship from the Queensland Government (RNB). All sources of funding are project grants or stipends for researchers. The funding bodies had no involvement in the study design, analysis or interpretation or data, the writing of the manuscript or in the decision to submit the manuscript for publication.

References

[1] S. Trost, "Discussion paper for the development of recommendations for children's and youths' participation in health promoting physical activity," Australian Department of Health and Ageing (Editor), 2005.

[2] Department of Health and Ageing, *National Physical Activity Recommendations for Children 0–5 Years*, Australian Department of Health and Ageing, 2010.

[3] J. N. Zwier, P. E. van Schie, J. G. Becher et al., "Physical activity in young children with cerebral palsy," *Disability and Rehabilitation*, vol. 32, no. 18, pp. 1501–1508, 2010.

[4] E. G. Fowler, T. H. Kolobe, D. L. Damiano et al., "Promotion of physical fitness and prevention of secondary conditions for children with cerebral palsy: section on pediatrics research summit proceedings," *Physical Therapy*, vol. 87, no. 11, pp. 1495–1510, 2007.

[5] D. L. Damiano, "Activity, activity, activity: rethinking our physical therapy approach to cerebral palsy," *Physical Therapy*, vol. 86, no. 11, pp. 1534–1540, 2006.

[6] C. J. Caspersen, K. E. Powell, and G. M. Christenson, "Physical activity, exercise and physical fitness: definitions and distinctions for health-related research," *Public Health Reports*, vol. 100, no. 2, pp. 126–131, 1985.

[7] International Classification of Functioning D.a.H.I., World Health Organization, Geneva, Switzerland, 2002.

[8] V. Penpraze, J. J. Reilly, C. M. MacLean et al., "Monitoring of physical activity in young children: how much is enough?" *Pediatric Exercise Science*, vol. 18, no. 4, pp. 483–491, 2006.

[9] G. J. Welk, C. B. Corbin, and D. Dale, "Measurement issues in the assessment of physical activity in children," *Research Quarterly for Exercise and Sport*, vol. 71, no. 2, supplement, pp. S59–S73, 2000.

[10] G. J. Welk, *Physical Activity Assessments for Health-Related Research*, Human Kinetics, 2002.

[11] J. R. Sirard and R. R. Pate, "Physical activity assessment in children and adolescents," *Sports Medicine*, vol. 31, no. 6, pp. 439–454, 2001.

[12] J. J. Reilly, "Physical activity, sedentary behaviour and energy balance in the preschool child: opportunities for early obesity prevention," *Proceedings of the Nutrition Society*, vol. 67, no. 3, pp. 317–325, 2008.

[13] S. I. de Vries, H. W. J. E. M. van Hirtum, I. Bakker, M. Hopman-Rock, R. A. Hirasing, and W. Van Mechelen, "Validity and reproducibility of motion sensors in youth: a systematic update," *Medicine and Science in Sports and Exercise*, vol. 41, no. 4, pp. 818–827, 2009.

[14] L. A. Prosser, R. T. Lauer, A. F. VanSant, M. F. Barbe, and S. C. K. Lee, "Variability and symmetry of gait in early walkers with and without bilateral cerebral palsy," *Gait and Posture*, vol. 31, no. 4, pp. 522–526, 2010.

[15] C. M. Capio, C. H. P. Sit, B. Abernethy, and E. R. Rotor, "Physical activity measurement instruments for children with cerebral palsy: a systematic review," *Developmental Medicine and Child Neurology*, vol. 52, no. 10, pp. 908–916, 2010.

[16] K. M. Clanchy, S. M. Tweedy, and R. Boyd, "Measurement of habitual physical activity performance in adolescents with cerebral palsy: a systematic review," *Developmental Medicine and Child Neurology*, vol. 53, no. 6, pp. 499–505, 2011.

[17] Department of Health and Ageing, *National Physical Activity Recommendations for Children 6–12 Years*, C.o.A. Department of Health and Ageing, 2010.

[18] L. B. Mokkink, C. B. Terwee, D. L. Patrick et al., "The COSMIN checklist for assessing the methodological quality of studies on measurement properties of health status measurement instruments: an international Delphi study," *Quality of Life Research*, vol. 19, no. 4, pp. 539–549, 2010.

[19] C. B. Terwee, L. B. Mokkink, D. L. Knol et al., "Rating the methodological quality in systematic reviews of studies on measurement properties: a scoring system for the COSMIN checklist," *Quality of Life Research*, vol. 21, no. 4, pp. 651–657, 2012.

[20] C. B. Terwee, S. D. M. Bot, M. R. de Boer et al., "Quality criteria were proposed for measurement properties of health status questionnaires," *Journal of Clinical Epidemiology*, vol. 60, no. 1, pp. 34–42, 2007.

[21] L. B. Mokkink, C. B. Terwee, D. L. Knol et al., "The COSMIN checklist for evaluating the methodological quality of studies on measurement properties: a clarification of its content," *BMC Medical Research Methodology*, vol. 10, article 22, 2010.

[22] M. Brandes, S. Heikens, W. Zijlstra et al., "Basic gait parameters of healthy and cp children assessed by accelerometry," in *Proceedings of the International Society of Biomechanics XXth Congress*, Cleveland, Ohio, USA, 2005.

[23] Y. L. Kuo, K. M. Culhane, P. Thomason, O. Tirosh, and R. Baker, "Measuring distance walked and step count in children with cerebral palsy: an evaluation of two portable activity monitors," *Gait and Posture*, vol. 29, no. 2, pp. 304–310, 2009.

[24] S. L. Stevens, E. A. Holbrook, D. K. Fuller, and D. W. Morgan, "Influence of age on step activity patterns in children with cerebral palsy and typically developing children," *Archives of Physical Medicine and Rehabilitation*, vol. 91, no. 12, pp. 1891–1896, 2010.

[25] C. M. McDonald, L. M. Widman, D. D. Walsh, S. A. Walsh, and R. T. Abresch, "Use of step activity monitoring for continuous physical activity assessment in boys with Duchenne muscular dystrophy," *Archives of Physical Medicine and Rehabilitation*, vol. 86, no. 4, pp. 802–808, 2005.

[26] R. Aviram, M. Belokopytov, S. Ben-Chaim, and A. Rotstein, "Evaluation of energy expenditure in children with cerebral palsy using a multi-sensor accelerometer," *The Journal of Sports Medicine and Physical Fitness*, vol. 51, no. 3, pp. 506–514, 2011.

[27] M. Brandes, W. Zijlstra, S. Heikens, R. van Lummel, and D. Rosenbaum, "Accelerometry based assessment of gait parameters in children," *Gait and Posture*, vol. 24, no. 4, pp. 482–486, 2006.

[28] K. Bjornson, K. Song, J. Lisle et al., "Measurement of walking activity throughout childhood: influence of leg length," *Pediatric Exercise Science*, vol. 22, no. 4, pp. 581–595, 2010.

[29] K. M. Song, K. F. Bjornson, T. Cappello, and K. Coleman, "Use of the stepwatch activity monitor for characterization

of normal activity levels of children," *Journal of Pediatric Orthopaedics*, vol. 26, no. 2, pp. 245–249, 2006.

[30] C. M. McDonald, L. Widman, R. T. Abresch, S. A. Walsh, and D. D. Walsh, "Utility of a step activity monitor for the measurement of daily ambulatory activity in children," *Archives of Physical Medicine and Rehabilitation*, vol. 86, no. 4, pp. 793–801, 2005.

[31] K. F. Bjornson, B. Belza, D. Kartin, R. Logsdon, and J. F. McLaughlin, "Ambulatory physical activity performance in youth with cerebral palsy and youth who are developing typically," *Physical Therapy*, vol. 87, no. 3, pp. 248–257, 2007.

[32] J. M. Bland and D. G. Altman, "Statistical methods for assessing agreement between two methods of clinical measurement," *The Lancet*, vol. 1, no. 8476, pp. 307–310, 1986.

[33] L. Lin, "Overview of agreement statistics for medical devices," *Journal of Biopharmaceutical Statistics*, vol. 18, no. 1, pp. 126–144, 2008.

[34] K. Linnet, "Limitations of the paired *t*-test for evaluation of method comparison data," *Clinical Chemistry*, vol. 45, no. 2, pp. 314–315, 1999.

[35] H. C. W. de Vet, C. B. Terwee, D. L. Knol, and L. M. Bouter, "When to use agreement versus reliability measures," *Journal of Clinical Epidemiology*, vol. 59, no. 10, pp. 1033–1039, 2006.

[36] M. Oliver, G. M. Schofield, G. S. Kolt, and P. J. Schluter, "Pedometer accuracy in physical activity assessment of preschool children," *Journal of Science and Medicine in Sport*, vol. 10, no. 5, pp. 303–310, 2007.

[37] E. van Cauwenberghe, J. Gubbels, I. de Bourdeaudhuij, and G. Cardon, "Feasibility and validity of accelerometer measurements to assess physical activity in toddlers," *International Journal of Behavioral Nutrition and Physical Activity*, vol. 8, no. 1, article 67, 2011.

[38] K. M. Clanchy, S. M. Tweedy, R. N. Boyd, and S. G. Trost, "Validity of accelerometry in ambulatory children and adolescents with cerebral palsy," *European Journal of Applied Physiology*, vol. 111, no. 12, pp. 2951–2959, 2011.

[39] C. M. Capio, C. H. Sit, and B. Abernethy, "Physical activity measurement using MTI (actigraph) among children with cerebral palsy," *Archives of Physical Medicine and Rehabilitation*, vol. 91, no. 8, pp. 1283–1290, 2010.

[40] K. Bell, "Energy expenditure, body composition and physical activity levels of children with cerebral palsy," *The American Journal of Clinical Nutrition*, vol. 32, pp. 313–319, 2005.

[41] V. A. Stallings, B. S. Zemel, J. C. Davies, C. E. Cronk, and E. B. Charney, "Energy expenditure of children and adolescents with severe disabilities: a cerebral palsy model," *The American Journal of Clinical Nutrition*, vol. 64, no. 4, pp. 627–634, 1996.

[42] R. M. Angulo-Barroso, C. W. Tiernan, L. C. Chen, D. Ulrich, and H. Neary, "Treadmill responses and physical activity levels of infants at risk for neuromotor delay," *Pediatric Physical Therapy*, vol. 22, no. 1, pp. 61–68, 2010.

Hindawi Publishing Corporation
International Journal of Pediatrics
Volume 2012, Article ID 387280, 7 pages
doi:10.1155/2012/387280

Research Article

Play and Be Happy? Leisure Participation and Quality of Life in School-Aged Children with Cerebral Palsy

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Received 4 April 2012; Accepted 21 June 2012

Academic Editor: Maria A. Fragala-Pinkham

The objective of this study was to examine the association between leisure participation and quality of life (QoL) in school-age children with cerebral palsy (CP). Leisure participation was assessed using the Children's Assessment of Participation and Enjoyment (CAPE) and QoL using the Pediatric Quality of Life Inventory (PedsQL). Pearson correlation coefficients were calculated to examine the association between CAPE and PedsQL scores, and a multiple linear regression model was used to estimate QoL predictors. Sixty-three children (mean age 9.7 ± 2.1 years; 39 male) in GMFCS levels I–V were included. Intensity of participation in active-physical activities was significantly correlated with both physical ($r = 0.34$, $P = 0.007$) and psychosocial well-being ($r = 0.31$, $P = 0.01$). Intensity and diversity of participation in skill-based activities were negatively correlated with physical well-being ($r = -0.39$, $P = 0.001$, and $r = -0.41$, $P = 0.001$, resp.). Diversity and intensity of participation accounted for 32% ($P = 0.002$) of the variance for physical well-being and 48% ($P < 0.001$) when age and gross motor functioning were added. Meaningful and adapted leisure activities appropriate to the child's skills and preferences may foster QoL.

1. Introduction

Cerebral palsy (CP) is a broad term that describes a set of conditions that is associated with major physical impairments and other developmental deficits and arises in the early stages of brain development [1]. CP is the most common type of physical disability affecting children in developed countries [2] with an estimated prevalence of 2.0 to 2.5/1000 children [3]. Despite the nonprogressive nature of the condition, the nature of functional impairments may change as the child develops. As a consequence, evolving limitations in everyday activities may be experienced, with possible impact on the individual's overall health and well-being [4, 5]. The type and distribution of movement disorder in CP may be categorized as spastic, dyskinetic, hypotonic, or mixed. Spastic patterns are the most common and may be further differentiated as diplegia, quadriplegia, hemiplegia, or monoplegia, relating to the limbs involved [6, 7].

Individuals with CP have been an important target population requiring rehabilitation services. The traditional focus of health care services for this population has been primarily directed at rehabilitation interventions that address the underlying motor and other developmental impairments, such as abnormal muscle tone, decreased attention span, poor dexterity, or difficulties with perceptual concepts, as well as limitations in essential daily self-care skills and mobility. In the last decade, however, increasing interest has been attributed to the quality of life (QoL) and participation of children with CP. Researchers and clinicians are concerned with the extent to which children with CP have

the opportunity to be involved and enjoy leisure activities at home and in the community and the extent to which they report a good QoL [8, 9].

QoL is a broad concept encompassing many components of overall health and well-being (e.g., physical, psychosocial, economic, and cultural). It is influenced by the context of the culture and value systems in which the individual lives and relates to the individual's goals, expectations, standards, and concerns [10]. The QoL of children who live with a neurological condition can be impacted at different levels, including physical (e.g., physical health, independence in basic functional activities), psychological (e.g., mental status, positive self-perception), and psychosocial dimensions (e.g., forming friendships, leisure time, finding a partner) [11]. Identifying the factors that are associated with better or poorer QoL is important [12] as this can be used to guide program planning and the allocation of resources, thus optimizing the well-being of these children.

Studies have shown that individuals with disabilities do not necessarily have a lower QoL and a diminished perception of their well-being or general dissatisfaction with life [13, 14]. Recent studies are exploring a variety of factors that may contribute to a good QoL in children with neurodevelopmental disabilities; however, most studies explore aspects related to body function and activity limitations.

Participation in leisure activities includes participation in sports, arts, entertainment, social, self-improvement, and religious activities [15]. Engaging in leisure activities may be influenced by the child's personal factors, environmental factors, and underlying health condition [15]. By participating in leisure activities, children may develop competencies, achieve mental and physical health, gain an understanding of their strengths and abilities, and form lasting meaningful friendships and relationships. It is through participating that children make contributions to their community, learn about themselves and the expectations of society, and develop skills needed to become successful and autonomous in their home, school, and community [16, 17]. For these reasons, participation is increasingly considered as one of the primary aims of pediatric rehabilitation and is believed to contribute to child health, development, and QoL [16, 18]. Researchers have shown that multiple factors may contribute to a perceived good QoL in children with physical disabilities [17], including the child's participation in a variety of leisure activities [19].

Participation in leisure is an objective, tangible outcome that can be incorporated and measured in rehabilitation and health programs, while QoL is a subjective outcome [20]. Although frequently assumed, the association between leisure participation and QoL has not been clearly delineated [21]. The objective of this study was to examine the association between involvement in leisure activities and QoL in children between 6 to 12 years of age with CP.

2. Methods

2.1. Population. This study used a cross-sectional design involving a historical cohort of children with CP seen by a single neurologist over a 10-year period (1991–2001) in a variety of settings (private office, hospital, neonatal clinic, suburban private clinic). This sample has been described previously [22–25]. This sample is representative of children with CP from a local community who are routinely sent to a pediatric neurologist for diagnosis, etiological determination, and referral for rehabilitation services. Exclusion criteria were children presenting progressive disorders, disorders of noncerebral origin, and specific syndromes. Parents who could not easily read or converse in English or French were excluded from participation.

A total of 63 children and their parents completed the leisure participation and QoL questionnaires. Twenty-one participants who could not actively participate in the completion of the leisure participation questionnaire were excluded. Those participants were children with more severe cognitive impairment and therefore who could not actively contribute to the completion of these self-report questionnaires. These children were more likely to be level IV or V GMFCS. Demographic and clinical characteristics of the participants and also of the children who could not complete the questionnaires (nonparticipants) are described in Table 1.

2.2. Procedures. Scientific and ethical approval was obtained through the McGill University Health Centre, Montreal Children's Hospital's institutional review board. Information letters describing the study were sent to parents of children with a diagnosis of CP between the ages of 6 and 12 years. Informed signed consent was obtained from a parent, and assent was obtained from children when feasible. A 3-hour visit was scheduled at the Montreal Children's Hospital to conduct required assessments. Participants completed a series of developmental evaluations administered by a neurologist and a physical therapist and/or occupational therapist. As part of this study, the neurologist assessed the level of gross motor function for each participant using the 5-level Gross Motor Function Classification System (GMFCS) [26]. The Gross Motor Function Measure (GMFM-66) [27] was administered by a physical or occupational therapist during the visit. Parents (and children when feasible) completed questionnaires assessing leisure participation and QoL and also completed a demographic questionnaire.

2.3. Outcome Measures. The Pediatric Quality of Life Inventory (PedsQL) Generic Core Scales [28] were used to assess quality of life in the physical, emotional, social, and school domains. This questionnaire includes 23 items, and the parent proxy-report version was used in this study. The child-report version was also used when feasible, but, due to smaller numbers, the parent report was used for analysis in this paper due to the small sample of children who were able to complete the child version independently. Individual domain scores were calculated, as well as psychosocial and physical summary scores. The psychosocial well-being summary score represents the way the child feels about their social life, their school functioning, and their emotional well-being. The physical summary score represents the ease in which one can get around and do basic activities, without pain while maintaining a good energy level. Internal

Table 1: Characteristics of participants.

$N = 63$		Study participants ($n = 63$)	Nonparticipants ($n = 21$)
Mean age		9.7 ± 2.1 years	8.69 ± 1.9 years
Gender		62% male	52% male
Gross Motor Function Classification System (GMFCS)	Level I	59%	10%
	Level II	22%	—
	Level III–V	18%	90%
CP distribution	Spastic quadriplegia	24%	61%
	Spastic hemiplegia	35%	9.5%
	Spastic diplegia	24%	9.5%
	Other	17%	4.8%
School setting	Regular school	56%	24%
	Special school	44%	76%
Rehabilitation services	Received rehabilitation services in the past 6 months	84%	100%
Socioeconomic status (combined household income before taxes)	0–$19,000	12%	13%
	$20,000–$39,000	27%	27%
	$40,000–$59,000	21%	20%
	$60,000–$79,000	14%	27%
	$80,000+	26%	13%

consistency reliability of the PedsQL is excellent (alphas > 0.90). The validity of the PedsQL Generic Core Scales has been demonstrated through known groups comparisons and correlations with other measures of disease burden [28].

The Children's Assessment of Participation and Enjoyment (CAPE) [29] was used to assess participation in leisure activities. This 55-item child self-report questionnaire was designed for children and youth with and without disabilities between the ages of 6 and 21 years. It provides information on five dimensions of participation (diversity, intensity, where, with whom, and enjoyment) and five types of activities (recreational, active-physical, social, skill-based, and self-improvement). Current evidence supports the tool's validity, and reliability is adequate [30]. Parental assistance was sought when children had difficulty completing this questionnaire; however, children actively contributed in the completion of the CAPE. For the purposes of this study, intensity (how often does the child participate in each activity) and diversity (how many different activities a child engages in) scores in the five different activity types were used.

2.4. Data Analysis. Descriptive statistics were used to describe the sample. Participants (children with CP) who could not actively participate in completion of the CAPE were excluded from analysis. Pearson correlation coefficients were computed to examine the association between leisure participation (CAPE) and QoL (PedsQL). A multiple linear regression model was estimated with QoL (psychosocial and physical summary scores) as the dependent variable and leisure participation as the independent variable. Models were also derived including age and level of motor function using GMFM scores.

Diagnostic tests were used to check for violation of the assumptions inherent in linear regression models. Due to the lack of evidence on specific leisure domains that may predict QoL, forced entry regression was used to determine the significant predictors of psychosocial and physical QoL. This method is likely not influenced by random variation in the data and is therefore appropriate for using theory testing [31].

3. Results

When reporting their child's QoL, about half of the parents reported that their child had low well-being (more than one standard deviation below normative means) in the physical and psychosocial well-being domains. Well-being in emotional functioning and school functioning was within normative means for about 60% of the children as reported by their parents (Table 2). Children participated in a variety of leisure activities, but mostly in informal activities. Intensity scores measure how often the child has engaged in a given activity in the past four months (ranging from once in the past four months to everyday or more). The intensity of recreational activities (e.g., doing crafts) was higher than the other four activity domains, followed by social activities (e.g., talking on the phone) and self-improvement activities (e.g., reading) (Table 3). A detailed description of participation in leisure activities and QoL for this population has been published elsewhere [22–24].

The relationship between involvement in leisure activities (i.e., diversity and intensity of participation in activities) and parent-proxy report of child's QoL was tested. Intensity of participation, but not diversity, in active-physical activities such as team sports, bicycling, water and snow sports,

Table 2: Performance on PedsQL domains.

PedsQL parent-report $N = 63$	Score per domain mean (range)	Abnormal mean score (<1 SD from normative data)
Emotional functioning	68.5 (40–100)	39.7%
Social functioning	58.3 (25–100)	50.8%
School functioning	62.9 (25–100)	39.7%
Physical functioning	62.1 (12.5–100)	50.8%
Physical summary score	62.1 (12.5–100)	50.8%
Psychosocial summary score	63.2 (35–100)	55.6%

Table 3: Mean scores of domains of participation in leisure activities as assessed by the CAPE.

Domain of participation $N = 63$	Diversity mean (range)	Frequency mean (range)	Enjoyment mean (range)
Recreational activities	9.34 (5–12)	4.10 (1.6–6)	4.28 (2.9–5.1)
Active physical activities	3.03 (0–7)	1.68 (0–4.33)	4.49 (2.6–5.3)
Social activities	6.87 (4–9)	3.12 (1.5–4.7)	4.37 (2.7–5.3)
Skill-based activities	2.8 (0–6)	1.45 (0–3.8)	4.12 (1–5)
Self-improvement activities	5.58 (1–9)	2.82 (0.6–5.6)	3.5 (1.5–5)
Formal activities	3.87 (0–9)	1.28 (0–3)	4.06 (0–5)
Informal activities	23.77 (13–31)	3.3 (1.8–4.8)	4.16 (3–5)

Scoring: Diversity (number of different activities) = number of activities involved in compared to total number of activities. Intensity (how often) = (1) 1x/4 months, (2) 2x/4 months, (3) 1x/month, (4) 2-3x/month, (5) f1x/week, (6) 2-3x/week, (7) daily. Enjoyment (how much do you enjoy) = (1) not at all, (2) somewhat, sort of, (3) pretty much, (4) very much, (5) love it.

and other individual physical activities was significantly correlated with physical well-being ($r = 0.34$, $P = 0.007$). Intensity and diversity of involvement in skill-based activities, that is, of activities such as dancing, arts, and music classes done with an instructor, were negatively correlated with physical well-being ($r = -0.39$, $P = 0.001$ for intensity and $r = -0.41$, $P = 0.001$ for diversity, resp.). Intensity of participation in active-physical activities also accounted for better psychosocial well-being ($r = 0.31$, $P = 0.01$) of children with CP.

In a multiple regression model, diversity and intensity of participation in five domains of leisure accounted for 32% ($P = 0.002$) of the variation in physical well-being. However, when age and gross motor functioning (GMFM score) were included in the model, this value increased to 48% ($P < 0.001$) of the variance in the physical well-being domain (Table 4). None of the multiple regression models was significant for psychosocial well-being.

4. Discussion

This study describes the association between participation in leisure activities and QoL in school-aged children with CP. Results indicate a positive association between engagement in physical activities and both physical and psychosocial well-being. These findings suggest that school-aged children with CP who participate more actively in physical activities get around more easily to do basic activities, without pain and with a good energy level, and feel better about their social life, school functioning, and their emotions, according to parent-report. Inversely, it is possible that children who generally feel better in these domains naturally participate more in active physical activities. Skill-based activities and physical well-being were negatively related, suggesting that other factors may play a role in this relationship. For instance, motor functioning alone accounted for a high variance in the outcome variable in the multivariate model. Indeed, child attributes such as the severity of motor dysfunction, age, and gender were previously described as important predictors of physical well-being, a component of a child's QoL [22, 32, 33]. Studies have shown that, with increasing age, children tend to decrease their participation in out of school leisure activities [17]; therefore, it is important to motivate them to maintain and increase their activity level as they develop.

Other personal and environmental factors may mediate the association between leisure participation and QoL. For instance, children and families who do not have the environmental supports and adaptations they require may not be able to participate in leisure activities of their own choosing, which is related to the child's QoL. Furthermore, children with more severe motor limitations often attend segregated school environments where they are exposed to more intense rehabilitation services and adapted leisure activities as compared to children in an integrated school setting. Children in special schools may have the opportunity to participate in activities such as adapted horseback riding, adapted arts, and swimming programs. Half of the parents of school-aged children with CP in our study reported their children as having poorer physical well-being (<1 SD of the normative mean) compared to parents of typically developing peers. This finding is probably related to their motor limitations, which may also explain our result that higher levels of participation in skill-based activities were associated with lower physical well-being. Skill-based activities (e.g., dance lessons, karate lessons) may be challenging for children with disabilities, especially if the demands of the particular

Table 4: Best predictive models of physical well-being (PedsQL).

Model variables ($N = 60$)	R^2 (P)	Best predictors beta (P value)
Diversity of leisure activities (all domains)	0.23 (<0.005)	Skill-based activities −0.49 (<0.001)
Diversity and intensity of leisure activities, age, and motor functioning	0.48 (<0.001)	GMFM-66 0.61 (<0.001)

task are not adapted to the child's capabilities. Families should be assisted in choosing appropriate and meaningful activities and learning about available adapted sports and recreation programs in their community to foster a sense of mastery and competence. In this regard, it is interesting to observe that children in adapted or segregated schools may have more opportunities to participate in skill-based activities as adapted skill-based activities may be provided by the school or referred to by professionals in the school, in spite of their higher motor limitations.

Physical activity has substantial benefits to the health and well-being of children and adolescents. Active-physical pursuits are known to improve cardiovascular and emotional health, motivating children to stay physically active and contributing to a sense of well-being [34, 35]. Participation in leisure activities also fosters friendships and other social relationships, often creating a social support network that may contribute to the individual's overall well-being [19]. In congruence with the findings of our study, a recent study with 120 school-aged children with CP using different measurement tools reported a significant relationship between the intensity of participation in leisure activities with physical well-being, emotional well-being, and social support and peers [32]. This particular study measured leisure participation more globally; however, we applied a measure with five subscales of different types of leisure activities. Our study indicates that it is the active-physical leisure activities specifically that are most strongly related to aspects of QoL.

Associations between emotional and psychosocial well-being and participation in leisure have also been reported when children engage in everyday activities or social activities [32, 36]. Qualitative studies have shown how children and adolescents with disabilities place a high value on participating in leisure activities. Youth with CP and their parents reported that participation in chosen and enjoyable activities has a positive impact on their well-being [37–42]. A recent analysis of the association between leisure participation and QoL [19] in children with neurodevelopmental disabilities found evidence from quantitative, qualitative, and mixed-methods studies supporting the relationship between participation in leisure and different domains of QoL [32, 33, 35–38]. Similarly to our study, others have found a positive association between engagement in active physical activities and physical well-being in children and youth with CP [32, 35, 43]. However, the measurement of both constructs (QoL and participation) is very variable across studies, limiting interpretability for clinical practice. Our study, however, is the first to report on the relationship between psychosocial and physical domains of QoL, specifically with respect to leisure participation in school-age children with CP.

5. Limitations

Measurement issues should be taken into consideration when analyzing findings of this study. The CAPE measures the activities that the child actually does (i.e., performance), which may or may not be in line with their preferences or personal choices. A strong linear relationship between CAPE diversity and intensity scores may account for a limited capacity of the model to represent the sample and limit the size of regression coefficients in the regression models. A moderate correlation between levels of actual involvement in active-physical pursuits with the level of preferences for these types of activities was previously shown [25], implying that children may be participating in activities that they do not necessarily enjoy. Programs and interventions should consider the preferences of children and families in order to individually tailor the level of engagement in activities to their preferences (i.e., through adapted programs or those that have no environmental barriers) and thus foster better overall well-being. Parental report was used for the QoL measure in this study. Interrater reliability between parental and child report of QoL was previously described for this sample and shown to be very good for the physical domain (ICC = 0.72, $P < 0.0001$), but less for the psychosocial aspect of QoL (ICC = 0.54, $P < 0.0001$) [24]. Unfortunately, 21 children were either too young or had cognitive and/or language deficits that limited their ability to self-report. Whereas the CAPE can be completed with the help of an interviewer and uses pictorial and verbal depiction of the items, the self-report version of the PedsQL requires more ability to be completed. Therefore, we chose to use the PedsQL parent-report version in the analysis. This is a frequently reported challenge in measurement of subjective constructs in children with complex disabilities. Children may place a higher value than their parents on engagement in leisure activities, and an association between the child's self-report leisure participation and their QoL may be stronger than findings using parental report. The fact that only children who could self-report were considered in this study clearly indicates a need to explore other measurement alternatives for children with more severe impairments in order to promote QoL for children across all severity levels. Due to the cross-sectional design of this study, a causal relationship between leisure participation and QoL cannot be inferred. For instance, children who participate in more leisure activities may experience better QoL than children

who participate in fewer or less frequent activities. The opposite may also be true, such that children who experience a higher sense of physical and psychosocial well-being may engage in more leisure activities.

6. Conclusion

Our findings indicate that leisure participation in active-physical activities is positively associated with both physical and psychosocial well-being in school-aged children with CP. Meaningful and adapted leisure activities appropriate to the child's skills and preferences may foster QoL. Further studies are needed to explore the temporal and possible causal connections between these aspects of children's lives. QoL and participation are important complex, multidimensional conceptual constructs that encompass many subjective aspects, including individual preferences, the influence of environmental factors, but also objective aspects such as the availability, accessibility, and affordability of preferred activities, the school setting, and the family's involvement in specific activities. Assessment of both constructs and interventions aiming to improve the QoL of children with CP should include the promotion of meaningful and adapted leisure activities appropriate to the child's skills and preferences, especially active-physical activities.

Acknowledgments

The authors would like to thank the children and their families for participating in this research study. Appreciation is given to Nick Hall and Anna Radzioch for research assistance in the project and to all the occupational and physical therapists who evaluated the participants. This study was funded by the United Cerebral Palsy Research & Educational Foundation, USA. This work was supported by infrastructure from the Montreal Children's Hospital-Research Institute and Centre for Interdisciplinary Research in Rehabilitation of Greater Montreal, both of which are funded by the Fonds de la Recherche en Santé du Québec. The authors would like to acknowledge the following funding agencies for doctoral support: the Canadian Child Health Clinician Scientist Program, NeuroDevNet, and the Montreal Hospital Research Institute-Foundation of Stars for Keiko Shikako-Thomas, and the Fonds de la Recherche en Santé du Québec and Montreal Children's Hospital Research Institute-Foundation of Stars for Noémi Dahan-Oliel.

References

[1] P. Rosenbaum, N. Paneth, A. Leviton, M. Goldstein, and M. Bax, "A report: the definition and classification of cerebral palsy April 2006," *Developmental Medicine and Child Neurology*, vol. 49, no. 2, pp. 8–14, 2007.

[2] F. Stanley, E. Blair, and E. Alberman, *Cerebral Palsies: Epidemiology and Causal Pathways*, Mac Keith Press, London, UK, 2000.

[3] C. Missiuna, C. Smits, P. Rosenbaum, J. Woodside, and M. Law, *Report To the Ministry of Health on the Incidence and Prevalence of Childhood Disability*, Hamilton, Canada, 2001.

[4] A. Majnemer and B. Mazer, "New directions in the outcome evaluation of children with cerebral palsy," *Seminars in Pediatric Neurology*, vol. 11, no. 1, pp. 11–17, 2004.

[5] M. I. Shevell and J. B. Bodensteiner, "Cerebral palsy: defining the problem," *Seminars in Pediatric Neurology*, vol. 11, no. 1, pp. 2–4, 2004.

[6] W. L. Minear, "A classification of cerebral palsy," *Pediatrics*, vol. 18, no. 5, pp. 841–852, 1956.

[7] M. I. Shevell, L. Dagenais, and N. Hall, "The relationship of cerebral palsy subtype and functional motor impairment: a population-based study," *Developmental Medicine and Child Neurology*, vol. 51, no. 11, pp. 872–877, 2009.

[8] L. Adamson, "Self-image, adolescence, and disability," *American Journal of Occupational Therapy*, vol. 57, no. 5, pp. 578–581, 2003.

[9] J. Specht, G. King, E. Brown, and C. Foris, "The importance of leisure in the lives of persons with congenital physical disabilities," *American Journal of Occupational Therapy*, vol. 56, no. 4, pp. 436–445, 2002.

[10] World Health Organization, *WHOQOL User Manual*, WHO, Geneva, Switzerland, 1998.

[11] R. Voll, "Aspects of the quality of life of chronically ill and handicapped children and adolescents in outpatient and inpatient rehabilitation," *International Journal of Rehabilitation Research*, vol. 24, no. 1, pp. 43–49, 2001.

[12] I. B. Wilson and P. D. Cleary, "Linking clinical variables with health-related quality of life: a conceptual model of patient outcomes," *Journal of the American Medical Association*, vol. 273, no. 1, pp. 59–65, 1995.

[13] H. Dickinson, K. Parkinson, V. McManus et al., "Assessment of data quality in a multi-centre cross-sectional study of participation and quality of life of children with cerebral palsy," *BMC Public Health*, vol. 6, article no. 273, 2006.

[14] P. L. Rosenbaum, M. H. Livingston, R. J. Palisano, B. E. Galuppi, and D. J. Russell, "Quality of life and health-related quality of life of adolescents with cerebral palsy," *Developmental Medicine and Child Neurology*, vol. 49, no. 7, pp. 516–521, 2007.

[15] World Health Organization, *International Classification of Functioning. Disability and Health—Child and Youth Version*, Geneva, Switzerland, 2007.

[16] G. King, M. Law, S. King, P. Rosenbaum, M. K. Kertoy, and N. L. Young, "A conceptual model of the factors affecting the recreation and leisure participation of children with disabilities," *Physical and Occupational Therapy in Pediatrics*, vol. 23, no. 1, pp. 63–90, 2003.

[17] M. Law, G. King, S. King et al., "Patterns of participation in recreational and leisure activities among children with complex physical disabilities," *Developmental Medicine and Child Neurology*, vol. 48, no. 5, pp. 337–342, 2006.

[18] G. King, M. A. Tucker, P. Baldwin, K. Lowry, J. LaPorta, and L. Martens, "A life needs model of pediatric service delivery: services to support community participation and quality of life for children and youth with disabilities," *Physical and Occupational Therapy in Pediatrics*, vol. 22, no. 2, pp. 53–77, 2002.

[19] N. Dahan-Oliel, K. Shikako-Thomas, and A. Majnemer, "Quality of life and leisure participation in children with neurodevelopmental disabilities: a thematic analysis of the literature," *Quality of Life Research*, vol. 21, no. 3, pp. 427–439, 2012.

[20] A. Colver, "Quality of life and participation," *Developmental Medicine and Child Neurology*, vol. 51, no. 8, pp. 656–659, 2009.

[21] S. Paisley, A. Booth, and S. Mensinkai, "Chapter 12: Health-related quality of life studies. Etext on Health Technology Assessment (HTA) Information Resources," 2005, http://www.nlm.nih.gov/archive/20060905/nichsr/ehta/chapter12.html.

[22] A. Majnemer, M. Shevell, P. Rosenbaum, M. Law, and C. Poulin, "Determinants of life quality in school-age children with cerebral palsy," *The Journal of Pediatrics*, vol. 151, no. 5, pp. 470–e1, 2007.

[23] A. Majnemer, M. Shevell, M. Law et al., "Participation and enjoyment of leisure activities in school-aged children with cerebral palsy," *Developmental Medicine and Child Neurology*, vol. 50, no. 10, pp. 751–758, 2008.

[24] A. Majnemer, M. Shevell, M. Law, C. Poulin, and P. Rosenbaum, "Reliability in the ratings of quality of life between parents and their children of school age with cerebral palsy," *Quality of Life Research*, vol. 17, no. 9, pp. 1163–1171, 2008.

[25] A. Majnemer, K. Shikako-Thomas, N. Chokron et al., "Leisure activity preferences for 6- to 12-year-old children with cerebral palsy," *Developmental Medicine and Child Neurology*, vol. 52, no. 2, pp. 167–173, 2010.

[26] R. Palisano, P. Rosenbaum, S. Walter, D. Russell, E. Wood, and B. Galuppi, "Development and reliability of a system to classify gross motor function in children with cerebral palsy," *Developmental Medicine and Child Neurology*, vol. 39, no. 4, pp. 214–223, 1997.

[27] D. J. Russell, P. L. Rosenbaum, L. M. Avery, and M. Lane, *Gross Motor Function Measure (GMFM-66 and GMFM-88)*, Mac Keith Press, London, UK, 2002.

[28] J. W. Varni, M. Seid, and P. S. Kurtin, "PedsQL 4.0: reliability and validity of the pediatric quality of life inventory version 4.0 generic core scales in healthy and patient populations," *Medical Care*, vol. 39, no. 8, pp. 800–812, 2001.

[29] G. King, M. Law, S. King et al., *Children's Assessment of Participation and Enjoyment (CAPE) and Preferences for Activities of Children (PAC)*, Harcourt Assessment, San Antonio, Tex, USA, 2004.

[30] C. Imms, "Review of the children's assessment of participation and enjoyment and the preferences for activity of children," *Physical and Occupational Therapy in Pediatrics*, vol. 28, no. 4, pp. 389–404, 2008.

[31] A. Field, *Discovering Statistics Using SPSS*, Sage, London, UK, 2009.

[32] V. Mc Manus, P. Corcoran, and I. J. Perry, "Participation in everyday activities and quality of life in pre-teenage children living with cerebral palsy in South West Ireland," *BMC Pediatrics*, vol. 8, article 50, 2008.

[33] M. Pirpiris, P. E. Gates, J. J. McCarthy et al., "Function and well-being in ambulatory children with cerebral palsy," *Journal of Pediatric Orthopaedics*, vol. 26, no. 1, pp. 119–124, 2006.

[34] J. Darrah, J. Wessel, P. Nearingburg, and M. O'Connor, "Evaluation of a community fitness program for adolescents with cerebral palsy," *Pediatric Physical Therapy*, vol. 11, no. 1, pp. 18–23, 1999.

[35] O. Verschuren, M. Ketelaar, J. W. Gorter, P. J. M. Helders, C. S. P. M. Uiterwaal, and T. Takken, "Exercise training program in children and adolescents with cerebral palsy: a randomized controlled trial," *Archives of Pediatrics and Adolescent Medicine*, vol. 161, no. 11, pp. 1075–1081, 2007.

[36] K. F. Bjornson, B. Belza, D. Kartin, R. Logsdon, J. McLaughlin, and E. A. Thompson, "The relationship of physical activity to health status and quality of life in cerebral palsy," *Pediatric Physical Therapy*, vol. 20, no. 3, pp. 247–253, 2008.

[37] E. Davis, B. Davies, R. Wolfe et al., "A randomized controlled trial of the impact of therapeutic horse riding on the quality of life, health, and function of children with cerebral palsy," *Developmental Medicine and Child Neurology*, vol. 51, no. 2, pp. 111–119, 2009.

[38] E. Davis, A. Shelley, E. Waters et al., "Quality of life of adolescents with cerebral palsy: perspectives of adolescents and parents," *Developmental Medicine and Child Neurology*, vol. 51, no. 3, pp. 193–199, 2009.

[39] D. G. Groff, N. R. Lundberg, and R. B. Zabriskie, "Influence of adapted sport on quality of life: perceptions of athletes with cerebral palsy," *Disability and Rehabilitation*, vol. 31, no. 4, pp. 318–326, 2009.

[40] H. McBurney, N. F. Taylor, K. J. Dodd, and H. K. Graham, "A qualitative analysis of the benefits of strength training for young people with cerebral palsy," *Developmental Medicine and Child Neurology*, vol. 45, no. 10, pp. 658–663, 2003.

[41] K. Shikako-Thomas, A. Majnemer, M. Law, and L. Lach, "Determinants of participation in leisure activities in children and youth with cerebral palsy: systematic review," *Physical and Occupational Therapy in Pediatrics*, vol. 28, no. 2, pp. 155–169, 2008.

[42] B. Young, H. Rice, M. Dixon-Woods, A. F. Colver, and K. N. Parkinson, "A qualitative study of the health-related quality of life of disabled children," *Developmental Medicine and Child Neurology*, vol. 49, no. 9, pp. 660–665, 2007.

[43] L. M. Buffart, R. J. G. Van Den Berg-Emons, J. V. Meeteren, H. J. Stam, and M. E. Roebroeck, "Lifestyle, participation, and health-related quality of life in adolescents and young adults with myelomeningocele," *Developmental Medicine and Child Neurology*, vol. 51, no. 11, pp. 886–894, 2009.

Hindawi Publishing Corporation
International Journal of Pediatrics
Volume 2012, Article ID 329284, 6 pages
doi:10.1155/2012/329284

Research Article

Accelerometry: A Feasible Method to Quantify Physical Activity in Ambulatory and Nonambulatory Adolescents with Cerebral Palsy

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Received 29 December 2011; Accepted 23 April 2012

Academic Editor: Roslyn Boyd

Objective. To determine the feasibility of physical activity monitoring in adolescents with cerebral palsy (CP). *Methods*. A convenience sample of ambulatory and non-ambulatory adolescents ($N = 23$; 17 males, 6 females; mean age 13.5 y, SD 2.6 y; Gross Motor Function Classification System (GMFCS) distribution: $n = 9$ Level I, $n = 5$ Level II, $n = 5$ Level III, $n = 4$ Level IV) was recruited. Physical activity (PA) was objectively assessed using the *ActiGraph* GT1M activity monitor. Discomfort or adverse effects of wearing the accelerometers were recorded by participants. Levels of physical activity were determined as total PA, light PA (LPA), moderate PA (MPA), moderate-to-vigorous (MVPA), and vigorous PA (VPA) using cut-points recently validated for CP. *Results*. Most participants showed little reluctance. Mean daily MVPA for all participants was 30.7 minutes (SD 30.3), which corresponded to 2.7 (SD 2.4) minutes of MVPA per hour or 4.5% (SD 3.9) of the total monitoring time. Total PA and MVPA were greatest in ambulatory youth (GMFCS levels I and II) compared with youth who use a walking aid or wheelchair (GMFCS levels III and IV) ($P < 0.05$). *Conclusion(s)*. The results support the use of the accelerometer as a feasible and useful measure of activity in ambulatory and nonambulatory adolescents with CP.

1. Introduction

Impaired motor function is the hallmark of cerebral palsy (CP). As a result, children and adolescents with CP are at particular risk for inactivity and the associated negative health impacts (e.g., obesity with exacerbated cardiovascular dysfunction) [1–3]. Not surprisingly, physical activity for children and adolescents with CP is commonly prescribed in clinical practice [4]. However, recent intervention studies have not been able to show a sustained increased activity level in the home, school, or community settings [5, 6].

We are currently designing the Stay-FIT intervention study to implement and evaluate a community-delivered physical activity programme for youth with CP across the severity spectrum [7]. This intervention study is part of a translational research programme focusing on physical activity in individuals with CP (http://www.canchild.ca/en/ourresearch/stay_fit.asp). The Stay-FIT programme will examine issues of physical activity covering several domains described by the World Health Organization's International Classification of Functioning (ICF) [8], ranging from cardiovascular function and structure [9] to performance in physical activity. This programme reflects the recent paradigm shift in therapy for youth with CP by using physical activity as both the focus of intervention and the primary outcome.

In order to design these future interventions, an acceptable outcome measure for physical activity is needed. Accelerometer-based activity monitoring provides an excellent measure of daily physical activity as it can be used to measure the amount, intensity, and pattern of both activity

and sedentary behavior [10–13]. In 2010, Capio et al. published a validation study on the use of a uniaxial accelerometer (MTI) to monitor activity in 31 ambulatory children with CP, Gross Motor Function Classification System (GMFCS) levels I-III [11]. Although this study did not examine the feasibility of an accelerometer systematically, the authors reported that the participants did not manifest any indications of intolerance with the device. The accelerometer was able to capture raw activity volume in unstructured free play and in six structured activities of increasing intensities including sitting, walking, and jogging. The validity of this device as a measure of PA volume was confirmed by its linear association with measured heart rate and observed PA. GMFCS levels, however, explained 0 and less than 1% of the variance in activity during structured and free play activities, respectively. It must be noted that the participants' accelerometer data demonstrated a large degree of variance, as shown by the high standard deviations. Therefore, the authors deemed it inappropriate to use regression equations to predict an activity cut-point for MVPA. In 2011, a study using the *ActiGraph* 7164 accelerometer was published by Clanchy et al. including mainly ambulatory children and adolescents (mean age 12.6 ± 2.0 years) classified at GMFCS level I or II with only a small number of GMFCS III subjects [12]. This study showed that the *ActiGraph* 7164 is able to differentiate between different intensities of walking in children and adolescents with CP. The validity of the *ActiGraph* accelerometer as a measure of PA was confirmed by using directly measured oxygen uptake as a criterion measure. Unfortunately the data did not provide sufficient power to perform meaningful subgroup analyses by GMFCS level. Van den Berg-Emons et al. reported in 2011 on the physical activity measurement in adults with various diagnoses [10]. In this study a small number of nonambulatory adults with CP were included ($n = 4$) with limited wear time of 2 consecutive days (48 hours). The authors were able to show that activity volume and intensity could be measured in nonambulatory adults with CP during various activities including wheelchair driving. To our knowledge, there have been no studies to date that have evaluated the psychometric properties of accelerometer-based assessments of habitual physical activity in non-ambulant adolescents with CP (GMFCS level IV and V) [13].

Despite the growing interest in physical activity and the use of accelerometers in the CP population, there remains a gap in our knowledge on assessment of habitual physical activity at home, at school, and in the community in adolescents with CP, particularly for those with more severe functional limitations [11–13]. In this paper we present the results of the Stay-FIT *pilot* study that was developed to test the feasibility of accelerometry for use in ambulatory and nonambulatory adolescents with CP.

2. Materials and Methods

2.1. Participants. Adolescents were recruited through regional spasticity and teenager-transition clinics of a university medical center. Participants met the following inclusion criteria: (1) age between 10 and 20 years; (2) a definite diagnosis of CP; (3) a GMFCS level I, II, III, or IV. Between October 2009 and January 2011, 31 children and adolescents with CP were identified and agreed to be contacted with respect to study participation. Of these 31, four candidates opted not to participate, three candidates agreed to participate but later withdrew from the study, and one participant was diagnosed with an acquired brain injury and was subsequently excluded. As a result, 23 children and adolescents (17 males, 6 females; mean age 13.5 y, SD 2.6 y) completed the study, of which nine were classified at GMFCS level I, five level II, five level III, and four level IV. For classification of severity we used the GMFCS Expanded and Revised version (GMFCS-ER) that has excellent interrater reliability for use in adolescents (12–18 years) [14]. All parents/guardians and participants provided written consent/assent to participate in this study approved by the Faculty of Health Sciences/Hamilton Health Sciences Research Ethics Board, Hamilton, Canada.

2.2. Assessment of Physical Activity. Habitual physical activity was objectively assessed using the *ActiGraph* GT1M activity monitor. This device was chosen for the purposes of this study for its ability to measure activity over a relatively prolonged period while remaining unobtrusive. The *ActiGraph* GT1M accelerometer weighs 27 g with dimensions of 3.8 × 3.7 × 1.8 cm (i.e., about the size of a matchbox) and measures and records acceleration in the vertical plane ranging from ~0.05 to 2 G in magnitude. The acceleration is sampled and digitized by a 12-bit analog-to-digital converter at a rate of 30 Hz. This signal is passed through a digital filter that eliminates nonhuman motion and then stored in user-defined intervals (i.e., epochs). Given the brief, intermittent, and spontaneous nature of activity reported in youth, physical activity was recorded in 3-second recording intervals or epochs [15].

2.3. Procedure. Participants were instructed to wear the accelerometer over the right hip during all waking hours for seven consecutive days, except when engaging in water activities so as not to damage the equipment. A seven-day period was selected to ensure that measured activity was representative of habitual levels of physical activity [16]. A log book was kept with the intent of tracking all times (and reasons) the device was removed and replaced. Figure 1 shows an example of a logbook and accelerometer output for a typical monitoring day for one participant. Upon completion of the seven days, log books and accelerometers were obtained from the participants and downloaded for further analysis. The research coordinator discussed any difficulties encountered and concerns that may have arisen with the participant and their parents/guardian.

2.4. Physical Activity Analysis. Accelerometer data were visually inspected to ensure that information recorded in the log book corresponded with the accelerometer output. Any activity recorded during periods of nonwear time, as indicated by the participant in the log book, was deleted. Observations of consecutive epochs of "0" counts were

Event	Monday Day 2 Date: Feb 8
Time the device was put on	7:23
Times the device may have been taken off and put back on and reason(s) (e.g. nap, swimming, shower, etc)	
	Shower 7:05
	7:50 Put back on
Time the device was taken off before bed	8:23

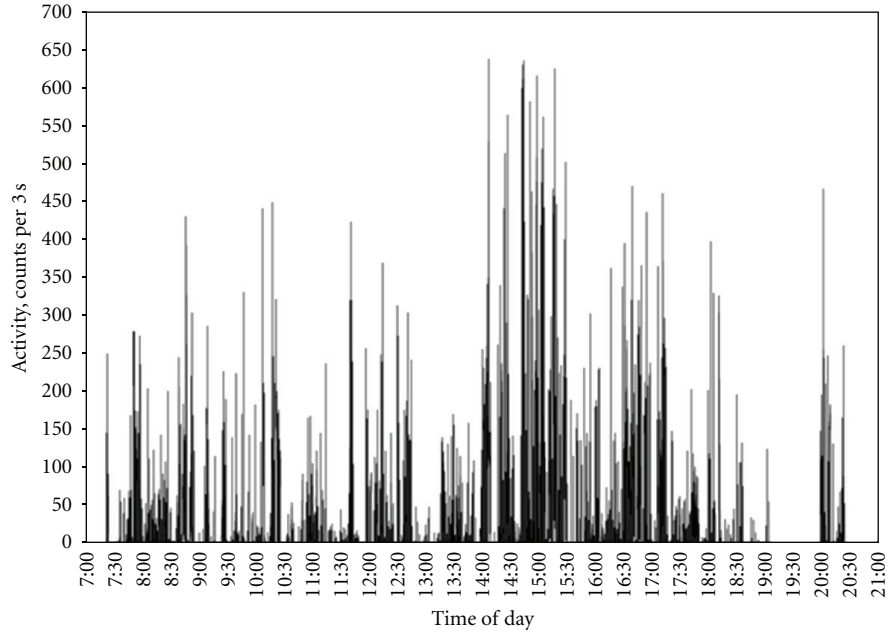

Figure 1: An example of a typical monitoring day for one participant with CP (female, 11 years of age, GMFCS level I). The table is a sample from the participant's log book in which they were asked to record the times the device was put on and taken off for each of the monitoring days. The figure to the right represents the accelerometer output for the corresponding day. No activity was recorded by the device from 7:05 to approximately 7:56 pm, which corresponds to the time the participants indicated they removed the device.

considered sedentary time unless otherwise stated in the log book. Only participants with ≥5 hours of monitoring time on ≥4 days were included in the analyses. These criteria were selected to maximize participant inclusion and were based on the minimal allowable time previously used to estimate habitual physical activity [16, 17]. The data were then uploaded to a Microsoft Excel-based Visual Basic data reduction program to determine total monitoring time and total activity. The program also distinguished light physical activity (LPA), moderate physical activity (MPA), moderate-to-vigorous activity (MVPA), and vigorous physical activity (VPA). Activity intensity was examined using the cut-points developed by Clanchy et al. and Evenson et al., which were recently validated for use in children and adolescents with CP [12, 18].

2.5. Descriptive Statistics. Levels of physical activity (total, LPA, MPA, VPA, and MVPA) between the four GMFCS levels were compared using one-way analysis of variance (ANOVA) in STATISTICA (StatSoft, Tulsa, Okla., USA). Analyses of covariance (ANCOVA) were also performed with chronological age as the covariate so as to account for the distribution in age among participants. Tukey's honestly significant post hoc tests were performed when necessary. Given the small number of participants in each of the GMFCS levels, Kendall's Tau was used to assess the relationship between MVPA and GMFCS level in SPSS (Version 17.0, Chicago, Ill., USA). Descriptive statistics were used to calculate the proportion of participants meeting the Canadian physical activity guideline recommendations for youth (≥60 min MVPA per day). Statistical significance for all analyses was set at $P \leq 0.05$.

3. Results

3.1. Feasibility. We found that refusal to participate was most often based upon the lack of parental enthusiasm and the youth's perception that they might look "different" in various social settings. No participants reported discomfort or adverse effects of wearing the accelerometer throughout the duration of the study. One participant exposed the device to water on day 5 of wear, which resulted in highly erratic data. Therefore, that participant's data from only days 1 to 4 were included in the analyses.

3.2. Physical Activity Levels. None of the participants were excluded on the basis of failure to wear the accelerometer for the minimum required period (≥5 hours on ≥4 days). On average, the device was worn for 6 of the 7 required monitoring days, with the monitoring period ranging from 540.5 to 859.2 minutes per day (mean ± SD: 707.7 ± 81.2 min). On a daily basis, our participants engaged in (mean ± SD) 89.5 ± 47.1 min of LPA, 17.8 ± 16.9 min of MPA, 12.0 ± 14.4 min of VPA, and 30.7 ± 30.3 min of MVPA. To account for differences in wear time, data were also examined as minutes of activity per hour of monitoring time (Table 1). Both ANOVA and ANCOVA suggested that youth classified at GMFCS level IV presented with lower levels of LPA, MPA, and MVPA compared with level I ($P < 0.05$). Similarly, youth at level III demonstrated lower levels

Table 1: Minutes of activity per hour and per day of monitoring time.

		GMFCS				
		Level I ($n = 9$)	Level II ($n = 5$)	Level III ($n = 5$)	Level IV ($n = 4$)	Total ($n = 23$)
LPA	min/day	121.5 (38.7)	95.7 (43.7)	66.7 (33.5)	32.1 (18.2)*	88.4 (47.8)
	min/hr	10.2 (3.4)	8.2 (3.6)	5.6 (2.3)	1.6 (1.2)*†	7.3 (4.2)
MPA	min/day	33.0 (15.6)	16.4 (12.5)	6.0 (5.7)*	2.7 (3.7)*	18.2 (17.1)
	min/hr	2.7 (1.2)	1.4 (1.0)	0.5 (0.4)*	0.2 (0.3)*	1.5 (1.4)
MVPA	min/day	56.0 (28.4)	26.6 (24.7)	9.3 (10.4)*	5.8 (8.0)*	30.7 (30.3)
	min/hr	4.5 (2.1)	2.2 (2.1)	1.5 (1.5)*	0.5 (0.7)*	2.6 (2.4)

Data are presented as mean (SD). *indicates significant difference from level I, †indicates significant difference from level II, $P < 0.05$.
Minutes of activity per hour and per day of monitoring time.
LPA: light physical activity; MPA: moderate physical activity; MVPA: moderate-to-vigorous physical activity; CP: cerebral palsy; GMFCS: Gross Motor Function Classification System; min: minutes; hr: hour.

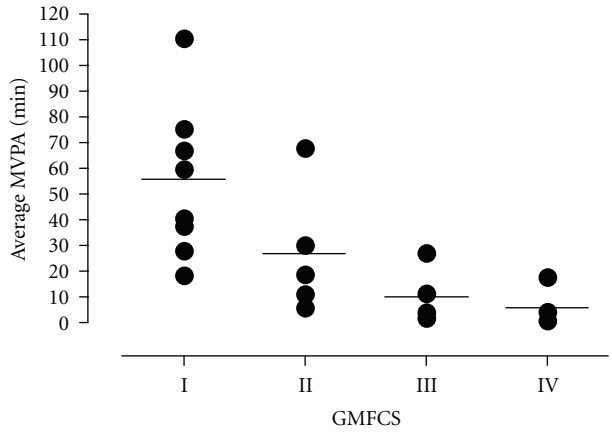

Figure 2: Moderate-to-vigorous activity time in adolescents with CP by GMFCS level. MVPA: moderate-to-vigorous physical activity; CP: cerebral palsy; GMFCS: Gross Motor Function Classification System; min: minutes.

of MPA and MVPA compared with level I. No differences were seen between levels I and II for any intensity, nor were there significant differences between levels II, III, and IV. A significant negative correlation was seen between MVPA, in both minutes/day and minutes/hour, and GMFCS levels (minutes/day: $\tau = -0.65$, minutes/hour: $\tau = -0.61$, $P < 0.001$, Figure 2).

4. Discussion

This study aimed to assess the ability of accelerometers to measure the duration, intensity, and timing of physical activity in the home, community, and at school of ambulatory and nonambulatory adolescents with CP. The use of the accelerometer worn around the waist to measure habitual physical activity has been shown to be feasible and unobtrusive to participants, including adolescents classified as GMFCS level IV. Our results are in line with recently published data on the *ActiGraph* in children and teenagers up to age 16 years with CP, GMFCS level I and II [11, 12] and add supportive data for its use in adolescents with more functional limitations (GMFCS level III and IV) and older adolescents (up to age 20 years).

While our findings are promising, a number of limitations both inherent to the accelerometer and related to our analysis should be noted. First, our small sample size may explain the lack of significant findings when comparing activity by GMFCS level. Moreover, the activity cut-points selected were based on the work by Clanchy et al. [12], which was performed only in ambulatory youth with CP. It is unknown whether these same cut-points are applicable in nonambulatory adolescents with CP. Our assessment of activity is further complicated by the use of waist-worn accelerometer in nonambulatory adolescents. More specifically, it is impossible to determine whether the use of these waist-worn accelerometers accurately captured all activity performed by participants classified as GMFCS levels III and IV, particularly in those youth who are able to self-propel their wheelchair. Future work should employ a similar design to that utilized by Clanchy et al. [12] in which the oxygen cost of movement is assessed in conjunction with accelerometer recordings. Finally, it is important to note that the sensitivity of the accelerometer to water presents a particular challenge since aquatic exercise seems to be a preferred activity of youth with CP [19]. Despite this limitation, we believe the ability of accelerometers to measure the duration, intensity, and timing of physical activity in the home, community, and at school outweighs the limitation of missing relatively small amounts of time in a pool.

With respect to activity monitoring, of the 23 participants, none engaged in ≥60 min MVPA per day recommended in the Canadian Physical Activity Guidelines for youth on a daily basis, 17 (74%) participants did not achieve 60 min MVPA on any monitoring day, and the remaining 6 participants engaged in ≥60 min MVPA on 3–5 days out of 7. Even in participants at the most functional ability level (GMFCS level I) we observed a highly sedentary lifestyle. The minimal amount of active time found in this pilot study highlights the dire need for intervention studies.

We believe the *ActiGraph* accelerometer is ready to use as a measure of habitual physical activity at home, at school, and in the community in children and adolescents with CP. The recent validation of cut-points for classification of

activity levels in this population has provided a great opportunity to use accelerometers as an outcome measure in highly needed intervention studies that emphasize the integration of physical activity and participation into daily lifestyle.

5. Conclusion

This study shows promise for using the *Actigraph* GT1M accelerometer as a feasible and meaningful measure in daily activity in adolescents with CP across GMFCS levels I–IV.

Abbreviations

ANOVA:	One-way analysis of variance
ANCOVA:	Analyses of covariance
CP:	Cerebral palsy
GMFCS:	Gross Motor Function Classification System
GMFCS-E&R:	Gross Motor Function Classification System, expanded and revised version
LPA:	Light physical activity
MPA:	Moderate physical activity
MVPA:	Moderate-to-vigorous physical activity
PA:	Physical activity
VPA:	Vigorous physical activity.

Acknowledgments

This pilot-project is part of the Stay-FIT study and is funded by the Ontario Federation for Cerebral Palsy. The following members of the Stay-FIT study Group participated in the research program: Jan Willem Gorter, Maureen MacDonald, Robert Palisano, Peter Rosenbaum, Brian Timmons, and Virginia Wright. The authors thank Barb Galuppi, Research Coordinator of the Stay-FIT study, for her contributions to the project. Special thanks are due to the participating adolescents with CP and to their parents as well. A preliminary report of the findings of this study was presented by Stephen Noorduyn at the 2nd Joint Meeting of the North American Society for Pediatric Exercise Medicine (NASPEM) and the European Group of Pediatric Work Physiology in Niagara-on-the-Lake, ON, Canada in September 2010 and by Jan Willem Gorter at the annual meeting of the Ontario Association of Children's Services (OACRS) in Toronto on 7 November 2011.

References

[1] C. A. Maher, M. T. Williams, T. Olds, and A. E. Lane, "Physical and sedentary activity in adolescents with cerebral palsy," *Developmental Medicine and Child Neurology*, vol. 49, no. 6, pp. 450–457, 2007.

[2] K. F. Bjornson, B. Belza, D. Kartin, R. Logsdon, J. McLaughlin, and E. A. Thompson, "The relationship of physical activity to health status and quality of life in cerebral palsy," *Pediatric Physical Therapy*, vol. 20, no. 3, pp. 247–253, 2008.

[3] N. Owen, P. B. Sparling, G. N. Healy, D. W. Dunstan, and C. E. Matthews, "Sedentary behavior: emerging evidence for a new health risk," *Mayo Clinic Proceedings*, vol. 85, no. 12, pp. 1138–1141, 2010.

[4] E. G. Fowler, T. H. A. Kolobe, D. L. Damiano et al., "Promotion of physical fitness and prevention of secondary conditions for children with cerebral palsy: section on pediatrics research summit proceedings," *Physical Therapy*, vol. 87, no. 11, pp. 1495–1510, 2007.

[5] O. Verschuren, M. Ketelaar, T. Takken, P. J. M. Helders, and J. W. Gorter, "Exercise programs for children with cerebral palsy: a systematic review of the literature," *American Journal of Physical Medicine and Rehabilitation*, vol. 87, no. 5, pp. 404–417, 2008.

[6] C. A. Maher, M. T. Williams, T. Olds, and A. E. Lane, "An internet-based physical activity intervention for adolescents with cerebral palsy: a randomized controlled trial," *Developmental Medicine and Child Neurology*, vol. 52, no. 5, pp. 448–455, 2010.

[7] A. A. O. M. Claassen, J. W. Gorter, D. Stewart, O. Verschuren, B. E. Galuppi, and L. J. Shimmell, "Becoming and staying physically active in adolescents with cerebral palsy: protocol of a qualitative study of facilitators and barriers to physical activity," *BMC Pediatrics*, vol. 11, article no. 1, 2011.

[8] World Health Organization, *The International Classification of Functioning, Disability, and Health*, World Health Organization, Geneva, Switzerland, 2001.

[9] A. A. Martin, L. M. Cotie, B. W. Timmons, J. W. Gorter, and M. J. MacDonald, "Arterial structure and function in ambulatory adolescents with cerebral palsy are not different from healthy controls," *International Journal of Pediatrics*, vol. 2012, Article ID 168209, 8 pages, 2012.

[10] R. J. Van Den Berg-Emons, A. A. L'Ortye, L. M. Buffart et al., "Validation of the physical activity scale for individuals with physical disabilities," *Archives of Physical Medicine and Rehabilitation*, vol. 92, no. 6, pp. 923–928, 2011.

[11] C. M. Capio, C. H. Sit, and B. Abernethy, "Physical activity measurement using MTI (actigraph) among children with cerebral palsy," *Archives of Physical Medicine and Rehabilitation*, vol. 91, no. 8, pp. 1283–1290, 2010.

[12] K. M. Clanchy, S. M. Tweedy, R. N. Boyd, and S. G. Trost, "Validity of accelerometry in ambulatory children and adolescents with cerebral palsy," *European Journal of Applied Physiology*, vol. 111, no. 12, pp. 2951–2959, 2011.

[13] K. M. Clanchy, S. M. Tweedy, and R. Boyd, "Measurement of habitual physical activity performance in adolescents with cerebral palsy: a systematic review," *Developmental Medicine and Child Neurology*, vol. 53, no. 6, pp. 499–505, 2011.

[14] J. W. Gorter, J. Slaman, D. Bartlett, and H. J. G. Van den Berg-Emons, "Reliability of the gross motor function classification system expanded and revised (GMFCS-ER) when used with adolescents and young adults with cerebral palsy," *Developmental Medicine & Child Neurology*, vol. 53, supplement 5, pp. 42–43, 2011.

[15] G. Baquet, S. Berthoin, and E. Van Praagh, "Are intensified physical education sessions able to elicit heart rate at a sufficient level to promote aerobic fitness in adolescents?" *Research Quarterly for Exercise and Sport*, vol. 73, no. 3, pp. 282–288, 2002.

[16] S. G. Trost, R. R. Pate, P. S. Freedson, J. F. Sallis, and W. C. Taylor, "Using objective physical activity measures with youth: how many days of monitoring are needed?" *Medicine and Science in Sports and Exercise*, vol. 32, no. 2, pp. 426–431, 2000.

[17] K. A. Pfeiffer, M. Dowda, K. L. McIver, and R. R. Pate, "Factors related to objectively measured physical activity in preschool children," *Pediatric Exercise Science*, vol. 21, no. 2, pp. 196–208, 2009.

[18] K. R. Evenson, D. J. Catellier, K. Gill, K. S. Ondrak, and R. G. McMurray, "Calibration of two objective measures of physical activity for children," *Journal of Sports Sciences*, vol. 26, no. 14, pp. 1557–1565, 2008.

[19] J. W. Gorter and S. J. Currie, "Aquatic exercise programs for children and adolescents with Cerebral Palsy: what do we know and where do we go?" *International Journal of Pediatrics*, vol. 2011, Article ID 712165, 7 pages, 2011.

Hindawi Publishing Corporation
International Journal of Pediatrics
Volume 2012, Article ID 541363, 11 pages
doi:10.1155/2012/541363

Review Article

A Review of the Potential for Cardiometabolic Dysfunction in Youth with Spina Bifida and the Role for Physical Activity and Structured Exercise

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Received 8 December 2011; Revised 5 April 2012; Accepted 20 April 2012

Academic Editor: Maria A. Fragala-Pinkham

Children and adolescents who have decreased mobility due to spina bifida may be at increased risk for the components of metabolic syndrome, including abdominal obesity, insulin resistance, and dyslipidemia due to low physical activity. Like their nondisabled peers, adolescents with spina bifida that develop metabolic risk factors early in life have set the stage for adult disease. Exercise interventions can improve metabolic dysfunction in nondisabled youth, but the types of exercise programs that are most effective and the mechanisms involved are not known. This is especially true in adolescents with spina bifida, who have impaired mobility and physical function and with whom there have been few well-controlled studies. This paper highlights the current lack of knowledge about the role of physical activity and the need to develop exercise strategies targeting the reduction of cardiometabolic risk and improving quality of life in youth with spina bifida.

1. Introduction

Spina bifida (SB) is a congenital abnormality characterized by the incomplete closure of the spinal column. The majority (>90%) of cases are classified as meningomyelocele, in which the spinal cord protrudes through the spinal column, resulting in nerve damage and physical disabilities including lower limb paralysis and disrupted bladder or bowel function. Widespread public advertising and clinical advocacy campaigns have been used to promote the intake of adequate dietary folic acid during pregnancy, since this strategy has been shown to reduce the risk of developing SB. Nevertheless, the current prevalence of SB in the United States is estimated to be close to 1 in 1,000 pregnancies [1]. With better surgical treatments and early medical care infant survival rates rose from 83% in 1979–83 to 91% in 1989–94 [2]. A larger national survey covering 1995–2001 reported infant survival rates of 92% [3]. Thus, in the United States ~25,000 children ages 0–19 years and ~166,000 total people are currently affected by SB [4, 5].

As a result of improved surgical treatments and early medical care, children with SB can expect longer lives today than in the past [2]. In recent decades spinal repair and shunting techniques have improved, complications have declined, and survival rates have increased [1, 2, 6, 7]. Although life span for people with SB has increased, there remain physical limitations and mobility issues that require attention. Additionally, a new health challenge that must be considered is the potential for cardiometabolic disease risk, which may result from the physical deconditioning that occurs in people with disabilities [8]. As described by Rimmer et al. [8] in their recent review, people with physical disabilities spend less time performing physical activities than their nondisabled peers. The consequences of a sedentary lifestyle in all people include physical deconditioning and increased risk of developing obesity,

insulin resistance, and cardiovascular disease. People with disabilities such as SB or spinal cord injury (SCI), especially those confined to wheelchairs, are prone to develop what Rimmer et al. [8] termed "disability-associated low energy expenditure deconditioning syndrome," in which sedentary lifestyle creates a vicious cycle of further deconditioning, disability, and disease risk. This cyclic problem worsens with advancing age, but strategies such as exercise programs tailored to meet the specific needs of the individual can be an effective approach to attenuate or reverse the progression of functional disability and disease risk. To date, the presence of cardiometabolic disease risk in people with SB is not well described. Additionally, there have been only a few published studies that reported the results of exercise training programs for children with SB. Those studies, however, have been small and primarily focused on physical function rather than cardiometabolic health. In fact, a recent systematic review concluded that most of the publications on exercise programs for SB were not of high quality due to small sample size and poorly defined outcomes [9].

The purpose of this narrative review is to describe the current scientific literature on cardiometabolic risk in youth with SB and the potential for structured exercise intervention programs to improve health and function in that population. The volume of existing literature in this area is relatively small and therefore neither a structured systematic review of the evidence, nor a meta-analysis of the results of multiple exercise studies was feasible. Thus, our goal is to outline what is known about the potential cardiometabolic consequences of SB, to highlight both the strengths and weaknesses of selected studies, to identify gaps in knowledge, and to suggest future directions that would produce the type of evidence that will ultimately guide improved clinical care and lifestyle recommendations for people with SB. Our review of the literature covers primarily papers published with in the past 25 years that could be identified through PubMed and cross-referencing citations of published papers. We included some studies with small sample sizes because so little has been published to date, but excluded individual case studies. Although the focus of this narrative review is on SB, we have included selected results obtained from studies of SCI and childhood obesity because of their relevance to understanding the potential consequences of physical disability, deconditioning, and cardiometabolic risk in youth with SB.

2. Body Composition, Physical Function, and Cardiometabolic Risk

2.1. Health and Function in Children with SB. Although the prevalence of diabetes and cardiovascular disease in the SB population is not known, children and adults with SB may be at increased risk for metabolic and vascular dysfunction because of their body composition, physical function, and clinical blood test results (Table 1). One of the most common findings has been that body mass index (BMI) and percent body fat are increased in people with SB relative to people without SB [10–16]. Although it has been reported that overweight and obesity prevalence are increased in both children and adults with SB [10–14, 16], Dosa et al. [15] found in their study of 203 people that obesity prevalence, based on BMI, for children 6–19 years old with SB (8–18%) was similar to current general population values for children in the United States. However, they found that for adults >20 years old obesity rates in people with SB (37%) were higher than in the general population [15]. The sample size in that study was larger than most others in the SB literature, but it is unclear if the results are generalizable since, to our knowledge, there are no large-scale surveys of obesity prevalence in people with SB. It is important to note, though, that classifying overweight and obesity in people with SB according to BMI standards, as it is done in the nondisabled population, may underestimate the true extent of obesity in people with SB. Nelson et al. [12] proposed defining obesity in children affected by SB or SCI as the presence of abdominal fat measured by dual X-ray absorptiometry that exceeds 30% of tissue mass in boys or 35% in girls, respectively. Using this approach they reported that 18 out of 34 (~53%) children with SB tested in their center met the criterion for obesity based on standardized BMI z-score, but that 25 out of 34 (~74%) would be classified as obese based on abdominal fat content. Those results underscore the fact that BMI does not provide information about the composition of body mass and can often mask offsetting changes in the proportions of fat and lean tissue that occur over time within individuals, or differences that exist between comparison groups. Furthermore, BMI standards developed for nondisabled children have not been validated for children with SB. To do so would require careful control for the level of ambulation and spinal cord involvement and might prove difficult because of the wide range of physical abilities and spinal cord deficits in people with SB. Adopting a classification system for overweight or obesity based on body composition, however, is also challenging in clinical settings since there are several potential methods (e.g., skinfolds, air displacement plethysmography, underwater weighing, bioelectrical impedance, dual energy X-ray absorptiometry, and magnetic resonance imaging) that require expertise and resources that may not be available in some centers. Nevertheless, in research settings, assessing body composition, particularly abdominal fat, should be included in future assessments of metabolic risk in youth with SB.

In addition to increased body fatness, children and young adults with SB typically are reported to have reduced aerobic fitness and muscular strength [10, 11, 17]. These deficits in physical capacity are reported in both ambulatory individuals tested using leg cycling or on a treadmill, and in nonambulatory individuals tested using arm crank ergometry, respectively. The study of Widman et al. [10] was notable because there were 37 nonambulatory adolescents with SB who completed assessments of upper body muscle strength and maximal aerobic capacity testing, and the results were compared to 34 age-matched control adolescents without SB who completed the same tests in the same research center. In two other studies [11, 17], measurements of muscle strength, and aerobic capacity during leg cycling were compared

against previously published data from nondisabled people and found to be up to 30–40% lower in people with SB. These differences are large enough to support that physical function deficits are present in people with SB. However, reliability and normative values for body composition and physical function can vary among laboratories due to differences in test equipment, test operators, and regional population characteristics. Thus, research studies on SB should ideally include comparison data from age- and sex-matched controls studied in the same laboratory. Comparison values for aerobic capacity during arm crank ergometry were not provided in two of the studies cited previously [11, 17]. This is a limitation since the number of nonambulatory people with SB is substantial; for example, ~90% of SB patients in our clinical center rely on wheelchairs and are nonambulatory. Thus, further descriptive data are needed to fully characterize the upper body exercise capacity of youth with SB.

As a result of low aerobic capacity and muscle strength, activities of daily living may be relatively more difficult for people with SB. Bruinings et al. [18] reported that the oxygen cost (work economy) of walking or wheelchair use at fixed speeds for children and young adults with SB was similar to, or even less than that of age-matched people without SB. However, for ambulatory people with SB, self-selected walking speed was 14% slower compared to nondisabled controls [18]. More importantly, because of their reduced peak aerobic capacity, activities of daily living were relatively more demanding since they required a higher portion of aerobic reserve capacity for both ambulatory and nonambulatory people with SB. Thus, it is not surprising that the amount of daily physical activity performed by children and adults with SB is lower than nondisabled comparison groups [11, 19, 20]. Likewise, the reported quality of life is lower in people with SB, particularly in those who are nonambulatory [21–24]. In the study by Abresch et al. [21], quality of life was similarly reduced in children with either SB or SCI compared to healthy controls. Interestingly, though, the presence of obesity adversely affected quality of life scores in healthy control children, it had no further effect on the scores of children with SB or SCI in that study. It remains to be shown whether quality-of-life assessments are improved following physical activity interventions.

Given the increased presence of obesity and decreased physical activity and physical function, it is plausible to expect that the components of metabolic risk would be adversely affected by the presence of SB. There are, however, only a few studies that have measured the components of metabolic syndrome or associated risk factors in children or adults with SB. The available reports showed that fasting glucose, insulin, cholesterol, triglycerides, and blood pressure were, on average, not different in people with SB compared to either non-SB control groups or published normal values (Table 1) [12, 25, 26]. Some studies have reported values for clinical blood tests but did not include group means and control values, so the effect of SB was difficult to determine [27]. Exceptions to those trends were reported by Rendeli and colleagues, who found that total- and VLDL-cholesterol were increased in girls with SB, though not in boys with SB [26], and that circulating homocysteine was increased in boys and girls with SB [28]. The difference in total cholesterol outcomes between boys and girls in the former study [26] appeared to be attributable to differences in walking ability. Total- and VLDL-cholesterol values were higher in girls who did not walk compared to those who walked independently or with assistance, but this effect of walking ability was not apparent in boys. This observation may be related to the finding that girls are, in general, less likely to be physically active than boys during childhood and adolescence [29]. It should also be noted, however, that the data in the study by Rendeli et al. [26] were not corrected for potential differences in age or maturation between sexes and therefore require confirmation.

Although the group means reported in most of the studies cited above revealed little or no differences between people with and without SB, the results require confirmation in larger studies, and there is still reason for concern. For example, assessment of vascular function in a small group of men with SB demonstrated that arterial diameters were reduced and sheer stress on the vascular wall was increased compared to nondisabled controls, a result that is predictive of endothelial dysfunction [30]. Additionally, in a Dutch cohort of 31 adolescents and young adults with SB, ~90% had one or more risk factors for cardiovascular disease based on the Framingham Heart Study criteria, which includes assessments of total- and HDL-cholesterol, systolic blood pressure, and smoking [27]. Similarly, Nelson et al. [12] reported that fasting insulin resistance (measured using the calculation of homeostatic model of assessment for insulin resistance, HOMA-IR) was significantly increased in children and young adults with SB or SCI who were obese. Furthermore, 70%–75% of study participants with SB and 80–100% of participants with SCI had low HDL-cholesterol, independent of the presence of obesity. Metabolic syndrome (defined as the presence of three or more risk factors from a panel including obesity assessed by BMI, percent body fat or waist circumference; insulin resistance assessed by fasting glucose, fasting insulin, or impaired glucose tolerance; hypertriglyceridemia; low HDL-cholesterol; hypertension) was present in 32% of people with SB and 50% of people with SCI, compared to 20% of nondisabled controls [12]. The presence of those adverse clinical outcomes in the SB and SCI groups could be predominantly ascribed to the copresence of obesity, just as it was in the nondisabled control group. However, because youth with SB (and SCI) had higher rates of metabolic syndrome, which represents the clustering of adverse clinical conditions, and because the prevalence of obesity is elevated in the SB population, the results of that study [12] and the work of Buffart et al. [27] suggest that clinicians should carefully monitor young people with SB for these signs of disease risk.

2.2. Additional Insights into Health and Function from Studies of SCI. Although the etiology of their disabilities is clearly different from SB, people with limited use of their legs due to spinal cord injury (SCI) and who rely on wheelchairs for mobility face many of the same health

concerns as people with SB. In the United States, there are >1.2 million people with spinal cord injury (SCI) and ~1,500–2,000 new pediatric cases of SCI per year [31]. Because of similar physical challenges and sedentary lifestyle, individuals with SCI, like those with SB, face an increased risk of cardiometabolic disease. As with SB, children with SCI are likely to have obesity characterized by increased abdominal fat and reduced lean mass [16, 32]. In the study by Liusuwan et al. [16] the magnitude of increase in body fat and decrease in lean body mass relative to normal weight or overweight nondisabled control groups was greater in youth with SB than with SCI. This finding suggests that the long-term health consequences of obesity for SB may be more pronounced than for SCI.

Although there are relatively few studies of metabolic risk factors in adults with SB, there are relevant data in the SCI population that could be used to predict longer-term health risks in the SB population. For example, a recent analysis of 134 Swedish adults with SCI revealed that one-third of people met the criteria for cardiovascular risk intervention based on the Framingham Risk Equation and the Systemic Coronary Risk Evaluation models, which include measures of cholesterol, blood pressure smoking, age, and sex [34]. When the authors of that study included BMI > 22 kg/m^2 as a potential measure of overweight deemed appropriate for people with SCI, nearly 80% of the group reached the higher risk classification worthy of clinical intervention [34]. Additionally, as shown in children and young adults with SB, HDL-cholesterol has been reported to be reduced in people with SCI [35]. Additional markers of cardiometabolic disease risk that are reported to be increased in people with SCI include the inflammatory proteins, C-reactive protein, and interleukin-6 [35, 36], neither of which have been reported in people with SB to our knowledge. Furthermore, Nash et al. [37] made the observation in young adults with SCI that fasting triglycerides were normal, but the elevation in triglycerides was ~35% higher following a high-fat meal compared to recreationally active nondisabled men. They proposed that the meal stimulus revealed a type of dyslipidemia that is undetected when only fasting measurements are used. This situation is analogous to carbohydrate metabolism, whereby fasting glucose may be in the normal range but postprandial blood glucose is elevated, revealing impaired glucose tolerance. The finding by Nash et al. [37], however, was based on data on only three young men with SCI and therefore needs to be replicated in both the SCI and SB populations. Postprandial hyperlipidemia could be an important clinical outcome as it has been shown to be increased in nondisabled people with hypertension and cardiovascular disease [38] and may therefore represent an early marker for disease risk applicable to adolescents with SB.

2.3. Health Consequences of Obesity and/or Sedentary Lifestyle in Nondisabled Youth. The potential metabolic risk in children with SB resembles in many ways the current problem of obesity and sedentary lifestyle in all adolescents. According to body mass index criteria for obesity (≥the 95th percentile for age and sex) ~18% of children in the United States are obese [39]. While overnutrition is a key contributor to the development of childhood obesity, there is also a role for physical activity, and half of American children fail to reach the recommended 60 minutes per day of moderate-to-vigorous physical activity [29, 40]. Abdominal adiposity in children is a strong predictor of insulin resistance and is associated with features of metabolic syndrome such as dyslipidemia, hypertension, and increased circulating inflammatory markers, all of which have been more widely studied in nondisabled children than children with SB [41–46]. It is estimated that 25–50% of overweight adolescents meet the criteria for metabolic syndrome [47]. More importantly, longitudinal studies show that obesity and dyslipidemia that are present during childhood often persist into adulthood and predict the development of cardiovascular disease [48–50], which makes obesity and sedentary lifestyle in today's youth such a widespread concern. Thus, lifestyle or other clinical strategies need to be developed to address these concerns for youth in general and tailored to meet the special needs of children with SB.

3. Exercise as a Means of Lowering Metabolic Risk

3.1. Exercise Programs for People without SB. It is well established that exercise, alone or in combination with dietary strategies aimed to reduce body weight, can be effective for the prevention of diabetes and metabolic syndrome in nondisabled children and adults [51–55]. Extensive literature has also shown that exercise performed at least 2-3 days per week can improve insulin sensitivity, reduce abdominal fat, and increase HDL-cholesterol, in addition to providing numerous other health benefits in nondisabled children and adults [56–63]. Similarly, when exercise is performed up to 15 hours before a meal the circulating postprandial triglyceride concentration is reduced, a result that is attributed to activation of lipoprotein lipase by muscle contraction [64–67]. Unfortunately, in the United States, recent observational studies have reported that less than half of children and adults meet the current recommendations of 300 and 150 minutes per week, respectively, of moderate-to-vigorous physical activity [29, 40, 68]. Likewise, due to the physical limitations described a common clinical observation is that many people with SB have low physical activity. The extent of this perceived problem is not yet quantified, since descriptive data for the participation rates in physical activity, including structured exercise or sports or activities of daily living, are to our knowledge not available for the SB population.

3.2. Exercise for Children with SB. Recent work suggests that some adolescents with SB can successfully participate in sports and should be encouraged to do so for psychosocial reasons [69, 70]. Buffart et al. [22] reported that children and young adults with SB had lower reported quality of life scores than a nondisabled comparison group, and ~60% had difficulty with activities of daily living. However,

Table 1: Summary of published outcomes for body composition, physical function, and metabolic and vascular risk factors in people with spina bifida (SB).

Outcome	Increased in SB	Decreased in SB	Not different in SB
Body mass index, for age and sex	- Non-amb children [10] - Amb children [11]* - Children and young adults [12] - Amb and non-amb adults [15] - Children [16]		- Amb and non-amb children [15]
Total body fat (%)	- Non-amb children [10] - Children and young adults [12] - Children [13, 14, 16]		- Amb children [11]*
Abdominal fat (%)	- Children and young adults [12] - Obese children [25]*		
Lean body mass (%)		- Children [14, 16]	
Aerobic capacity		- Non-amb children [10] - Amb children [11]* - Non-amb children and young adults [17]* - Amb children and young adults [17]* - Amb children [33]	
Muscle strength	- Non-amb children [10] - Amb children [11]* - Non-amb children and young adults [17]* - Amb children and young adults [17]*		
Absolute energy cost of walking or wheelchair use		- Non-amb children and young adults [18]	- Amb children and young adults [18]
Requirement of physical reserve for activities of daily living	- Non-amb children and young adults [18] - Amb children and young adults [18]		
Total daily physical activity	- Amb children [11]* - Non-amb children [19] - Amb children [19] - Non-amb children and young adults [20]* - Amb children and young adults [20]*		
Glucose		- Children and young adults [12]	- Obese children [25]*
Insulin			- Children and young adults [12]
HOMA-IR	- Obese children [25]*		- Children and young adults [12]
Total cholesterol	- Amb and non-amb girls [26]		- Children and young adults [12] - Amb and non-amb girls [26]

Table 1: Continued.

Outcome	Increased in SB	Decreased in SB	Not different in SB
LDL-cholesterol			- Children and young adults [12] - Amb and non-amb girls [26]
HDL-Cholesterol			- Children and young adults [12] - Amb and non-amb girls [26]
Triglycerides			- Amb and non-amb children [26]
Homocysteine	- Amb and non-amb children [28]		
Blood pressure	- Amb and non-amb children and young adults [27]		- Obese children [25]*

All comparisons refer to outcomes for people with SB relative to people without SB. Results from people with SCI or other spinal disorders are not included in this table according to the descriptions presented in the individual studies. *Comparison is relative to published or unpublished values from prior studies; all other investigations used a specifically recruited comparison group within their study design. Amb: ambulatory; Non-amb: nonambulatory. If not specified, ambulatory status was not stated in the cited study. In some studies "amb and non-amb" designation is used because the results for ambulatory and nonambulatory participants were not presented separately. The designation for children and adults in the cited studies is defined as <18 or ≥18 years old, respectively. In some studies, results from children and adults were not presented separately.

after controlling for ambulation status and sex (walkers and males reported fewer difficulties, resp.), daily physical activity and aerobic fitness were positively correlated with quality of life and inversely correlated with physical difficulties. These results support the possibility that appropriately tailored physical activity programs could improve perceived quality of life for young people who currently have low habitual activity. However, the most feasible and effective types of exercise for people with SB are not well defined.

To date, physical therapy strategies employed with people affected by SB have focused on muscle strengthening exercises and orthopedic supports and assistive devices meant to aid ambulation and postural control [71–73]. Similarly, the few exercise intervention trials performed with people with SB-targeted improvements in muscle size, muscle function, or aerobic capacity since these outcomes are likely to translate to better mobility and performance of activities of daily living [74–77]. However, most of those studies used small sample size and required replication.

The first study to our knowledge to report the feasibility and effectiveness of exercise in SB was published by Ekblom and Lundberg in 1968 [74]. In that study, seven adolescents with SB, seven with cerebral palsy, and three with other forms of spinal paraplegia, completed a low volume (30 minutes twice per week for 6 weeks) of moderate-intensity wheelchair exercise consisting of wheelchair driving and upper body strengthening activities. This program resulted in improvements in upper body fitness in the SB group as shown by a 10-11% reduction in oxygen uptake and heart rate and a 40% reduction in blood lactate during submaximal arm crank exercise. During maximal arm crank exercise, work output was 21% higher after training although peak oxygen uptake did not change; suggesting that both submaximal and maximal work economy was increased. Since that report, however, there have been only two investigations in which nonambulatory children or adults with SB performed upper body exercise training and fitness outcomes were reported. Andrade et al. [75] had eight children with SB, five who were ambulatory, and three who wheelchair users, complete a once-weekly supervised program of upper body aerobic and resistance exercise, which resulted in improvements in upper body strength after 10 weeks. Similarly, O'Connell and Barnhart [76] enrolled three children with SB and three with cerebral palsy, all of whom were wheelchair users, in a program of upper body resistance training performed three times per week for nine weeks. Despite a wide range of ages (5–16 years old) and abilities of the participants, the exercise program resulted in an increase in upper body strength (70–200% improvement) and self-propelled wheelchair distance covered in 12 minutes (29%). Although each of those studies demonstrate the potential benefits of upper body endurance and strengthening exercises on physical function for children with SB, the results are difficult to generalize due to the small sample sizes and heterogeneity of the participants. None of the studies assessed the impact of the exercise program on body composition or cardiometabolic risk factors such as lipids, glucose, insulin, blood pressure, or vascular function. Additionally, the pretraining familiarization and testing protocols were not well described so it is possible that at least part of the improvements ascribed to exercise training may be due to learning effects that can occur when novice participants gain increased awareness to study goals and are more familiar with the testing environment at the posttest compared to the baseline measurement.

More recently, de Groot et al. [77] conducted a home-based walking trial for ambulatory children with SB. Children in the intervention group were provided with a treadmill and instructed to exercise for up to 30 minutes per session in two days per week for 12 weeks in addition to their normal daily activities, while children in the standard-of-care control group maintained their habitual lifestyle patterns. At the end of the program, the walking group increased the distance they could walk during a 6-minute test by 38%, and this improvement was maintained for at least three months, whereas the walking distance in the control group was unchanged. Similarly, peak aerobic capacity and walking economy improved in the walking group but not the control group. This study demonstrates that children with SB can

improve their functional ability by performing a structured, progressive intensity exercise program. It remains to be shown, however, if these positive results can be extended to people with SB who are nonambulatory, and whether the benefits of the exercise translate to improvement in metabolic and vascular outcomes.

3.3. Exercise Programs for People with SCI. The value of considering exercise intervention programs for people with SCI is that there have been far more studies conducted compared to the situation for SB [78, 79]. Supporting the small evidence base for SB, the more extensive literature on SCI indicates that regular exercise is beneficial for people with limited mobility, which has led to the development of structured exercise recommendations for spinal cord injured people [80–82]. In adults with SCI cross-sectional studies demonstrate that the volume of daily physical activity is inversely associated with obesity and several metabolic risk factors, and positively related to aerobic fitness and quality-of-life assessments [83–85]. Likewise, beneficial changes in health and functional outcomes are observed when adults with SCI complete exercise training or functional electrical stimulation protocols [35, 79, 86–90]. Some of these studies used small sample sizes [87, 90, 91], but despite that limitation, the potential for metabolic benefits is promising. For example, de Groot et al. [87] showed that after 8 weeks of upper body exercise, men with SCI who performed high-intensity interval training had better improvement in aerobic capacity and fasting lipid profile than those who performed continuous moderate-intensity exercise. Although that study only included 3 men per group and requires confirmation, the results are consistent with several recent reports demonstrating that, compared to moderate-intensity exercise, aerobic interval training produced equal or greater fitness and metabolic benefits for obese adolescents and adults with heart disease or metabolic syndrome [92–96]. In the home-based walking program for children with SB described previously, de Groot et al. [77] used a modified version of this type of aerobic interval exercise. Thus, using short segments of higher intensity exercise could be an effective, time-efficient means to promote favorable fitness and metabolic adaptations in youth with SB. This and other exercise strategies need to be tested systematically, with the types of adaptations required to accommodate the needs and abilities of the individual carefully documented and disseminated.

4. Summary and Recommendations

In summary, although it may seem intuitive that exercise should be a cornerstone of healthy lifestyle recommendations for SB patients, there are so few studies on the effects of exercise in this population that it is unclear whether specific types of activity are more effective than others. Recent exercise recommendations have been developed for adults with SCI and consist of at least 20 min of moderate-to-vigorous intensity aerobic activity performed two times per week, and strength training exercises performed two times per week, comprised of three sets of 8–10 repetitions of exercises targeting each major muscle group [80]. Although it is plausible that this recommendation is appropriate and effective for children with SB, it is still unclear whether adjustments are needed to address the physical challenges that accompany SB or to meet specific health and function needs. For youth with SB who have limited use of their lower limbs, exercise must necessarily rely on the upper body and trunk muscles. For wheelchair users, this often means that there are fewer opportunities for physical activity and sports. Children with SB face barriers of access to appropriate exercise facilities and must rely on adults to help organize and supervise sports or physical play opportunities, which may be lacking in many communities. Thus, there is a potential role for healthcare providers to work with local fitness centers and schools to develop activity programs that incorporate structural exercise, sports programs like wheelchair basketball, and age- and function-appropriate movement games. Since upper body exercise activates less muscle mass than typical leg exercise it may be necessary to carefully regulate intensity or duration of activity, or to incorporate electrical stimulation of the lower limb musculature to ultimately reduce body fatness, insulin resistance, inflammation, and/or hyperlipidemia. Our premise is that distinct exercise approaches that vary in the volume, intensity, frequency, and/or mode of activity could be developed to target different outcomes in physical function and metabolic health. Achieving that goal will require detailed investigations that address the feasibility and effectiveness of exercise for children with SB. In the meantime, clinicians that work with patients with SB should encourage them to strive to increase their habitual physical activity on most or all days of the week, since the literature supports that this may help, improve physical function and perceived quality of life.

Conflict of Interests

The authors have no conflict of interests to declare.

Acknowledgments

This project was supported by National Center for Research Resources grants P20-RR024215 to K.R. Short. The content is solely the responsibility of the authors and does not necessarily represent the official views of the National Center for Research Resources or the National Institutes of Health.

References

[1] C. M. Shaer, N. Chescheir, and J. Schulkin, "Myelomeningocele: a review of the epidemiology, genetics, risk factors for conception, prenatal diagnosis, and prognosis for affected individuals," *Obstetrical and Gynecological Survey*, vol. 62, no. 7, pp. 471–479, 2007.

[2] L. Y. C. Wong and L. J. Paulozzi, "Survival of infants with spina bifida: a population study, 1979–94," *Paediatric and Perinatal Epidemiology*, vol. 15, no. 4, pp. 374–378, 2001.

[3] K. A. Bol, J. S. Collins, and R. S. Kirby, "Survival of infants with neural tube defects in the presence of folic acid fortification," *Pediatrics*, vol. 117, no. 3, pp. 803–813, 2006.

[4] M. Shin, L. M. Besser, C. Siffel et al., "Prevalence of spina bifida among children and adolescents in 10 regions in the United States," *Pediatrics*, vol. 126, no. 2, pp. 274–279, 2010.

[5] Spina Bifida Association, 2012, http://www.spinabifidaassociation.org.

[6] N. Akalan, "Myelomeningocele (open spina bifida)—surgical management," *Advances and Technical Standards in Neurosurgery*, vol. 37, pp. 113–141, 2011.

[7] S. L. Huang, W. Shi, and L. G. Zhang, "Characteristics and surgery of cervical myelomeningocele," *Child's Nervous System*, vol. 26, no. 1, pp. 87–91, 2010.

[8] J. H. Rimmer, W. Schiller, and M. D. Chen, "Effects of disability-associated low energy expenditure deconditioning syndrome," *Exercise and Sport Sciences Reviews*, vol. 40, no. 1, pp. 22–29, 2012.

[9] L. M. Dagenais, E. R. Lahay, K. A. Stueck, E. White, L. Williams, and S. R. Harris, "Effects of electrical stimulation, exercise training and motor skills training on strength of children with meningomyelocele: a systematic review," *Physical and Occupational Therapy in Pediatrics*, vol. 29, no. 4, pp. 445–463, 2009.

[10] L. M. Widman, R. T. Abresch, D. M. Styne, and C. M. McDonald, "Aerobic fitness and upper extremity strength in patients aged 11 to 21 years with spinal cord dysfunction as compared to ideal weight and overweight controls," *Journal of Spinal Cord Medicine*, vol. 30, no. 1, pp. S88–S96, 2007.

[11] M. A. G. C. Schoenmakers, J. F. de Groot, J. W. Gorter, J. L. M. Hillaert, P. J. M. Helders, and T. Takken, "Muscle strength, aerobic capacity and physical activity in independent ambulating children with lumbosacral spina bifida," *Disability and Rehabilitation*, vol. 31, no. 4, pp. 259–266, 2009.

[12] M. D. Nelson, L. M. Widman, R. T. Abresch et al., "Metabolic syndrome in adolescents with spinal cord dysfunction," *Journal of Spinal Cord Medicine*, vol. 30, no. 1, pp. S127–S139, 2007.

[13] K. Mita, K. Akataki, K. Itoh, Y. Ono, N. Ishida, and T. Oki, "Assessment of obesity of children with spina bifida," *Developmental Medicine and Child Neurology*, vol. 35, no. 4, pp. 305–311, 1993.

[14] K. Shepherd, D. Roberts, S. Golding, B. J. Thomas, and R. W. Shepherd, "Body composition in myelomeningocele," *American Journal of Clinical Nutrition*, vol. 53, no. 1, pp. 1–6, 1991.

[15] N. P. Dosa, J. T. Foley, M. Eckrich, D. Woodall-Ruff, and G. S. Liptak, "Obesity across the lifespan among persons with spina bifida," *Disability and Rehabilitation*, vol. 31, no. 11, pp. 914–920, 2009.

[16] R. A. Liusuwan, L. M. Widman, R. T. Abresch, D. M. Styne, and C. M. McDonald, "Body composition and resting energy expenditure in patients aged 11 to 21 years with spinal cord dysfunction compared to controls: comparisons and relationships among the groups," *Journal of Spinal Cord Medicine*, vol. 30, no. 1, pp. S105–S111, 2007.

[17] L. M. Buffart, R. J. G. van den Berg-Emons, M. S. van Wijlen-Hempel, H. J. Stam, and M. E. Roebroeck, "Health-related physical fitness of adolescents and young adults with myelomeningocele," *European Journal of Applied Physiology*, vol. 103, no. 2, pp. 181–188, 2008.

[18] A. L. Bruinings, H. J. G. van den Berg-Emons, L. M. Buffart, H. C. M. van der Heijden-Maessen, M. E. Roebroeck, and H. J. Stam, "Energy cost and physical strain of daily activities in adolescents and young adults with myelomeningocele," *Developmental Medicine and Child Neurology*, vol. 49, no. 9, pp. 672–677, 2007.

[19] H. J. G. van den Berg-Emons, J. B. Bussmann, A. S. Brobbel, M. E. Roebroeck, J. van Meeteren, and H. J. Stam, "Everyday physical activity in adolescents and young adults with meningomyelocele as measured with a novel activity monitor," *Journal of Pediatrics*, vol. 139, no. 6, pp. 880–886, 2001.

[20] L. M. Buffart, M. E. Roebroeck, M. Rol, H. J. Stam, and R. J. G. van den Berg-Emons, "Triad of physical activity, aerobic fitness and obesity in adolescents and young adults with myelomeningocele," *Journal of Rehabilitation Medicine*, vol. 40, no. 1, pp. 70–75, 2008.

[21] R. T. Abresch, D. A. McDonald, L. M. Widman, K. McGinnis, and K. J. Hickey, "Impact of spinal cord dysfunction and obesity on the health-related quality of life of children and adolescents," *Journal of Spinal Cord Medicine*, vol. 30, no. 1, pp. S112–S118, 2007.

[22] L. M. Buffart, R. J. G. van den Berg-Emons, J. V. Meeteren, H. J. Stam, and M. E. Roebroeck, "Lifestyle, participation, and health-related quality of life in adolescents and young adults with myelomeningocele," *Developmental Medicine and Child Neurology*, vol. 51, no. 11, pp. 886–894, 2009.

[23] H. A. Barf, M. W. M. Post, M. Verhoef, A. Jennekens-Schinkel, R. H. J. M. Gooskens, and A. J. H. Prevo, "Restrictions in social participation of young adults with spina bifida," *Disability and Rehabilitation*, vol. 31, no. 11, pp. 921–927, 2009.

[24] C. Rendeli, E. Ausili, F. Tabacco et al., "Assessment of health status in children with spina bifida," *Spinal Cord*, vol. 43, no. 4, pp. 230–235, 2005.

[25] K. Casteels, S. Fieuws, M. van Helvoirt et al., "Metformin therapy to reduce weight gain and visceral adiposity in children and adolescents with neurogenic or myogenic motor deficit," *Pediatric Diabetes*, vol. 11, no. 1, pp. 61–69, 2010.

[26] C. Rendeli, M. Castorina, E. Ausili et al., "Risk factors for atherogenesis in children with spina bifida," *Child's Nervous System*, vol. 20, no. 6, pp. 392–396, 2004.

[27] L. M. Buffart, R. J. van den Berg-Emons, A. Burdorf, W. G. Janssen, H. J. Stam, and M. E. Roebroeck, "Cardiovascular disease risk factors and the relationships with physical activity, aerobic fitness, and body fat in adolescents and young adults with myelomeningocele," *Archives of Physical Medicine and Rehabilitation*, vol. 89, no. 11, pp. 2167–2173, 2008.

[28] C. Rendeli, E. Ausili, M. Castorina, D. Antuzzi, F. Tabacco, and M. Caldarelli, "Homocysteine, folate, lipid profile and MTHFR genotype and disability in children with myelomeningocele," *Child's Nervous System*, vol. 22, no. 10, pp. 1316–1321, 2006.

[29] P. R. Nader, R. H. Bradley, R. M. Houts, S. L. McRitchie, and M. O'Brien, "Moderate-to-vigorous physical activity from ages 9 to 15 years," *Journal of the American Medical Association*, vol. 300, no. 3, pp. 295–305, 2008.

[30] C. R. L. Boot, H. van Langen, and M. T. E. Hopman, "Arterial vascular properties in individuals with spina bifida," *Spinal Cord*, vol. 41, no. 4, pp. 242–246, 2003.

[31] Christopher & Dana Reeve Foundation, *One Degree of Separation: Paralysis and Spinal Cord Injury in the United States*, Christopher & Dana Reeve Foundation, Short Hills, NJ, USA, 2009.

[32] C. M. McDonald, A. L. Abresch-Meyer, M. D. Nelson, and L. M. Widman, "Body mass index and body composition measures by dual X-ray absorptiometry in patients aged 10

to 21 years with spinal cord injury," *Journal of Spinal Cord Medicine*, vol. 30, no. 1, pp. S97–S104, 2007.

[33] J. F. de Groot, T. Takken, M. A. G. C. Schoenmakers, L. Vanhees, and P. J. M. Helders, "Limiting factors in peak oxygen uptake and the relationship with functional ambulation in ambulating children with spina bifida," *European Journal of Applied Physiology*, vol. 104, no. 4, pp. 657–665, 2008.

[34] K. Wahman, M. S. Nash, J. E. Lewis, A. Seiger, and R. Levi, "Cardiovascular disease risk and the need for prevention after paraplegia determined by conventional multifactorial risk models: the Stockholm spinal cord injury study," *Journal of Rehabilitation Medicine*, vol. 43, no. 3, pp. 237–242, 2011.

[35] R. E. Cowan and M. S. Nash, "Cardiovascular disease, SCI and exercise: unique risks and focused countermeasures," *Disability and Rehabilitation*, vol. 32, no. 26, pp. 2228–2236, 2010.

[36] T. D. Wang, Y. H. Wang, T. S. Huang, T. C. Su, S. L. Pan, and S. Y. Chen, "Circulating levels of markers of inflammation and endothelial activation are increased in men with chronic spinal cord injury," *Journal of the Formosan Medical Association*, vol. 106, no. 11, pp. 919–928, 2007.

[37] M. S. Nash, J. DeGroot, A. Martinez-Arizala, and A. J. Mendez, "Evidence for an exaggerated postprandial lipemia in chronic paraplegia," *Journal of Spinal Cord Medicine*, vol. 28, no. 4, pp. 320–325, 2005.

[38] G. D. Kolovou, D. C. Daskalova, S. A. Iraklianou et al., "Postprandial lipemia in hypertension," *Journal of the American College of Nutrition*, vol. 22, no. 1, pp. 80–87, 2003.

[39] C. L. Ogden, M. D. Carroll, and K. M. Flegal, "High body mass index for age among US children and adolescents, 2003–2006," *Journal of the American Medical Association*, vol. 299, no. 20, pp. 2401–2405, 2008.

[40] D. K. Eaton, L. Kann, S. A. Kinchen et al., "Youth risk behavior surveillance—United States, 2009," *MMWR CDC Surveillance Summaries*, vol. 59, pp. 1–142, 2010.

[41] R. Sinha, G. Fisch, B. Teague et al., "Prevalence of impaired glucose tolerance among children and adolescents with marked obesity," *The New England Journal of Medicine*, vol. 346, pp. 802–810, 2002.

[42] C. W. Yeckel, S. E. Taksali, J. Dziura et al., "The normal glucose tolerance continuum in obese youth: evidence for impairment in β-cell function independent of insulin resistance," *Journal of Clinical Endocrinology and Metabolism*, vol. 90, no. 2, pp. 747–754, 2005.

[43] D. E. Williams, B. L. Cadwell, Y. J. Cheng et al., "Prevalence of impaired fasting glucose and its relationship with cardiovascular disease risk factors in US adolescents, 1999-2000," *Pediatrics*, vol. 116, no. 5, pp. 1122–1126, 2005.

[44] A. Syrenicz, B. Garanty-Bogacka, M. Syrenicz, A. Gebala, and M. Walczak, "Low-grade systemic inflammation and the risk of type 2 diabetes in obese children and adolescents," *Neuroendocrinology Letters*, vol. 27, no. 4, pp. 453–458, 2006.

[45] J. C. Winer, T. L. Zern, S. E. Taksali et al., "Adiponectin in childhood and adolescent obesity and its association with inflammatory markers and components of the metabolic syndrome," *Journal of Clinical Endocrinology and Metabolism*, vol. 91, no. 11, pp. 4415–4423, 2006.

[46] P. Balagopal, D. George, N. Patton et al., "Lifestyle-only intervention attenuates the inflammatory state associated with obesity: a randomized controlled study in adolescents," *Journal of Pediatrics*, vol. 146, no. 3, pp. 342–348, 2005.

[47] S. Lee, F. Bacha, N. Gungor, and S. Arslanian, "Comparison of different definitions of pediatric metabolic syndrome: relation to abdominal adiposity, insulin resistance, adiponectin, and inflammatory biomarkers," *Journal of Pediatrics*, vol. 152, no. 2, pp. 177–184.e3, 2008.

[48] M. Juonala, J. S. A. Viikari, M. Kahonen et al., "Childhood levels of serum apolipoproteins B and A-I predict carotid intima-media thickness and brachial endothelial function in adulthood: the cardiovascular risk in young Finns study," *Journal of the American College of Cardiology*, vol. 52, no. 4, pp. 293–299, 2008.

[49] D. S. Freedman, D. A. Patel, S. R. Srinivasan et al., "The contribution of childhood obesity to adult carotid intima-media thickness: the Bogalusa heart study," *International Journal of Obesity*, vol. 32, no. 5, pp. 749–756, 2008.

[50] S. R. Srinivasan, L. Myers, and G. S. Berenson, "Predictability of childhood adiposity and insulin for developing insulin resistance syndrome (syndrome X) in young adulthood: the Bogalusa heart study," *Diabetes*, vol. 51, no. 1, pp. 204–209, 2002.

[51] A. R. A. Adegboye, S. A. Anderssen, K. Froberg et al., "Recommended aerobic fitness level for metabolic health in children and adolescents: a study of diagnostic accuracy," *British Journal of Sports Medicine*, vol. 45, no. 9, pp. 722–728, 2011.

[52] D. E. Laaksonen, J. Lindstrom, T. A. Lakka et al., "Physical activity in the prevention of type 2 diabetes: the finnish diabetes prevention study," *Diabetes*, vol. 54, no. 1, pp. 158–165, 2005.

[53] Diabetes Prevention Program Research Group, "10-year follow-up of diabetes incidence and weight loss in the diabetes prevention program outcomes study," *The Lancet*, vol. 374, no. 9702, pp. 1677–1686, 2009.

[54] A. S. Thomas, L. F. Greene, J. D. Ard, R. A. Oster, B. E. Darnell, and B. A. Gower, "Physical activity may facilitate diabetes prevention in adolescents," *Diabetes Care*, vol. 32, no. 1, pp. 9–13, 2009.

[55] S. Lee, J. L. Kuk, P. T. Katzmarzyk, S. N. Blair, T. S. Church, and R. Ross, "Cardiorespiratory fitness attenuates metabolic risk independent of abdominal subcutaneous and visceral fat in men," *Diabetes Care*, vol. 28, no. 4, pp. 895–901, 2005.

[56] K. R. Short, J. L. Vittone, M. L. Bigelow et al., "Impact of aerobic exercise training on age-related changes in insulin sensitivity and muscle oxidative capacity," *Diabetes*, vol. 52, no. 8, pp. 1888–1896, 2003.

[57] B. A. Irving, K. R. Short, K. S. Nair, and C. S. Stump, "Nine days of intensive exercise training improves mitochondrial function but not insulin action in adult offspring of mothers with type 2 diabetes," *Journal of Clinical Endocrinology and Metabolism*, vol. 96, no. 7, pp. E1137–E1141, 2011.

[58] A. Igwebuike, B. A. Irving, M. L. Bigelow, K. R. Short, J. P. McConnell, and K. S. Nair, "Lack of dehydroepiandrosterone effect on a combined endurance and resistance exercise program in postmenopausal women," *Journal of Clinical Endocrinology and Metabolism*, vol. 93, no. 2, pp. 534–538, 2008.

[59] G. P. Nassis, K. Papantakou, K. Skenderi et al., "Aerobic exercise training improves insulin sensitivity without changes in body weight, body fat, adiponectin, and inflammatory markers in overweight and obese girls," *Metabolism*, vol. 54, no. 11, pp. 1472–1479, 2005.

[60] G. A. Kelley and K. S. Kelley, "Aerobic exercise and lipids and lipoproteins in children and adolescents: a meta-analysis of randomized controlled trials," *Atherosclerosis*, vol. 191, no. 2, pp. 447–453, 2007.

[61] C. A. Slentz, B. D. Duscha, J. L. Johnson et al., "Effects of the amount of exercise on body weight, body composition, and measures of central obesity: STRRIDE—a randomized controlled study," *Archives of Internal Medicine*, vol. 164, no. 1, pp. 31–39, 2004.

[62] J. O. Holloszy, "Exercise-induced increase in muscle insulin sensitivity," *Journal of Applied Physiology*, vol. 99, no. 1, pp. 338–343, 2005.

[63] G. J. van der Heijden, G. Toffolo, E. Manesso, P. J. J. Sauer, and A. L. Sunehag, "Aerobic exercise increases peripheral and hepatic insulin sensitivity in sedentary adolescents," *Journal of Clinical Endocrinology and Metabolism*, vol. 94, no. 11, pp. 4292–4299, 2009.

[64] F. Magkos, D. C. Wright, B. W. Patterson, B. S. Mohammed, and B. Mittendorfer, "Lipid metabolism response to a single, prolonged bout of endurance exercise in healthy young men," *American Journal of Physiology*, vol. 290, no. 2, pp. E355–E362, 2006.

[65] J. M. R. Gill and A. E. Hardman, "Postprandial lipemia: effects of exercise and restriction of energy intake compared," *American Journal of Clinical Nutrition*, vol. 71, no. 2, pp. 465–471, 2000.

[66] J. M. R. Gill, G. P. Mees, K. N. Frayn, and A. E. Hardman, "Moderate exercise, postprandial lipaemia and triacylglycerol clearance," *European Journal of Clinical Investigation*, vol. 31, no. 3, pp. 201–207, 2001.

[67] O. J. MacEneaney, M. Harrison, D. J. O'Gorman, E. V. Pankratieva, P. L. O'Connor, and N. M. Moyna, "Effect of prior exercise on postprandial lipemia and markers of inflammation and endothelial activation in normal weight and overweight adolescent boys," *European Journal of Applied Physiology*, vol. 106, no. 5, pp. 721–729, 2009.

[68] C. A. Macera, D. A. Jones, M. M. Yore et al., "Prevalence of physical activity, including lifestyle activities among adults: United States, 2000-2001," *Morbidity and Mortality Weekly Report*, vol. 52, pp. 764–769, 2003.

[69] L. M. Buffart, H. P. van der Ploeg, A. E. Bauman et al., "Sports participation in adolescents and young adults with myelomeningocele and its role in total physical activity behaviour and fitness," *Journal of Rehabilitation Medicine*, vol. 40, no. 9, pp. 702–708, 2008.

[70] J. An and D. L. Goodwin, "Physical education for students with spina bifida: mothers' perspectives," *Adapted Physical Activity Quarterly*, vol. 24, no. 1, pp. 38–58, 2007.

[71] K. A. Hinderer, S. R. Hinderer, and D. B. Shurtleff, "Myelodysplasia," in *Physical Therapy for Children*, S. K. Campbell, D. W. Van der Linden, and R. J. Palisano, Eds., pp. 735–789, Elsevier Saunders, Philadelphia, Pa, USA, 3rd edition, 2006.

[72] K. D. Ryan, C. Ploski, and J. B. Emans, "Myelodysplasia—the musculoskeletal problem: habilitation from infancy to adulthood," *Physical Therapy*, vol. 71, no. 12, pp. 935–946, 1991.

[73] J. M. Mazur and S. Kyle, "Efficacy of bracing the lower limbs and ambulation training in children with myelomeningocele," *Developmental Medicine and Child Neurology*, vol. 46, no. 5, pp. 352–356, 2004.

[74] B. Ekblom and A. Lundberg, "Effect of physical training on adolescents with severe motor handicaps," *Acta Paediatrica Scandinavica*, vol. 57, no. 1, pp. 17–23, 1968.

[75] C. K. Andrade, J. Kramer, M. Garber, and P. Longmuir, "Changes in self-concept, cardiovascular endurance and muscular strength of children with spina bifida aged 8 to 13 years in response to a 10-week physical-activity programme: a pilot study," *Child*, vol. 17, no. 3, pp. 183–196, 1991.

[76] D. G. O'Connell and R. Barnhart, "Improvement in wheelchair propulsion in pediatric wheelchair users through resistance training: a pilot study," *Archives of Physical Medicine and Rehabilitation*, vol. 76, no. 4, pp. 368–372, 1995.

[77] J. F. de Groot, T. Takken, M. van Brussel et al., "Randomized controlled study of home-based treadmill training for ambulatory children with spina bifida," *Neurorehabilitation and Neural Repair*, vol. 25, pp. 597–606, 2011.

[78] A. A. Phillips, A. T. Cote, and D. E. R. Warburton, "A systematic review of exercise as a therapeutic intervention to improve arterial function in persons living with spinal cord injury," *Spinal Cord*, vol. 49, no. 6, pp. 702–714, 2011.

[79] M. S. Nash, "Exercise as a health-promoting activity following spinal cord injury," *Journal of Neurologic Physical Therapy*, vol. 29, pp. 87–103, 2005.

[80] K. A. M. Ginis, A. L. Hicks, A. E. Latimer et al., "The development of evidence-informed physical activity guidelines for adults with spinal cord injury," *Spinal Cord*, vol. 49, pp. 1088–1096, 2011.

[81] K. A. Martin Ginis, A. E. Latimer, A. C. Buchholz et al., "Establishing evidence-based physical activity guidelines: methods for the study of health and activity in people with spinal cord injury (SHAPE SCI)," *Spinal Cord*, vol. 46, no. 3, pp. 216–221, 2008.

[82] R. E. Cowan, L. A. Malone, and M. S. Nash, "Exercise is medicine: exercise prescription after SCI to manage cardiovascular disease risk factors," *Topics in Spinal Cord Injury Rehabilitation*, vol. 14, no. 3, pp. 69–83, 2009.

[83] C. F. J. Nooijen, S. de Groot, K. Postma et al., "A more active lifestyle in persons with a recent spinal cord injury benefits physical fitness and health," *Spinal Cord*, vol. 50, pp. 320–323, 2012.

[84] A. C. Buchholz, K. A. M. Ginis, S. R. Bray et al., "Greater daily leisure time physical activity is associated with lower chronic disease risk in adults with spinal cord injury," *Applied Physiology, Nutrition and Metabolism*, vol. 34, no. 4, pp. 640–647, 2009.

[85] H. Liang, K. Tomey, D. Chen, N. L. Savar, J. H. Rimmer, and C. L. Braunschweig, "Objective measures of neighborhood environment and self-reported physical activity in spinal cord injured men," *Archives of Physical Medicine and Rehabilitation*, vol. 89, no. 8, pp. 1468–1473, 2008.

[86] X. Devillard, D. Rimaud, F. Roche, and P. Calmels, "Effects of training programs in spinal cord injury," *Annales de Readaptation et de Medecine Physique*, vol. 50, no. 6, pp. 480–489, 2007.

[87] P. C. E. de Groot, N. Hjeltnes, A. C. Heijboer, W. Stal, and K. Birkeland, "Effect of training intensity on physical capacity, lipid profile and insulin sensitivity in early rehabilitation of spinal cord injured individuals," *Spinal Cord*, vol. 41, no. 12, pp. 673–679, 2003.

[88] M. S. Nash, I. van de Ven, N. van Elk, and B. M. Johnson, "Effects of circuit resistance training on fitness attributes and upper-extremity pain in middle-aged men with paraplegia," *Archives of Physical Medicine and Rehabilitation*, vol. 88, no. 1, pp. 70–75, 2007.

[89] A. L. Hicks, K. A. Martin, D. S. Ditor et al., "Long-term exercise training in persons with spinal cord injury: effects on strength, arm ergometry performance and psychological well-being," *Spinal Cord*, vol. 41, no. 1, pp. 34–43, 2003.

[90] S. Turbanski and D. Schmidtbleicher, "Effects of heavy resistance training on strength and power in upper extremities in wheelchair athletes," *Journal of Strength and Conditioning Research*, vol. 24, no. 1, pp. 8–16, 2010.

[91] A. S. Gorgey, K. J. Mather, H. R. Cupp, and D. R. Gater, "Effects of resistance training on adiposity and metabolism after spinal cord injury," *Medicine & Science in Sports & Exercise*, vol. 44, no. 1, pp. 165–174, 2012.

[92] U. Wisløff, A. Støylen, J. P. Loennechen et al., "Superior cardiovascular effect of aerobic interval training versus moderate continuous training in heart failure patients: a randomized study," *Circulation*, vol. 115, no. 24, pp. 3086–3094, 2007.

[93] P. S. Munk, E. M. Staal, N. Butt, K. Isaksen, and A. I. Larsen, "High-intensity interval training may reduce in-stent restenosis following percutaneous coronary intervention with stent implantation. A randomized controlled trial evaluating the relationship to endothelial function and inflammation," *American Heart Journal*, vol. 158, no. 5, pp. 734–741, 2009.

[94] Ø. Rognmo, E. Hetland, J. Helgerud, J. Hoff, and S. A. Slordahl, "High intensity aerobic interval exercise is superior to moderate intensity exercise for increasing aerobic capacity in patients with coronary artery disease," *European Journal of Cardiovascular Prevention and Rehabilitation*, vol. 11, no. 3, pp. 216–222, 2004.

[95] A. E. Tjønna, S. J. Lee, Ø. Rognmo et al., "Aerobic interval training versus continuous moderate exercise as a treatment for the metabolic syndrome: a pilot study," *Circulation*, vol. 118, no. 4, pp. 346–354, 2008.

[96] A. E. Tjønna, T. O. Stølen, A. Bye et al., "Aerobic interval training reduces cardiovascular risk factors more than a multitreatment approach in overweight adolescents," *Clinical Science*, vol. 116, no. 4, pp. 317–326, 2009.

Hindawi Publishing Corporation
International Journal of Pediatrics
Volume 2012, Article ID 168209, 8 pages
doi:10.1155/2012/168209

Research Article

Arterial Structure and Function in Ambulatory Adolescents with Cerebral Palsy Are Not Different from Healthy Controls

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Received 1 December 2011; Revised 14 March 2012; Accepted 30 March 2012

Academic Editor: Margaret E. O'Neil

Physical inactivity in youth with cerebral palsy (CP) places them at increased risk of developing cardiovascular disease. The current study assessed indices of arterial health in adolescents with CP, classified as levels I-II of the Gross Motor Function Classification System (GMFCS) ($n = 11$, age 13.2 ± 2.1 yr), in comparison to age- and sex-matched controls ($n = 11$, age 12.4 ± 2.3 yr). Groups were similar in anthropometric measurements, resting blood pressures, and heart rates. There were no group differences in brachial flow-mediated dilation (11.1 ± 7.8 versus 6.1 ± 3.6), carotid intima-media thickness (0.42 ± 0.04 versus 0.41 ± 0.03 mm), and distensibility (0.008 ± 0.002 versus 0.008 ± 0.002 mmHg) or central (4.3 ± 0.6 versus 4.1 ± 0.9 m/s) and peripheral pulse wave velocity (7.1 ± 1.7 versus 7.6 ± 1.1 m/s); CP versus healthy controls, respectively. Vigorous intensity physical activity (PA) was lower in the CP group (CP: 38 ± 80 min versus controls: 196 ± 174 min); groups were similar in light and moderate intensity PA levels. Arterial health of ambulatory youth with CP is not different from a control group despite lower vigorous PA levels. Similar studies need to examine individuals with more pronounced mobility limitations (GMFCS level III–V).

1. Introduction

Cerebral Palsy (CP) is defined as a disorder of posture and movement due to a nonprogressive disturbance in the developing fetal or infant brain [1]. CP manifests as limitations in gross motor capacity [2], affecting performance in daily mobility over a lifespan [3]. Youth with CP are less physically active than their typically developing peers [4, 5] and show an inverse relationship between functional limitations and social participation [6]. Physical inactivity in youth places them at a greater risk of developing a variety of secondary health complications [7] and is also a major controllable risk factor for cardiovascular disease (CVD) [8].

It has been suggested that one mechanism by which physical activity (PA) exerts its protective effect on cardiovascular health is through positive effects on the endothelium [9], a single layer of cells responsible for the vasodilator response to increased conduit artery flow. A strong relationship between low levels of PA and endothelial dysfunction has been well documented in children [10], potentially predisposing youth with CP to an increased risk of endothelial dysfunction. Endothelial dysfunction is considered an early and integral manifestation of atherosclerotic disease, which can be evident in the first decade of life [11]. Endothelial dysfunction is an indicator of preclinical vascular disease and for youth with CP may act as a marker of early changes in vessel function, indicative of future atherosclerotic risk [12].

Pulse wave velocity (PWV) is a sensitive marker of arterial wall stiffness and subsequent marker of cardiovascular risk [13]. In children, PWV is positively correlated with body mass index (BMI), waist circumference (WC), and percentage body fat and negatively correlated with cardiorespiratory fitness and levels of PA [14]. Carotid artery distensibility and carotid artery intima-media thickness ($_c$IMT) are two additional indices of arterial health and their role in the development of CVD is widely accepted [15].

Strong relationships between cardiovascular risk factors identified in childhood and adolescence and the progression of atherosclerosis in adulthood are emerging [16]. Consistent, positive effects of habitual PA on vessel health have been demonstrated [14, 17]. Measuring indices of arterial stiffness and endothelial function are therefore important in this young, clinical population in order to identify changes in vascular health at the earliest stage possible.

To our knowledge, there is no study published assessing vessel health in general or its association with levels of habitual PA in youth with CP. Given the fact that children aged 5 to 7 years with CP have lower PA levels than typically developing peers [4, 5] we hypothesize that even the most functional adolescents with CP (GMFCS levels I-II) may have decreased levels of PA and altered arterial function and structure compared to an age- and sex-matched control sample.

2. Methods

2.1. Participants. Twenty-two adolescents (9–16 yrs) were recruited; of which, 11 individuals with CP (8 boys; mean ± SD age of 13.2 ± 2.1 yr) were recruited from the Spasticity Clinic and Teen Transition Clinic at McMaster Children's Hospital, Hamilton, Ontario, Canada. Inclusion criteria for the CP group included a classification of either a level I or II (GMFCS-Expanded & Revised) [18] indicating that all subjects with CP were ambulatory without use of mobility devices (level I $n = 7$, level II $n = 4$). Subjects were chronological age- and sex-matched to a healthy control group with a mean age of 12.4 ± 2.3 yr. Control subjects were healthy, with no known cardiovascular or metabolic conditions and studied without specific exclusion criteria. Experimental procedures were explained to participants and their guardians prior to obtaining written and verbal informed consent/assent from the parent/guardian and participant, respectively. Approval from the Hamilton Health Sciences and McMaster University Faculty of Health Sciences Research Ethics Board was obtained for the study.

2.2. Study Design. This study employed a cross-sectional design to characterize the differences in specific measures of vascular structure and function between children with CP and healthy controls. All measures were noninvasive and took place in a quiet, temperature-controlled room (23° ± 1°C) with the participant in a supine position. All subjects were instructed to abstain from vigorous PA 24 hours pre- and were tested 4 hours postprandialy [19].

2.2.1. Anthropometric Measurements. Sitting and standing height (cm) were measured to the nearest mm without shoes and in light clothing. Body mass was measured to the nearest 0.1 kg using a digital scale, and BMI was calculated. WC was measured 4 cm above the umbilicus at the end of a normal expiration [20]. Two measurements were taken for each variable with a third required if a difference greater than 4 mm for height and WC and 0.4 kg for weight [21, 22] existed. For height and weight, the average of the two measurements was reported, and the median value was reported if three measurements were obtained [23]. Waist-to-height ratio (WHR) was calculated as the WC divided by the height (cm). As a marker of biological maturity, each individuals' age at peak height velocity (APHV) and time from peak height velocity (TPHV) was calculated using a gender specific equation [24].

2.2.2. Resting Heart Rate and Blood Pressure. Testing sessions began with 10 min. of supine rest to ensure representative resting measurements prior to the commencement of the vascular assessment [25]. Continuous heart rate via a single-lead electrocardiograph and brachial blood pressure (BP) measurements via an automated applanation tonometer with oscillometric cuff calibration (model CBM-7000; Colin Medical Instruments, San Antonio, TX) were collected. All signals (including those described below) were acquired simultaneously using a commercially available data acquisition system (Powerlab model ML795, ADInstruments, Colorado Springs, USA) and software program (LabChart 7; ADInstruments Inc., Colorado Springs, CO, USA). At the end of the vascular assessment, four measurements of seated brachial artery pressure were obtained using an automated sphygmomanometer (Dinamap Pro 100, Critikon LCC, Tampa, FL). The first measurement was used for calibration purposes only and the average of the following three measures was reported [26].

2.3. Vascular Assessment

2.3.1. Pulse Wave Velocity. Baseline measurements of PWV were acquired through electrocardiography and photoplethysmography. Both central and peripheral PWV (cPWV and pPWV, resp.) were determined from 20 continuous heart cycles using the equation [13]:

$$\mathrm{PWV} = \frac{D}{\Delta t}, \tag{1}$$

where D is the distance between measurement sites and Δt is the pulse transit time. Arterial waveforms at the common carotid, femoral, and dorsalis pedis arteries were collected using photoplethysmograph (PPG) sensors (IR Plethysmograph; Model MLT1020PPG; ADInstruments, Colorado Springs) on the right side of the body. PPG signals were bandpass-filtered (5–30 Hz) with the lower (≤5 Hz) and higher frequencies (≥30 Hz) removed in order to assist in the detection of the foot of each waveform. The foot of each waveform was identified as the minimum value of the digitally filtered signal [27] and corresponds to the end of diastole, when the steep rise in the wave begins and appears as a sharp inflection of the original signal [28].

Central PTT was determined using the subtraction method [29]. Similarly, cPWV path length was calculated by subtracting the surface distance between the sternal notch and the carotid PPG placement from that of the sternal notch and the femoral PPG placement. Peripheral pulse transit time was determined as the time delay between the arrival of the femoral artery pulse wave and the dorsalis pedis artery pulse wave [19], with the path length measured as the distance between these two sites. Anthropometric measuring

tape was used to measure the straightline distances between skin sites (sternal notch to the placement of each PPG sensor) along the surface of the body.

2.3.2. Carotid Distensibility and Intima-Media Thickness. Direct measurements of carotid distensibility were acquired as previously described [30] using a combination of high-resolution, two-dimensional, B-mode ultrasound images (System FiVe; GE Medical Systems, Horten, Norway) and applanation tonometry (model SPT-301; Millar Instruments, Houston, TX, USA). A hand-held tonometer was positioned over the point of greatest pulsation and held in a fixed position for ten consecutive heart cycles while ultrasound images of the left common carotid artery were collected simultaneously. Absolute carotid artery systolic blood pressures were calculated by calibrating the relative values acquired using applanation tonometry to the calibrated brachial artery blood pressures acquired simultaneously [31, 32].

Ultrasound images were stored offline in Digital Image and Communications in Medicine (DICOM) format for later analysis using a semiautomated edge tracking system (AMS (Artery Measurement System) Image and Data Analysis. Tomas Gustavsson, gustav@alumni.chalmers.se) [30]. In each frame, carotid artery (minimum, mean, and maximum) lumen diameters were calculated from roughly 100 measurement markers along the vessel wall within the region of interest (ROI), for a total of 110 000 measures in a 10 heart cycle data sample. Distensibility was calculated using the equation [13]:

$$\text{Distensibility} = \frac{\prod (d_{\max}/2)^2 - \prod (d_{\min}/2)^2}{\prod (d_{\max}/2)^2 \times \text{PP}}, \tag{2}$$

where $d_{\max}$ is the maximum diameter, $d_{\min}$ is the minimum diameter, and PP is carotid pulse pressure, the change in pressure from DBP and SBP. The mean carotid diameter was calculated using the average of all diameters acquired throughout the ten heart cycles. The same software program and ultrasound images were used on the far wall of the carotid artery for measurement of the $_c$IMT.

2.3.3. Flow-Mediated Dilation Assessment. The flow-mediated dilation (FMD) assessment has been shown to be the most reproducible and least variable of the techniques used to measure endothelial function in children [33]. With the participant in the supine position, the left arm was positioned (roughly 80° from the torso) and immobilized so that an optimal image of the brachial artery could be obtained in a comfortable position [34]. A sphygmomanometric cuff was placed on the forearm, below the medial epicondyle [35], and remained deflated while baseline data were collected. B-mode ultrasound images of the left brachial artery were collected through two-dimensional grayscale ultrasound imaging using a 10 MHz linear array probe (System FiVe; GE Medical Systems, Horten, Norway). A baseline longitudinal image of the brachial artery (3 consecutive cardiac cycles) was acquired by a single ultrasonographer. An intensity-weighted sample volume was attained and the gate width was therefore adjusted accordingly.

To create the flow stimulus, the forearm cuff was instantaneously inflated to a standardized, suprasystolic pressure of 200 mmHg to ensure arterial inflow occlusion and ischemia of downstream vessels and tissue [35]. The cuff was instantaneously deflated after 5 min. of occlusion and the first 30 sec. of reactive hyperemic blood velocity signals were collected using pulsed-wave Doppler ultrasound. The forward and reverse frequency signals were processed by an external spectral analysis system (Neurovision 500 M, Multigon Ind; Yonkers NY) and an intensity-weighted calculated mean was output into a data acquisition system (Powerlab model ML795). B-mode ultrasound images of the brachial artery over three consecutive heart cycles were stored every 15 sec. from 30 sec. until 3 min. after cuff.

End-diastolic frames were extracted from each sequence of images using a DICOM editing software program (Sante DICOM Editor 3.1.13, Santesoft, Athens, Greece). The semi-automated edge detection software program (AMS) was used to detect the vessel diameters within a specific ROI for the three end-diastolic frames at each time point. The peak dilation of the vessel was established as the single largest end-diastolic diameter (mm) measured from 30 sec. to 3 min. after cuff release. From this data, the absolute FMD (mm) and relative FMD (%FMD) were calculated as follows [36]:

$$\begin{aligned} \text{Absolute FMD} &= \text{Peak Diameter (mm)} \\ &\quad - \text{Baseline Diameter (mm)}, \\ \text{Relative FMD} &= \left(\frac{\text{Absolute FMD}}{\text{Baseline Diameter}}\right) \times 100\%. \end{aligned} \tag{3}$$

The following equation was used to calculate shear rate (SR) for each participant [37]:

$$\text{Shear Rate} = 8 \times \left(\frac{\text{Velocity}}{\text{Diameter}}\right), \tag{4}$$

where velocity represents the mean blood flow velocity of the velocity profile of the first 30 sec. after cuff release and the baseline brachial diameter (mm) is used for the internal artery diameter value. The area under the curve of the shear rate was calculated from the mean of the first point, using the trapezoid rule to obtain the area under the entire curve (GraphPad Prism version 4.00 for Windows, GraphPad Software, San Diego California USA, http://www.graphpad.com/). Relative FMD (%FMD) was normalized to the area under the entire SR curve and reported as $\%\text{FMD}/\text{SR}_{\text{AUC}}$:

$$\text{Normalized FMD} = \left(\frac{\%\text{FMD}}{\text{SR}_{\text{AUC}}}\right). \tag{5}$$

This method of analysis provides values of absolute maximum dilation (mm), time to reach peak dilation (sec.), and a raw calculation of the SR stimulus (SR_{AUC}).

2.4. Physical Activity. Habitual PA patterns were assessed using the Exercise Questionnaire adopted from Brunton and Bartlett, used in a longitudinal study describing exercise participation of adolescents with CP [38]. This recall

questionnaire provides information regarding the frequency, duration, and intensity of PA performed in the previous week. This questionnaire, based on the "Previous Day Physical Activity Record" by Weston et al. (1997) [39], was used to assess PA in both the CP and control group.

2.5. Statistical Analysis. Statistical analyses were performed using SPSS Statistics, version 19.0 (SPSS, Inc., Chicago, IL). Data distribution was initially examined for normality using the Shapiro-Wilk's Test and homogeneity of variance using Levene's Test. Independent t-tests were used to compare group differences in all vascular indices, anthropometric measures, and levels of PA. Analyses of vascular indices were also completed with chronological age as a covariate. Statistical significance for all analyses was set at $P \leq 0.05$.

3. Results

Characteristics of the study population are described in Table 1. The control and CP group were of similar age, height, weight, WC, WHR, and BMI. There were no group differences in resting seated brachial systolic blood pressure, dias-tolic blood pressure, mean arterial pressure, or resting supine heart rate.

Outcomes of the flow-mediated dilation (FMD) assessment are reported in Table 2. It must be noted that one participant with CP was removed from all FMD analysis due to inadequate ultrasound image quality of postocclusion data. One control subject was also identified as an outlier (via box plot and a response greater than 2 SD above the mean) and removed from the analysis. Thus, all statistical analyses of endothelial function (Table 2) were performed with an $n = 10$ in each group, with the exception of the preocclusion brachial diameters ($n = 11$) as all pre-occlusion data remained acceptable for analysis. There were no differences between groups ($P > 0.05$) in pre-occlusion brachial diameter (mm) or peak diameter (mm) reached during reactive hyperemia (Table 2). There were no differences in the SR stimulus or time taken to reach peak diameter between groups (Table 2).

There were no differences in baseline measures of carotid distensibility, $_c$IMT, or baseline carotid diameter between groups (Table 3). One control subject was a significant outlier and removed from analysis of distensibility (CON, $n = 10$) and one CP subject could not be included in the analysis of $_c$IMT due to insufficient clarity of the far wall IMT for pro-per identification (CP, $n = 10$). No differences were seen in cPWV or pPWV or PTT between groups (Table 3). One individual from the control group could not be included in the analysis due to an arrhythmia that did not permit appropriate analysis of the PWV data (CON, $n = 10$).

The total number of minutes/week of PA in each intensity category is reported in Figure 1(a). There were no group differences in the total number of minutes spent in light and moderate PA. The CP group reported a significantly smaller amount of vigorous PA weekly than the control group (Figure 1(a)) with over 60% of individuals in the CP group reporting 0 minutes of total time spent performing vigorous PA in the previous week. Furthermore, when total PA time/week was calculated (combining each intensity of PA), there were no significant differences between groups (CP: 4260 min/week versus Controls: 4840 min/week) (Figure 1(b)).

Table 1: Subject characteristics.

	Control ($n = 11$)	CP ($n = 11$)	P value
Age, yrs	12.4 ± 2.3	13.2 ± 2.1	0.458
Height, m	1.6 ± 0.1	1.5 ± 0.1	0.169
Weight, kg	49.3 ± 14.2	41.4 ± 8.4	0.129
APHV	13.08 ± 0.9	14.02 ± 1.3	0.062
TPHV, yrs	−0.66 ± 2.1	−0.86 ± 1.7	0.809
WC, cm	69.8 ± 8.8	67.3 ± 7.2	0.478
WHR	0.44 ± 0.05	0.45 ± 0.06	0.750
BMI, kg/m^2	19.5 ± 3.7	18.4 ± 3.2	0.474
BMI percentile	57 ± 31	38 ± 33	0.178
Resting HR, bpm	68 ± 10	74 ± 13	0.278
Resting systolic BP, mmHg	113 ± 8	106 ± 12	0.164
Resting diastolic BP, mmHg	65 ± 5	62 ± 6	0.169
Resting MAP, mmHg	84 ± 3	81 ± 6	0.152

Values are represented as means ± SD. APHV: age at peak height velocity; TPHV: time to peak height velocity; WC: waist circumference; WHR: waist-to-height ratio; BMI: body mass index; HR: heart rate; BPM: beats per minute; BP: blood pressure; MAP: mean arterial pressure.

4. Discussion

Over time, decreased levels of PA are generally associated with impairments of vascular function and structure and increased cardiovascular risk. This becomes particularly important when PA levels are limited in children and adolescents with a physical disability, such as cerebral palsy. Thus, early vascular assessments in this at-risk population may assist in determining potential CV risk factors. In this study we purposefully studied vascular health in the most functional adolescents with CP to contrast their PA levels and vascular health with their healthy peers. The primary findings did not confirm our hypothesis that arterial function and structure in adolescents with CP (GMFCS level I-II) are different from a healthy control group despite individuals with CP spending significantly less time performing vigorous PA in comparison to their typically developing peers.

In this study, the primary risk factor (for future cardiovascular health) of interest was level of PA, as measured using the Exercise Questionnaire [38]. Both groups spent similar amounts of time performing light-to-moderate PA; however, the CP group spent a significantly less amount of time engaging in vigorous intensity PA. Despite this discrepancy in time spent in high intensity PA, no group differences were seen in any of the measured indices of vascular health. It has been suggested that the strongest relationships between exercise interventions (comparable to levels of PA) and enhanced endothelial function exist in groups with relatively impaired FMD a priori. The tightest correlations between PA and

Table 2: Group comparisons of brachial vascular dimensions and FMD response.

	Control ($n = 10$)	CP ($n = 10$)	P value
Preocclusion diameter, mm	3.20 ± 0.37	3.08 ± 0.48	0.803
RH peak diameter, mm	3.40 ± 0.39	3.48 ± 0.38	0.815
Absolute FMD, mm	0.19 ± 0.11	0.33 ± 0.21	0.075
Relative FMD (%FMD)	6.1 ± 3.6	11.1 ± 7.8	0.080
Normalized (%FMD/SR_{AUC})	0.0027 ± 0.0015	0.0046 ± 0.0033	0.126
Mean SR	530 ± 250	544 ± 198	0.788
Time to peak, s	110 ± 45	102 ± 46	0.826
PP, mmHg	48 ± 10	45 ± 13	0.523

Values are represented as means ± SD. RH: reactive hyperemia; FMD: flow mediated dilation; SR: shear rate; SR_{AUC}: shear rate area under the curve.
Note. $n = 11$ in both groups for baseline brachial diameter.

Table 3: Group comparisons of PTT, PWV, carotid vascular dimensions, and carotid distensibility.

	Control ($n = 10$)	CP ($n = 11$)	P value
Central PTT	0.103 ± 0.032	0.089 ± 0.013	0.454
cPWV (m/s)	4.1 ± 0.9	4.3 ± 0.6	0.977
Peripheral PTT	0.108 ± 0.012	0.111 ± 0.028	0.768
pPWV (m/s)	7.6 ± 1.1	7.1 ± 1.7	0.450
Baseline diameter, mm	5.73 ± 0.29	5.63 ± 0.74	0.690
$_{c}$IMT, mm	0.41 ± 0.03	0.42 ± 0.04	0.576
Wall/lumen ratio	0.072 ± 0.007	0.077 ± 0.012	0.832
Distensibility, $mmHg^{-1}$	0.008 ± 0.002	0.008 ± 0.002	0.474
Compliance, $mm^2/mmHg$	0.19 ± 0.03	0.17 ± 0.06	0.376
PP (mmHg)	36 ± 10	42 ± 11	0.208

Values are represented as means ± SD. PTT: pulse transit time; PWV: pulse wave velocity, distensibility and compliance: Control, $n = 10$; IMT: intima-media thickness and wall/lumen ratio: CP, $n = 10$; PP: pulse pressure.

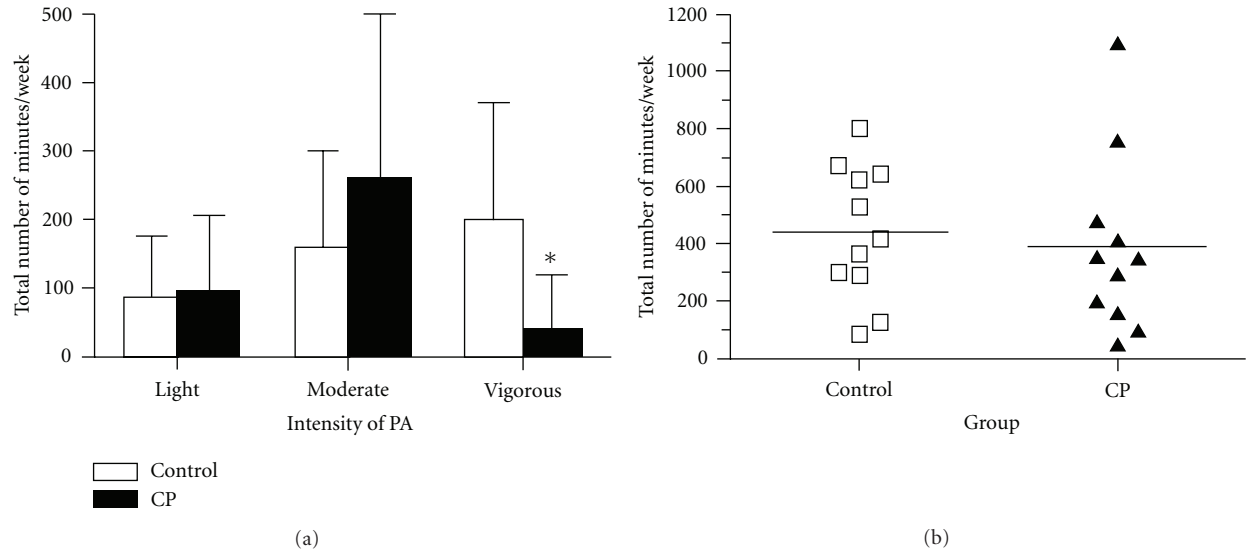

Figure 1: (a) Group comparisons of weekly PA according to intensity. (b) Group comparisons of total PA (summation of all three intensities) performed in one week, individual data and group means presented; $n = 11$ in each group.

FMD response have been shown to exist in the lowest tertiles of endothelial function [40]. Considering this, there is no reason to believe that the control group has experienced vascular dysfunction, which would predispose them to a positive vascular adaptation as a result of their higher levels of vigorous activity in comparison to the seemingly healthy CP group.

No significant differences between groups were found in cPWV or pPWV. These values were comparable to a previous study assessing PWV in a slightly younger group of healthy children (10.1 ± 0.3 yrs) who showed very similar cPWV values (4.2 ± 0.4 m/s) [14] to those in both groups in the current study (Table 3). This indicates preserved arterial stiffness at this time point for both the control and CP group. Similarities in PWV between groups in this study may be reflective of similar levels of low intensity PA, as indicated by the same amount of time spent in light and moderate intensity PA as well as the same total time spent performing PA per week (Figure 1(b)).

No differences were found between groups in either carotid distensibility or $_{c}$IMT. Throughout the lifespan, habitual PA has been shown to positively influence arterial distensibility [14, 18]. Age-related decreases in arterial distensibility and increases in stiffness have been reported [18]; however, increased levels of PA have been suggested to delay the age-dependent loss of arterial distensibility, in proportion to the amount and/or intensity of exercise [18, 41, 42]. Although there was no difference in distensibility between the CP and control group at this time, sufficient rationale is provided for this clinical group of adolescents to increase their levels of high intensity PA at an early stage and maintain these behaviours into adulthood in an attempt to mitigate these normative age-related changes.

$_{c}$IMT measurements were also similar between groups and were comparable to other control groups used in previous studies [43, 44]. Iannuzzi and colleagues (2004) [43] characterized the differences in $_{c}$IMT between obese children and age-matched control subjects (6–14 years) and showed a significantly greater IMT in the obese group in comparison to the healthy controls (0.55 ± 0.08 mm versus 0.49 ± 0.09 mm). The $_{c}$IMT of the obese children in the aforementioned study was approximately 24% and 25% greater than the $_{c}$IMT of the present study's CP and control group, suggesting healthy vascular structure in both groups in the current study.

In a previous study assessing the relationship between habitual PA (as measured using the double labeled water approach) and brachial FMD in 5–10-year-old children, a significant correlation was found ($r = 0.39$, $P = 0.007$), highlighting PA as the most influential variable in predicting the FMD response [9]. This group reported that physical fitness, as assessed using an incremental discontinuous treadmill-based exercise test, and levels of PA, as measured using Actigraph accelerometers, were lowest in the lowest %FMD and %FMD/SR_{AUC} tertile. These relationships between fitness, PA, and FMD response were significant, and it was concluded that PA measurements were the best predictors of endothelial (dys) function in this young group [40]. These data support the concept that PA exerts its protective effect on CV health via the endothelium and draws attention to the role of lifestyle modifications, specifically increases in levels of habitual PA in pediatric practice.

This cross-sectional study is the first to characterize indices of vascular health in higher functioning youth with CP and to make comparisons to a group of their typically developing peers. Children harbouring classic CV risk factors, including physical inactivity have been shown to exhibit impairments in vascular function and structure early in life and have an increased risk of premature atherosclerosis in adulthood [44]. It has been shown that levels of both PA and inactivity track significantly from adolescence (9 to 18 yrs) to young adulthood placing inactive children at an increased risk of becoming physically sedentary adults [45]. With evidence of physical inactivity being a significant precursor to CVD-related death and moderate levels of fitness providing protective effects against the influence of traditional risk factors on mortality [46], the value of well-established, healthy patterns of habitual PA in pediatric practice must not be overlooked. In a group of youth that may have increased susceptibility to physical inactivity, identifying any early alterations in vascular function and structure may assist in identifying preclinical vascular disease, allowing for intervention at the earliest stage possible.

5. Limitations

The FMD methodology used in the current study is relatively straightforward and noninvasive. However, limitations to the procedure are present. It is possible that during the FMD assessment peak dilation was underestimated as images were taken every 15 sec. for three heart cycles and not continuously for 3 min. following cuff release. This is a limitation of the storing capabilities of the equipment used; thus we chose to collect diameter data at fifteen-second intervals to attempt to represent the complete diameter profile following cuff release. The current results are limited to highly functioning, ambulatory individuals with CP and their typically developing peers. It is difficult to say if these results are applicable to prepubertal or postpubertal individuals as it can be assumed a mixed sample was represented. In this investigation, we did not control for or assess diet, vitamin ingestion or blood-borne CVD markers and therefore we cannot account for the contribution of these factors in any changes in vascular function.

One possible explanation for our finding of similar vascular structure and function despite differences in the amount of vigorous PA is that light-to-moderate PA is the main determinant of vascular health and that vigorous exercise is not necessary to maintain normal vascular structure and function in youth. It is also possible that the method used in the current study to assess PA (CP specific questionnaire) was not sensitive enough to determine absolute differences at all intensities or that confounding factors might result in a relative underestimation of the vigorous component of exercise for youth with CP. Future studies, which include more direct measurement of activity levels, may delineate the relationship between absolute activity levels and arterial health.

6. Conclusion

Although no differences in vascular structure or function between youth (9–16 years) with CP and typically developing peers were observed in the current study, the establishment of techniques to assess arterial health in youth with CP is critically important for determining future CV risk in this clinical population. This study confirms the feasibility of the use of these vascular assessment techniques in this population and presents potential for future, longitudinal assessments of individuals with CP across all levels of GMFCS classification. Each measurement of cardiovascular health was well tolerated and widely accepted by both participants and their parent/guardian(s). The consequences of significantly decreased amounts of time spent in vigorous PA for youth with CP, at this time and potentially into adulthood, remain unknown. For future research it is of interest to assess whether vessel health is compromised in youth and adults with more pronounced decreased levels of PA such as those in GMFCS levels III–V. Identifying these parameters may act as a tool for risk stratification in this population, thereby permitting identification of children who would benefit most from intensified PA and/or exercise interventions.

Acknowledgments

This paper is based on a master thesis in kinesiology at McMaster University written by the first author and funded by NSERC. This study was done as part of the Stay-FIT study that is funded by the Ontario Federation for Cerebral Palsy. Special thanks are due to the participants and their families for their valuable time and effort with this study.

References

[1] P. Rosenbaum, N. Paneth, A. Leviton et al., "A report: the definition and classification of cerebral palsy," *Developmental Medicine and Child Neurology*, vol. 109, pp. 8–14, 2007.

[2] P. L. Rosenbaum, S. D. Walter, S. E. Hanna et al., "Prognosis for gross motor function in cerebral palsy: creation of motor development curves," *Journal of the American Medical Association*, vol. 288, no. 11, pp. 1357–1363, 2002.

[3] J. W. Gorter, "Rehabilitative therapies for the child with cerebral palsy: focus on family, function and fitness," *Minerva Pediatrica*, vol. 61, no. 4, pp. 425–440, 2009.

[4] K. F. Bjornson, B. Belza, D. Kartin, R. Logsdon, and J. F. McLaughlin, "Ambulatory physical activity performance in youth with cerebral palsy and youth who are developing typically," *Physical Therapy*, vol. 87, no. 3, pp. 248–257, 2007.

[5] C. A. Maher, M. T. Williams, T. Olds, and A. E. Lane, "Physical and sedentary activity in adolescents with cerebral palsy," *Developmental Medicine and Child Neurology*, vol. 49, no. 6, pp. 450–457, 2007.

[6] R. Schenker, W. Coster, and S. Parush, "Participation and activity performance of students with cerebral palsy within the school environment," *Disability and Rehabilitation*, vol. 27, no. 10, pp. 539–552, 2005.

[7] M. J. LaMonte, S. N. Blair, and T. S. Church, "Physical activity and diabetes prevention," *Journal of Applied Physiology*, vol. 99, no. 3, pp. 1205–1213, 2005.

[8] S. N. Blair, H. W. Kohl, C. E. Barlow, R. S. Paffenbarger, L. W. Gibbons, and C. A. Macera, "Changes in physical fitness and all-cause mortality: a prospective study of healthy and unhealthy men," *Journal of the American Medical Association*, vol. 273, no. 14, pp. 1093–1098, 1995.

[9] R. A. Abbott, M. A. Harkness, and P. S. W. Davies, "Correlation of habitual physical activity levels with flow-mediated dilation of the brachial artery in 5-10 year old children," *Atherosclerosis*, vol. 160, no. 1, pp. 233–239, 2002.

[10] K. Pahkala, O. J. Heinonen, H. Lagström et al., "Vascular endothelial function and leisure-time physical activity in adolescents," *Circulation*, vol. 118, no. 23, pp. 2353–2359, 2008.

[11] H. C. Stary, "Evolution and progression of atherosclerotic lesions in coronary arteries of children and young adults," *Arteriosclerosis*, vol. 9, supplement 1, pp. I19–I32, 1989.

[12] J. A. Vita and J. F. Keaney, "Endothelial function: a barometer for cardiovascular risk?" *Circulation*, vol. 106, no. 6, pp. 640–642, 2002.

[13] M. F. O'Rourke, J. A. Staessen, C. Vlachopoulos, D. Duprez, and G. E. Plante, "Clinical applications of arterial stiffness; definitions and reference values," *American Journal of Hypertension*, vol. 15, no. 5, pp. 426–444, 2002.

[14] S. Sakuragi, K. Abhayaratna, K. J. Gravenmaker et al., "Influence of adiposity and physical activity on arterial stiffness in healthy children the lifestyle of our kids study," *Hypertension*, vol. 53, no. 4, pp. 611–616, 2009.

[15] D. H. O'Leary, J. F. Polak, R. A. Kronmal, T. A. Manolio, G. L. Burke, and S. K. Wolfson, "Carotid-artery intima and media thickness as a risk factor for myocardial infarction and stroke in older adults," *New England Journal of Medicine*, vol. 340, no. 1, pp. 14–22, 1999.

[16] O. T. Raitakari, M. Juonala, M. Kähönen et al., "Cardiovascular risk factors in childhood and carotid artery intima-media thickness in adulthood: the cardiovascular risk in young finns study," *Journal of the American Medical Association*, vol. 290, no. 17, pp. 2277–2283, 2003.

[17] D. R. Seals, C. A. DeSouza, A. J. Donato, and H. Tanaka, "Habitual exercise and arterial aging," *Journal of Applied Physiology*, vol. 105, no. 4, pp. 1323–1332, 2008.

[18] J. W. Gorter, J. Slaman, D. Bartlett, and H. J. G. Van den Berg-Emons, "Reliability of the gross motor function classification system expanded and revised (GMFCS-ER) when used with adolescents and young adults with cerebral palsy," *Developmental Medicine & Child Neurology*, vol. 53, supplement 5, pp. 42–43, 2011.

[19] M. Rakobowchuk, M. I. Stuckey, P. J. Millar, L. Gurr, and M. J. MacDonald, "Effect of acute sprint interval exercise on central and peripheral artery distensibility in young healthy males," *European Journal of Applied Physiology*, vol. 105, no. 5, pp. 787–795, 2009.

[20] M. C. J. Rudolf, J. Walker, and T. J. Cole, "What is the best way to measure waist circumference?" *International Journal of Pediatric Obesity*, vol. 2, no. 1, pp. 58–61, 2007.

[21] D. A. Bailey, "The Saskatchewan pediatric bone mineral accrual study: bone mineral acquisition during the growing years," *International Journal of Sports Medicine*, vol. 18, supplement 3, pp. S191–S194, 1997.

[22] L. B. Sherar, R. L. Mirwald, A. D. G. Baxter-Jones, and M. Thomis, "Prediction of adult height using maturity-based cumulative height velocity curves," *Journal of Pediatrics*, vol. 147, no. 4, pp. 508–514, 2005.

[23] W. Ross and M. Marfell-Jones, "Kinanthropometry," in *Physiological Testing of the High-Performance Athlete*, pp. 223–308, Human Kinetics Books, Champaign, Ill, USA, 1991.

[24] R. L. Mirwald, A. D. G. Baxter-Jones, D. A. Bailey, and G. P. Beunen, "An assessment of maturity from anthropometric measurements," *Medicine and Science in Sports and Exercise*, vol. 34, no. 4, pp. 689–694, 2002.

[25] R. A. Harris, S. K. Nishiyama, D. W. Wray, and R. S. Richardson, "Ultrasound assessment of flow-mediated dilation," *Hypertension*, vol. 55, no. 5, pp. 1075–1085, 2010.

[26] T. G. Pickering, J. E. Hall, L. J. Appel et al., "Recommendations for blood pressure measurement in humans: an AHA scientific statement from the Council on High Blood Pressure Research Professional and Public Education Subcommittee.," *Journal of Clinical Hypertension*, vol. 7, no. 2, pp. 102–109, 2005.

[27] R. Munakata, H. Katakai, T. Ueki, J. Kurosaka, K. I. Takao, and K. I. Tadano, "Total synthesis of (+)-macquarimicin A," *Journal of the American Chemical Society*, vol. 125, no. 48, pp. 14722–14723, 2003.

[28] W. W. Nichols and M. F. O'Rourke, *McDonald's Blood Flow in Arteries: Theoretical, Experimental and Clinical Principles*, A Hodder Arnold Publication, 5th edition, 2005.

[29] T. Weber, M. Ammer, M. Rammer et al., "Noninvasive determination of carotid-femoral pulse wave velocity depends critically on assessment of travel distance: a comparison with invasive measurement," *Journal of Hypertension*, vol. 27, no. 8, pp. 1624–1630, 2009.

[30] K. D. Currie, N. A. Proudfoot, B. W. Timmons, and M. J. MacDonald, "Noninvasive measures of vascular health are reliable in preschool-aged children," *Applied Physiology, Nutrition and Metabolism*, vol. 35, no. 4, pp. 512–517, 2010.

[31] R. Kelly and D. Fitchett, "Noninvasive determination of aortic input impedance and external left ventricular power output: a validation and repeatability study of a new technique," *Journal of the American College of Cardiology*, vol. 20, no. 4, pp. 952–963, 1992.

[32] B. Fernhall and S. Agiovlasitis, "Arterial function in youth: window into cardiovascular risk," *Journal of Applied Physiology*, vol. 105, no. 1, pp. 325–333, 2008.

[33] A. E. Donald, M. Charakida, T. J. Cole et al., "Non-invasive assessment of endothelial function. Which technique?" *Journal of the American College of Cardiology*, vol. 48, no. 9, pp. 1846–1850, 2006.

[34] G. Atkinson, A. M. Batterham, M. A. Black et al., "Is the ratio of flow-mediated dilation and shear rate a statistically sound approach to normalization in cross-sectional studies on endothelial function?" *Journal of Applied Physiology*, vol. 107, no. 6, pp. 1893–1899, 2009.

[35] M. C. Corretti, T. J. Anderson, E. J. Benjamin et al., "Guidelines for the ultrasound assessment of endothelial-dependent flow-mediated vasodilation of the brachial artery: a report of the international brachial artery reactivity task force," *Journal of the American College of Cardiology*, vol. 39, no. 2, pp. 257–265, 2002.

[36] T. J. Anderson, A. Uehata, M. D. Gerhard et al., "Close relation of endothelial function in the human coronary and peripheral circulations," *Journal of the American College of Cardiology*, vol. 26, no. 5, pp. 1235–1241, 1995.

[37] B. A. Parker, T. L. Trehearn, and J. R. Meendering, "Pick your Poiseuille: normalizing the shear stimulus in studies of flow-mediated dilation," *Journal of Applied Physiology*, vol. 107, no. 4, pp. 1357–1359, 2009.

[38] L. K. Brunton and D. J. Bartlett, "Description of exercise participation of adolescents with cerebral palsy across a 4-year period," *Pediatric Physical Therapy*, vol. 22, no. 2, pp. 180–187, 2010.

[39] A. T. Weston, R. Petosa, and R. R. Pate, "Validation of an instrument for measurement of physical activity in youth," *Medicine and Science in Sports and Exercise*, vol. 29, no. 1, pp. 138–143, 1997.

[40] N. D. Hopkins, G. Stratton, T. M. Tinken et al., "Relationships between measures of fitness, physical activity, body composition and vascular function in children," *Atherosclerosis*, vol. 204, no. 1, pp. 244–249, 2009.

[41] T. Kakiyama, M. Matsuda, and S. Koseki, "Effect of physical activity on the distensibility of the aortic wall in healthy males," *Angiology*, vol. 49, no. 9, pp. 749–757, 1998.

[42] D. R. Seals, A. E. Walker, G. L. Pierce, and L. A. Lesniewski, "Habitual exercise and vascular ageing," *Journal of Physiology*, vol. 587, no. 23, pp. 5541–5549, 2009.

[43] A. Iannuzzi, M. R. Licenziati, C. Acampora et al., "Increased carotid intima-media thickness and stiffness in obese children," *Diabetes Care*, vol. 27, no. 10, pp. 2506–2508, 2004.

[44] B. Hacihamdioğlu, V. Okutan, Y. Yozgat et al., "Abdominal obesity is an independent risk factor for increased carotid intima- media thickness in obese children," *Turkish Journal of Pediatrics*, vol. 53, no. 1, pp. 48–54, 2011.

[45] M. Juonala, J. S. A. Viikari, M. Kähönen et al., "Life-time risk factors and progression of carotid atherosclerosis in young adults: the Cardiovascular Risk in Young Finns study," *European Heart Journal*, vol. 31, no. 14, pp. 1745–1751, 2010.

[46] R. Telama, X. Yang, J. Viikari, I. Välimäki, O. Wanne, and O. Raitakari, "Physical activity from childhood to adulthood: a 21-year tracking study," *American Journal of Preventive Medicine*, vol. 28, no. 3, pp. 267–273, 2005.

Hindawi Publishing Corporation
International Journal of Pediatrics
Volume 2012, Article ID 504387, 11 pages
doi:10.1155/2012/504387

Research Article

Evaluation of Functional Electrical Stimulation to Assist Cycling in Four Adolescents with Spastic Cerebral Palsy

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Received 2 November 2011; Revised 12 January 2012; Accepted 13 February 2012

Academic Editor: Maria A. Fragala-Pinkham

Introduction. Adolescents with cerebral palsy (CP) often have difficulty participating in exercise at intensities necessary to improve cardiovascular fitness. Functional electrical stimulation- (FES-) assisted cycling is proposed as a form of exercise for adolescents with CP. The aims of this paper were to adapt methods and assess the feasibility of applying FES cycling technology in adolescents with CP, determine methods of performing cycling tests in adolescents with CP, and evaluate the immediate effects of FES assistance on cycling performance. *Materials/Methods*. Four participants (12–14 years old; GMFCS levels III-IV) participated in a case-based pilot study of FES-assisted cycling in which bilateral quadriceps muscles were activated using surface electrodes. Cycling cadence, power output, and heart rate were collected. *Results*. FES-assisted cycling was well tolerated ($n = 4$) and cases are presented demonstrating increased cadence (2–43 rpm), power output (19–70%), and heart rates (4-5%) and decreased variability (8–13%) in cycling performance when FES was applied, compared to volitional cycling without FES assistance. Some participants ($n = 2$) required the use of an auxiliary hub motor for assistance. *Conclusions*. FES-assisted cycling is feasible for individuals with CP and may lead to *immediate* improvements in cycling performance. Future work will examine the potential for long-term fitness gains using this intervention.

1. Introduction

Cerebral Palsy (CP) is a nonprogressive disorder that results from a disturbance in the fetal or infant brain [1, 2]. This disturbance, although varied in etiology, results in motor impairments in the developing child [3–6]. Individuals with CP have muscle weakness and abnormally high muscle spasticity in the affected extremities, which result in fine and gross motor developmental delays [2, 4, 5, 7]. Poor selective muscle control often results in coactivation of agonist and antagonist muscle groups [5]. Spasticity and abnormal tone that is present in the muscles of children with CP [7, 8] can cause abnormal forces at the joints, which can lead to bony deformity, joint instability, and muscle contractures as the child grows [7, 9–11]. The weakness that affects these muscles results in balance impairments and poor selective motor control which may lead to diminished independence and a lack of physical activity [12, 13]. Although CP is a nonprogressive injury of the brain, the impairments and functional limitations associated with CP can change over time, with many children becoming less independent with functional mobility as they enter their teenage years [14, 15]. Traditional means of exercise, such as running, jumping, and playing organized sports, may be difficult for individuals with such functional limitations and, unfortunately, many individuals with disabilities participate in less physical activity than people without disabilities [16–19].

Stationary recumbent cycling has been proposed as a feasible method to exercise in this population [20–29] because cycling does not require the dynamic balance that exercise

in a standing position requires. Cycling provides a potential means of improving strength, cardiovascular function, and range of motion, while exercising in a safe position and performing an activity that most children enjoy [26, 27, 29, 30]. Unfortunately, children with CP may not have the strength or coordination to cycle at the power intensities or sustained intervals required to achieve cardiovascular benefits from exercise [21, 31]. Many children with CP have difficulty with the motor performance of the cycling task because of unsmooth, asymmetrical cycling resulting from uncoordinated pushing and pulling on the pedals rather than cycling in a continuous manner [20, 21, 28]. Further difficulties include agonist/antagonist muscle coactivation, poor gross mechanical efficiency with the cycling task, and difficulties attaining threshold heart rates and cycling intensities necessary to achieve cardiorespiratory training effects and musculoskeletal changes [31]. Thus, additional means may be necessary to improve cycling ability for fitness attainment in children with CP.

Functional electrical stimulation (FES) has been used to facilitate cycling to improve cardiorespiratory fitness and to cause musculoskeletal gains in individuals with complete paralysis due to spinal cord injuries (SCI) [32–38]. In addition to cardiorespiratory and musculoskeletal gains, Krause et al. found that FES cycling may also moderate the excessive muscle tone that is present in individuals with SCI [39]. Children with spastic CP have lower levels of cardiovascular fitness [12], muscle weakness [3–5, 7, 8, 40], and elevated muscle tone [7, 11] that also could potentially respond to a FES cycling intervention [24, 41, 42]. The application of FES in individuals with CP, however, is fundamentally a different task than the application of FES in individuals with complete SCI because individuals with CP have varying degrees of volitional ability to pedal a cycle and additionally have sensate lower extremities. Preliminary work in our laboratory [24, 25] examined the development of a FES system for assisting and evaluating cycling in individuals with CP. Two recent publications have also reported the application of FES in individuals with CP [41, 42]. The first publication was a case study featuring training with FES cycling in an adult with CP [41]. The second publication reported on the use of FES in children with CP [42]; however, unlike in the current project, the participants were asked to passively allow FES to propel the crank rather than using volitional effort to contribute to the cycling task. The goal of FES assistance in the present study is to increase the cadence and power output that can be produced volitionally during cycling such that adolescents with CP can reach the heart rate thresholds necessary to gain cardiovascular benefits from exercise. There is also the potential that increasing cardiovascular fitness and strength may lead to improved function and quality of life. The aims of this study were to (1) adapt methods and assess the feasibility of applying FES cycling technology in adolescents with CP, (2) determine methods of performing cycling tests in adolescents with CP of GMFCS levels III-IV, and (3) Evaluate the *immediate* effects of FES assistance on cycling performance (i.e., cycling cadence and power output, coefficient of variance of cycling performance measures, and cardiovascular responses). Preliminary results have been presented elsewhere [25].

2. Materials and Methods

2.1. Participants. Participants were recruited from the Cerebral Palsy Clinic at Shriners Hospital for Children, Philadelphia, or through referral from community physical therapists. Adolescents with CP of Gross Motor Function Classification System (GMFCS) levels III-IV were recruited for this study [43]. Participants of GMFCS levels III-IV were targeted because these individuals have the physical capacity to learn how to cycle but they are often limited in their physical activity opportunities due to their lower level of independence with physical mobility. Individuals of GMFCS level III are able to ambulate with the use of an assistive device but may use a wheelchair for long distances. Individuals of GMFCS level IV have limited self-mobility and may require power mobility in the community. The participant inclusion and exclusion criteria are summarized in Table 1.

All participants were screened by an orthopedic surgeon to ensure that they were not at risk for hip dislocation and by a physical therapist to verify that they had sufficient passive range of motion to complete a revolution of the crank comfortably. The informed consent document, including all accompanying procedures and risks, was discussed with each participant and his/her parent or guardian. Sufficient time was provided for review of the document and to answer any questions. Informed consent and assent documents, approved by the governing Institutional Review Board, were signed by each participant and their parent or guardian.

A sample of convenience consisting of four participants (2 male) with spastic CP between the ages of 12–14 years (mean 13 ± 1.2 years) participated in this case-based pilot study (demographic information is provided in Table 2). Participant 1 had previous cycling experience in the community using an upright tricycle and had previously used neuromuscular electrical stimulation in an isometric strengthening protocol. Cycling and the application of surface electrical stimulation were novel to participants 2, 3, and 4.

2.2. Equipment. A previously described custom recumbent tricycle-based FES system was used for all testing sessions [24]. This tricycle-based system (a sport tricycle (KMXKarts; United Kingdom) mounted on a cycle trainer (Tacx; Wassenaar, The Netherlands) to allow for stationary cycling) was instrumented with a torque sensor and shaft encoder to allow for collection of torque, crank position and cadence, and consequently the calculation of power output during the cycling session (Figure 1). The tricycle-based system also allows for optional, direct drive pedaling assistance by an auxiliary hub motor directly coupled to the rear wheel's fixed gear. The use of this motor allows for controlled mechanical propulsion assistance in addition to the muscle contraction assistance provided by FES. Data were collected using custom software (MatLab, The Mathworks, Inc). The seat position, crank arm length, bottom bracket, and foot pedal position were adjusted to accommodate the leg length of each

Table 1: Inclusion and exclusion criteria for participation.

Inclusion	Exclusion
(i) 10–18 years of age	(i) Diagnosis of athetoid or ataxic CP
(ii) Diagnosis of spastic diplegic or quadriplegic CP	(ii) Significant scoliosis with primary curve >40 degrees
(iii) GMFCS level III (walks with assistive device; may use a wheelchair for long distances) or IV (self-mobility with limitations; transported or uses power mobility in the community)	(iii) Spinal fusion extending into the pelvis
(iv) Sufficient covering of the femoral head in the acetabulum (MIGR% < 40%)	(iv) Severe tactile hypersensitivity
(v) Adequate range of motion of the hips, knees, and ankles to allow pedaling	(v) Joint instability or dislocation in the lower extremities
(vi) Sufficient visuoperceptual skills and cognition/communication skills to participate in cycling trials	(vi) Surgery, traumatic or stress fractures in the last year
(vii) Seizure-free or well-controlled seizures	(vii) Botulinum toxin injections in the LE muscles in the past 6 months
(viii) Willingness to participate in testing at Shriners Hospital for Children, Philadelphia	(viii) Severe spasticity of the leg muscles (e.g., a score of >4 on the Modified Ashworth Scale)
(ix) Ability to communicate pain or discomfort with testing procedures	(ix) Joint pain or discomfort during cycling
(x) Ability to obtain parent/guardian consent and child assent/consent	(x) Severely limited range of motion/irreversible muscle contractures that prevent the subject from being able to be safely positioned on the cycling
	(xi) History of pulmonary disease limiting exercise tolerance (Asthma Control Test screen) or history of known cardiac disease (American Heart Association Screen)
	(xii) Pregnancy

Table 2: Participant description and FES parameters for cycling tests with FES.

Participant	Gender	Age (years)	Type of spastic CP	GMFCS level	Mode of community mobility
1	F	12	Diplegic	III	Anterior rolling walker
2	M	14	Diplegic	III	Posterior rolling walker
3	M	14	Quadriplegic	III	Posterior rolling walker; manual wheelchair at school
4	F	12	Quadriplegic	IV	Manual wheelchair

GMFCS refers to gross motor function classification scale level [43].

participant and any soft tissue contractures. In addition, shank guides previously used in our laboratory [22–24] were used to prevent excessive hip abduction and adduction during the cycling motion.

For this investigation, only the bilateral quadriceps femoris muscles were stimulated during the limb extension phase of the cycling crank rotation using a commercially-available stimulator with custom programming (Hasomed RehaStim, Magdeburg, Germany). Self-adhesive electrode sizes were selected for each individual to maximize the surface area over the quadriceps being activated in an effort to minimize stimulation current density and thereby maximize participant comfort. The appropriate electrode size for all participants was 7.5×10 cm (Axelgaard Manufacturing Co., Fall Brook, CA). The proximal electrode was positioned in an oblique orientation over the proximal rectus femoris and vastus lateralis heads of the quadriceps femoris and the distal electrode was positioned in an oblique orientation over the vastus medialis obliquus and distal vastus lateralis heads of the quadriceps femoris. Care was taken to ensure that electrodes were placed over an area of the skin free from blemishes or skin breakdown.

2.3. Stimulation Levels. The participants in this study completed training at a stimulation frequency of 33 Hz based on the parameters used in our laboratory for FES cycling in individuals with SCI [24, 35, 44]. A current intensity of 40 mA was well tolerated and allowed for easy facilitation of a fused quadriceps contraction using the electrical stimulation [24]. The level of stimulation applied was then varied by using a throttle (potentiometer) to increase or decrease the stimulus pulse width to control the strength of elicited muscle contractions. A custom program (MatLab, The MathWorks, Inc) controlled the on and off time for the stimulation applied to each leg based upon the position of the crank and the instantaneous cadence at which the

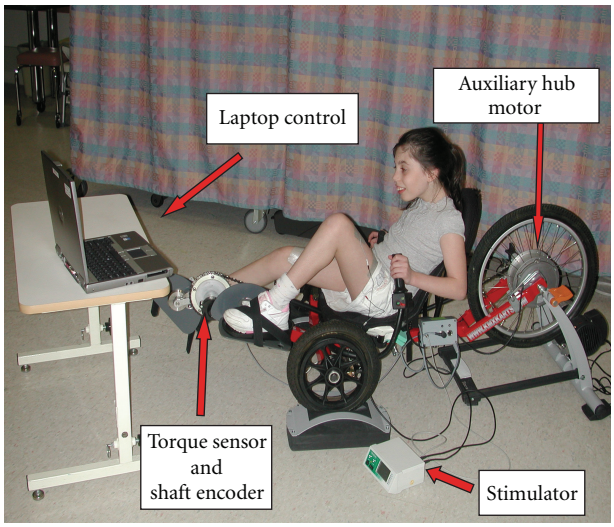


Figure 1: Tricycle components and participant set-up for FES-assisted cycling study [24]. The tricycle-based system is instrumented with a torque sensor and shaft encoder to allow for collection of torque, crank position and cadence, and consequentially the calculation of instantaneous power output, during the cycling session. The stimulator provides surface stimulation to bilateral quadriceps. The auxiliary hub motor was used for subjects 3 and 4 (please see text for details). A laptop computer is used for data acquisition, control of the stimulation timing, and control of the hub motor and to provide visual feedback on cycling performance to the cyclist.

participant was cycling [24]. Participants were instructed to tell investigators to turn down or turn off the stimulation at any time if they felt uncomfortable. Participants were also provided with a kill switch that could be pressed to terminate the stimulation at any time during the testing procedures.

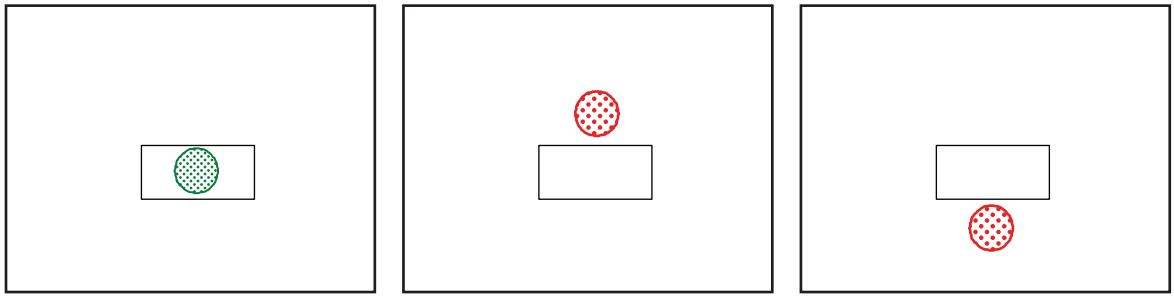
Motor level stimulation, defined as the level of stimulation required to cause a muscle contraction that moved the lower extremity through a pedaling arc of motion, was used for FES-assisted cycling trials. Participants were positioned on the bike with the leg being stimulated flexed at the knee and hip and the pedal positioned at the crank angle just prior to where active knee extension occurs. Participants were asked to relax their muscles and allow the electrical stimulation to move their legs. The stimulus pulse width was gradually increased until the limb moved through an arc of motion into extension. This procedure to determine the motor level response pulse width was then repeated until three successive trials were consistent (i.e., with motor level response pulse width values within 5% of each other). This procedure for determining motor level pulse width was then repeated for the opposite extremity. The motor level response pulse width for each individual was used for all FES-assisted cycling tests. Pulse width settings ranged from 90 to 200 μsec (160, 100, 90, and 200 μsec for participants 1, 2, 3, and 4, resp.).

2.4. Cycling Tests. To quantify the immediate effects of applying FES assistance, all participants completed cycling trials *with* and *without* the application of FES assistance. As part of the development of testing procedures, custom software was used to provide a simple feedback system for all participants. A laptop computer provided participants visual feedback of cycling performance with either power output or cadence targets to sustain (Figure 2).

Feedback targets were determined for each participant based on their cycling ability. Some individuals' cycling sessions were essential to the development of cycling performance testing procedures (e.g., use of hub motor to maintain a minimum cadence, discussed hereinafter), while the data from other individuals were used to determine the FES cycling techniques (e.g., application of alternating periods of stimulation on and off in participant 1). The specific tests and total number of tests each participant completed depended upon the stage of development of the project and aim being addressed when the participant participated and the number of times the participant was able to come to Shriners Hospital for testing (Table 3). Participants completed 2–4 sessions with at least 24 hours of rest between cycling sessions and with all testing occurring within a two-week period for each participant. Heart rate was recorded every 10 seconds, for the duration of the test, using a pulse oximeter.

2.4.1. Incremental Load Tests (Participants 1 and 4). The purpose of the incremental load test was to determine peak power output, cadence, and heart rate for each participant. Participants first completed a brief 30-second cycling trial to determine the appropriate power output or cadence increments. During the incremental load tests, the target was increased by equal increments at one-minute intervals. Tests lasted 8–12 minutes as recommended by traditional incremental exercise testing guidelines [45] and were sufficiently rigorous to determine the participant's peak power output.

2.4.2. Constant Load Tests (All Participants). The purpose of the constant load testing was to assess submaximal performance and determine the participant's ability to work at a steady-state level. For constant load trials, a single power output or cadence target was selected and the participant attempted to maintain the target level for the duration of the test. Constant load tests lasted 8–10 minutes and were sufficient in length for each participant to achieve steady-state cycling performance. For participants completing incremental testing prior to constant load testing, the target for the constant load test was set at 80% of the peak power output achieved during the incremental test. The target for the constant load test was based upon the method of Hunt et al. [46] who examined the energetics of FES cycling in individuals with paraplegia in which the work rate was based on the individual's maximal power on a prior test. Unpublished pilot work in our laboratory determined that exercising at 80% of the incremental load peak power output was sufficient to achieve steady-state oxygen uptake prior to lower extremity fatigue. For participants 2 and 3, who did not complete an incremental load test, the target for the constant load test was set at 80% of the peak power output achieved on the brief, maximal effort, 30-second cycling trial. At least 24



(a) (b) (c)

Figure 2: Visual feedback provided during cycling tests and training sessions. Participants are asked to cycle at a target power level or cadence which is represented by the white box on the screen. If the participant is successful, the ball stays within the box and turns green (a). If the participant cycles at a higher (b) or lower (c) power level or cadence, the ball moves out of the box and turns red.

Table 3: Overview of Tests Completed by Each Participant.

Participant	No motor constant load VOL	No motor constant load FES	No motor incremental load VOL	No motor incremental load FES	Motor constant load VOL	Motor constant load FES	Motor incremental load VOL	Other cycling trials completed
1			×	×				No motor constant load with FES alternating on:off in 1 minute increments
2	×	×						
3	×				×	×		
4					×	×	×	No motor-Brief trial without FES assistance

Participants are listed by row with "×" corresponding to each test a given subject completed. "No motor" refers to cycling tests without the use of the auxiliary hub motor to control cadence, while the hub motor was used in the "Motor" tests. VOL refers to tests without the use of FES assistance. FES refers to tests in which FES was applied. For the tests in which FES was applied, stimulation was applied at 33 Hz, 40 mA. Pulse width ranged from 90 to 200 μsec and corresponded to the participant-specific pulse width required to elicit a motor level contraction. Participants 1 and 2 used cadence as feedback on cycling performance, while participants 3 and 4 used power output for feedback due to requiring the use of the auxiliary hub motor (please see text for details).

hours of rest was provided between incremental and constant load cycling tests.

2.4.3. Auxiliary Hub Motor (Participants 3 and 4). Initial analysis of the first three participants determined that not all participants were able to complete a standard constant load or incremental test without the use of a motor to assist with propulsion. Participants 3 and 4 were unable to cycle consistently enough to maintain target cadences during constant load or incremental load tests. Consequently, the auxiliary hub motor located within the rear wheel of the cycle (Figure 1) was used during some of the testing for participants 3 and 4. The motor facilitated automatic control of cadence while simultaneously collecting power output data to provide information on cycling performance [24, 25]. Because of the poor cycling ability of these participants, the majority of the torque produced was resistive to forward motion of the crank which resulted in a negative net work rate. For these participants, in a period of passive cycling in which the participants allowed the motor to move their legs, negative power output data were collected. This represented how much the individual was resisting the passive movement of their legs and, in effect, how much work the motor had to do to overcome the weight and muscle tone in the participant's legs to turn the crank. After a period of passive cycling, the participants were asked to actively cycle and the difference between the passive phase and active phase power outputs was calculated to determine the net power output for the cyclist. Because cadence was controlled by the auxiliary hub motor for participants 3 and 4, power output targets were used for visual feedback during the cycling tests.

2.5. Data Analysis. Cadence and torque data were collected at 20 Hz using custom software (MatLab, The MathWorks, Inc) and used to calculate instantaneous power output. For participants completing constant load tests with and without FES assistance, paired t-tests were used to analyze differences between mean power output between tests with and without FES. Cycling performance was also analyzed by calculating the coefficient of variance of power output for each minute of the active cycling phase and then averaging across the total number of minutes to determine the coefficient of variance for the test. For participants completing the test using cadence as feedback (participants 1 and 2), the coefficient of variance of cadence was also calculated using the standard deviation of cadence and mean cadence in the calculation

instead of power output. Paired t-tests were performed to compare coefficient of variance between testing conditions with and without FES assistance.

Peak heart rate during incremental load testing was determined as the maximum of all heart rate data collected in a testing session. Peak heart rate was reported as a percentage above resting heart rate to account for slight within-participant variations in resting heart rate occurring between testing days. Average heart rate reflects an average of all heart rate data collected during the active cycling phase of testing within a single testing session.

3. Results

3.1. Application of FES Assistance. All participants were able to complete tests without difficulty and were able to tolerate the application of FES to bilateral quadriceps muscles. All participants were able to complete motor level stimulation thresholding and FES-assisted cycling trials without requiring additional acclimation to tolerate the stimulation. Additionally, although participants were provided with the kill switch, none of the participants chose to terminate the stimulation when FES was applied. The stimulus pulse width required to elicit a motor level response ranged from 90 to 200 μsec (see Section 2.3 for participant-specific pulse widths).

3.2. Cycling Performance. Participants 1 and 2 completed cycling tests *without* the use of the auxiliary hub motor to maintain a constant cadence. Cadence was used as a target for the constant load and incremental tests in these participants. Participant 1 completed two separate incremental tests (with and without FES assistance) and a constant load test in which FES assistance alternated between on and off at one-minute increments over the length of the test. Participant 2 completed constant load trials with and without FES assistance.

Participants 3 and 4 completed cycling tests *with* the auxiliary hub motor assistance. Power was used as the feedback target for the constant load and incremental tests for these participants. Baseline passive cycling data were collected to determine the amount of work the motor needed to perform to passively move the legs through a range of motion without the participant assisting with the task. Participant 3 completed three constant load tests: one *without* the use of the auxiliary hub motor and *without* FES assistance, one *with* the use of the auxiliary hub motor and *without* FES assistance, and one test *with* both the auxiliary hub motor and FES assistance. Participant 4 completed brief cycling trials with and without the use of the auxiliary hub motor and she completed three tests *with* the use of the auxiliary hub motor: an incremental test *without* FES assistance and constant load tests *with* and *without* FES assistance.

3.2.1. Incremental Load Test Results (Participants 1 and 4). Only participant 1 completed incremental load tests with and without FES assistance. During incremental cycling tests, this participant achieved higher peak cadence and heart rate values during the test with FES assistance (Figures 3(a) and 3(b)). The primary objective of the incremental test was to determine peak heart rate and power output values; however, a secondary analysis examined average heart rate and power output across the tests to determine the relative level of exertion at which the participant was working over the duration of the test (Figures 3(c) and 3(d)).

Cycling performance was also analyzed by calculating the coefficient of variance (averaged over each minute during the cycling trial) in power output and cadence. Participant 1 demonstrated a decrease in the variability in cadence (mean 16.3 ± 4.3% without FES assistance versus 7.8 ± 2.1% with FES assistance) and power output (mean of 28.8 ± 4.5% without FES assistance versus 16.5 ± 2.5% with FES assistance) in the cycling test with FES applied.

The incremental load test completed by participant 4 was performed as part of development of incremental load testing procedures using the auxiliary hub motor. The participant was able to complete the testing and appropriately respond to the increasing target on the computer screen. He did not complete a FES-assisted incremental load test that could be used for comparison.

3.2.2. Constant Load Test Results (All Participants). Participant 1 completed a constant load test in which FES assistance was alternated in one-minute increments of being on and off. The coefficient of variance for this participant's power output and cadence were calculated across each minute. Overall, the participant demonstrated an immediate decrease in the variability in cadence (mean 11.5 ± 4.1% without FES assistance versus 8 ± 2% with FES assistance) and variability of power output (mean 12.8 ± 2.6% without FES assistance versus 9.9 ± 2% with FES assistance) during the periods when FES assistance was applied (all P values > 0.3) (Figure 4). Although changes in variability were not statistically significant, it did appear that participant 1 cycled more smoothly with FES.

The application of FES assistance led to immediate changes in cycling performance for participant 2, who had no cycling experience and he was unable to complete a crank revolution without assistance. As described previously, Participant 2 completed constant load trials (full tests were unable to be completed due to poor cycling ability and this participant participating prior to the implementation of the auxiliary hub motor) with and without FES assistance. During volitional trials without FES assistance, his lower extremities would get stuck during his attempt to pedal forward and he would alternate between pedaling approximately 180 degrees forward and backward. The forward and backward motion of the crank resulted in no net power generation (Figure 5(a): prior to the application of FES assistance) and a cadence that fluctuated from positive to negative (Figure 5(b): prior to the application of FES assistance). Once FES assistance was applied, the participant was able to successfully pedal the cycle (Figure 5).

Participant 3 required the use of the hub motor to perform constant load cycling tests. During the volitional cycling test, the participant's power output was negative throughout

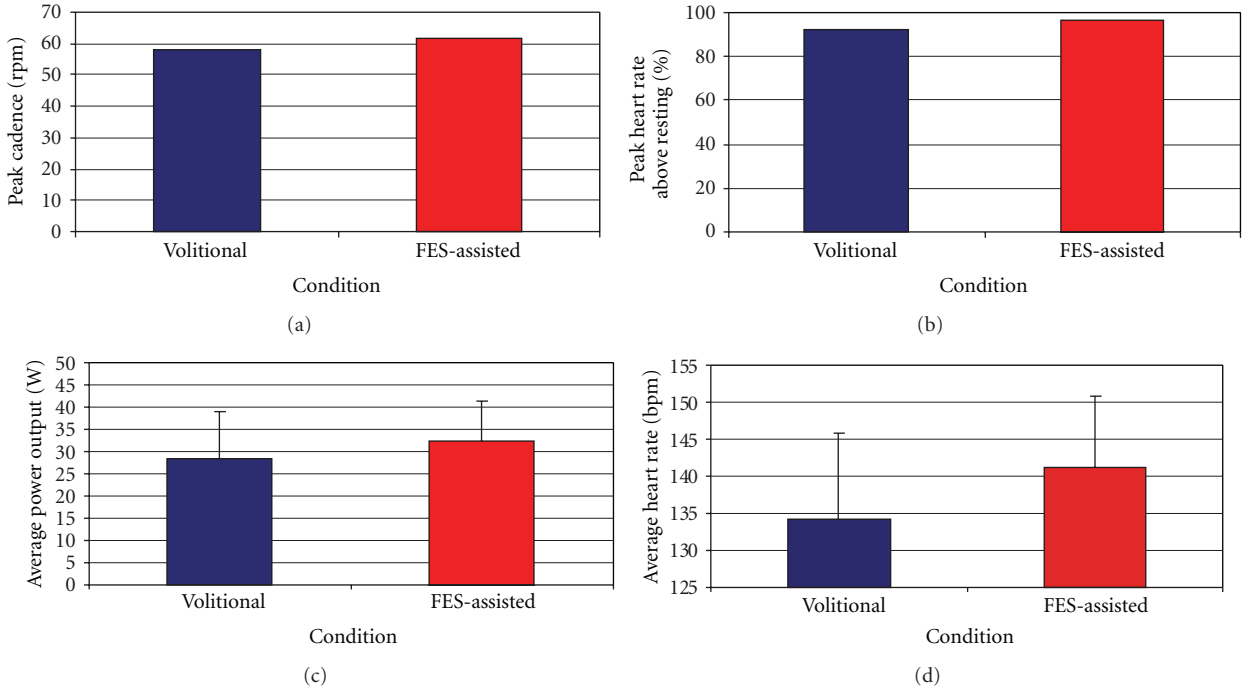


Figure 3: Cycling performance during incremental cycling tests with and without FES assistance in a child who is well adept at cycling (participant 1). The graphs illustrate peak cadence (a), peak heart rate as a percentage of resting heart rate (b), average power output (c), and average heart rate (d) during the tests. For average power output (c) and average heart rate (d) standard deviation values are shown. The blue bars represent volitional cycling and the red bars represent FES-Assisted cycling trials.

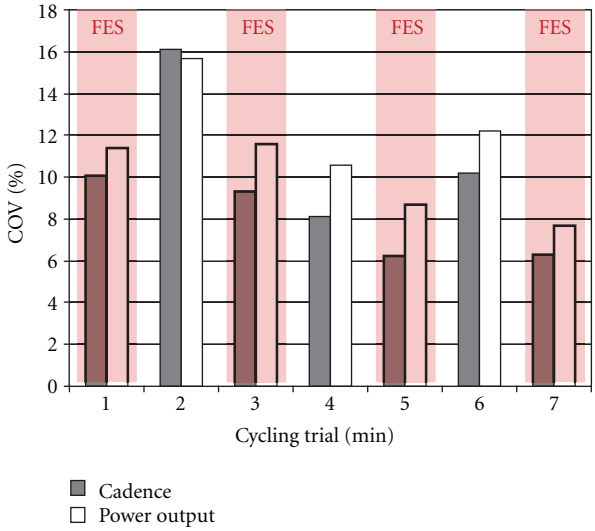


Figure 4: Coefficient of variance of cycling cadence and power output during a constant load cycling test in an individual with CP who is adept at cycling (Participant 1). The coefficient of variance for each variable was calculated over 1-minute periods in which FES assistance to the quadriceps muscles was either turned on (red shaded areas) or off (areas of the graph without red shading).

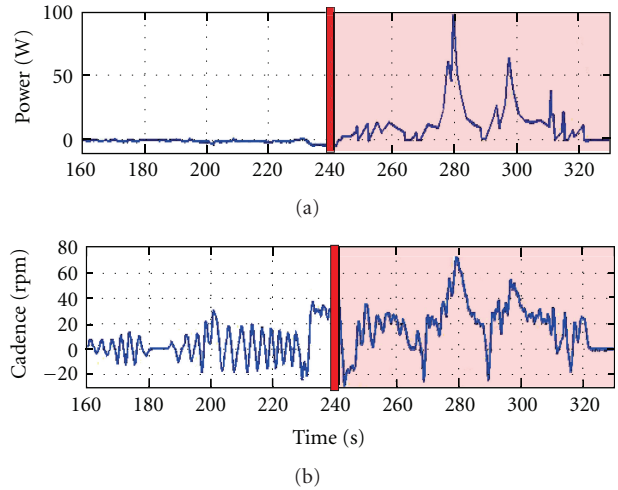


Figure 5: Cycling performance during the initial application of FES in a child for whom cycling was a novel task (participant 2). The top trace (a) illustrates his power output and the bottom trace (b) illustrates his cadence. The red vertical bars at 240 s indicate when FES assistance began and FES remained on during the red-shaded portion of the graph. Data were smoothed for analysis using a second-order lowpass Butterworth filter with a cutoff frequency of 0.1 Hz.

the majority of the cycling task, although positive power output was achieved during portions of the FES-assisted constant load test (Figure 6). Participant 4 also completed constant load trials with and without FES assistance; paired samples t-tests comparing average power output during two constant load cycling test conditions (with and without FES

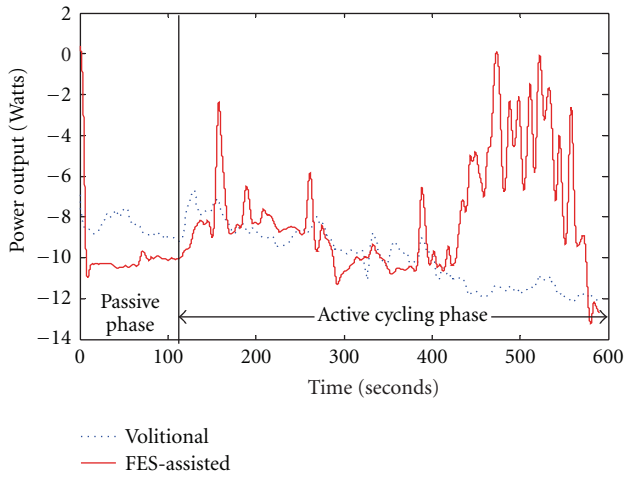


Figure 6: Power output during constant load cycling tests with (red line) and without (blue line) FES assistance. This data are from a child without cycling experience (participant 3) and the auxiliary hub motor was required for testing. The vertical black line at 120 seconds denotes the transition from a passive cycling phase (in which the motor moved the child's legs while he rested) to an active phase (in which the motor continued to control cadence, and the participant assisted with the cycling effort). In the FES assistance condition, the stimulation was applied only during the active cycling phase. Data were smoothed for analysis using a second-order lowpass Butterworth filter with a cutoff frequency of 0.1 Hz.

assistance) for participants 3 and 4 were not statistically significant ($P = 0.846$).

4. Discussion

FES-assisted cycling was well tolerated by the participants in this study and demonstrated the feasibility of applying FES assistance in children with CP. The use of an auxiliary motor to control cadence was necessary to allow for meaningful assessment of cycling performance in some participants. FES-assisted cycling resulted in increased cadence, power output, and heart rates and decreased variability in cycling performance compared with volitional cycling without FES assistance. Such improvements in cycling ability with the application of FES assistance may make cycling for fitness attainable for individuals with CP who have impaired cycling ability.

4.1. Application of FES Assistance. All participants were able to tolerate the application of FES and complete testing. Only one participant (participant 1) had previous experience with surface electrical stimulation while the other three did not. Unlike in individuals with complete SCI who are unable to volitionally contribute to the cycling task, our participants were able to cycle volitionally, although inefficiently, and were provided with visual feedback on the computer screen to encourage cycling at a target cadence or power output (Figure 2). The use of this feedback system may also have applications in studies involving children with incomplete SCI or poststroke because these individuals may have some ability to contribute to the cycling task [47]. In a recent study by Trevisi et al., participants with CP were asked to not contribute to the cycling effort and to allow the cycle to move their legs during the passive cycling and FES cycling phases [42]. Although the authors did find that FES in conjunction with traditional rehabilitation led to greater improvements in quadriceps strength and motor control than traditional rehabilitation alone, these gains may have been even greater had the participants trained using volitional effort combined with FES assistance. In addition, unlike the stationary ergometers used in previously published studies on FES cycling in individuals with CP [41, 42], the tricycle-based system in this study is capable of overground applications [24].

4.2. The Use of an Auxiliary Hub Motor (Participants 3 and 4). This work demonstrated that not all children with CP are able to pedal a tricycle without assistance. The use of the auxiliary hub motor allowed us to run quantitative exercise tests at clearly defined power increments. The work of Johnston et al. and our own pilot work have demonstrated the erratic cycling patterns used by children with CP of GMFCS levels III and IV [21, 24]. By using the auxiliary hub motor, we were able to quantify cycling performance in individuals with poor cycling ability. This information is difficult or impossible to gather without the use of the motor assistance due to the low, erratic, or negative work rates. For example, participant 3 was unable to cycle at a target power output of 0 Watts without motor assistance. Thus, he did not have the strength and coordination to volitionally cycle while working against only the resistance of his own legs. This resistance can be thought of as retarding torque which can be caused by muscle coactivation and muscle spasm, friction from the cycle drive train, and inertial resistance from the mass of the limbs. The cycling system, motor, and software, however, allowed us to collect power output data, despite the fact that the power output was less than 0 W. We discovered that during passive cycling, with the motor propelling the individual's legs, the power recorded was −8 W, indicating that the motor assistance required to passively move his limbs through the revolution was 8 W (Figure 6). With the use of the auxiliary motor and software, we were able to determine that the individual can contribute approximately 4 W of cycling effort for a net power output of −4 W. The contributing effort of the individual would be immeasurable with a nonmotorized system because of the inability to record negative power output for the individual unable to complete a crank revolution without motor assistance.

4.3. Cycling Performance. Using FES-assisted cycling technology in adolescents with CP is a novel translational approach from the treatment of individuals with SCI [24, 25]. The results demonstrated that FES-assisted cycling can be used to facilitate cycling in children who cannot cycle independently (Figure 5). Although in some cases the application of FES produced immediate increases in power output

(Figure 5(a)), in others it took several minutes to see a significant increase in power (Figure 6). In the participant who required a few minutes of FES assistance to demonstrate increased power output (Figure 6), however, the early phase of active FES-assisted cycling also demonstrated a decrease in his resistive torque (he was fighting the passive movement of his legs less than in the volitional test).

4.3.1. Incremental Load Tests. The application of FES assistance led to increased peak power output and heart rate during incremental cycling tests (Figures 3(a) and 3(b)). This participant also maintained a higher average heart rate when cycling with FES assistance (Figure 3(d)). The data from this participant demonstrates that FES assistance can facilitate an elevated heart rate while cycling. Thus, we hypothesize that FES-assisted cycling training has the potential to allow children with CP to cycle more vigorously and achieve therapeutic levels of effort which could result in improved cardiovascular fitness in individuals with CP who are not able to exercise in a traditional manner [12, 16–18].

Additionally, FES assistance also decreased the variability in cycling cadence and power output. Decreased variability in cadence signifies improved cycling performance and better potential for carryover into prolonged endurance cycling and overground cycling applications.

4.3.2. Constant Load Tests. Participants performing constant load tests also demonstrated increased power output and decreased variability in cadence and power output, which were immediate, when FES assistance was applied (Figures 4 and 6). In participant 3, the power output recorded in the volitional effort test (Figure 6—blue trace) decreases following the passive cycling phase; thus the motor had to work harder to turn the crank when the individual was helping than it did when he was resting and allowed the motor to move his legs passively. This observation demonstrates participant 3's inefficiency and unproductive volitional cycling ability, and we hypothesize that this is due to the increased coactivation and mechanical inefficiency of cycling which results in a lower net torque and an increase in the resistive torque. The decrease in power output observed in the volitional test (Figure 6—blue trace) was not present when FES assistance was applied to assist with muscle contraction timing (Figure 6—red trace). This participant was able to use less resistive torque when the FES assistance was applied. We hypothesize that with further training and FES assistance, this participant may have the potential to begin to produce positive torque and power output when cycling with the use of the auxiliary hub motor. Although the differences between FES-assisted and volitional cycling conditions were not statistically different, we anticipate that changes would be present with a larger sample size.

4.4. Limitations. The conclusions on the application of FES assistance on cycling in children with CP are limited due to the small sample size and the pilot nature of this preliminary work. The investigators acknowledge the risk of selection bias because (1) the participants were not randomly selected, (2) participants were required to have sufficient cognition and communication skills to participate in testing and training procedures, and (3) patients and families volunteering to participate may not be representative of the general population of children with CP. In addition, due to participant availability, not all participants completed a full series of constant load and incremental tests with and without FES assistance. This work was also limited in that metabolic data were not captured to provide additional insight on oxygen consumption and metabolic efficiency during cycling tests with and without FES assistance.

5. Conclusions

The objectives of this study were to (1) adapt methods and assess the feasibility of applying FES cycling technology in adolescents with CP, (2) determine methods of performing cycling tests in adolescents with CP of GMFCS levels III-IV, and (3) evaluate the *immediate* effects of FES assistance on cycling performance. Functional electrical stimulation-assisted cycling was tolerated well and resulted in increased cadence, power output, and heart rates and decreased variability in cycling performance. Such improvements in cycling ability with the application of FES assistance may make cycling for fitness attainable for individuals with CP with impaired cycling ability. The use of an auxiliary motor to control cadence may be necessary to perform quantitative cycling tests in individuals with CP with poor cycling ability. This study was designed to evaluate FES-assisted cycling as a method of improving cycling ability and to develop evaluation methods for use in future studies designed to assess potential benefits of this type of exercise intervention. Novel techniques such as FES-assisted cycling have the potential to provide a method for exercise and fitness for individuals with CP whose physical impairments limit their level of physical activity. With FES-assisted training and the immediate improvement in cycling ability it produces, there is the potential for carryover into overground cycling to provide recreational opportunities for individuals with CP. Future work will focus on determining the optimum stimulation settings for this approach and an evaluation of the effect of FES-assisted cycling training on cycling and cardiovascular performance.

Acknowledgments

A. T. Harrington submitted this paper as partial fulfillment of the requirements for the Ph.D. degree at the University of Delaware. This paper was supported in part by The Foundation for Physical Therapy PODS II Scholarship (Tokay Harrington); Shriners Hospital for Children Fellowship Grant 10650 (McRae), Shriners Hospitals for Children Research Grant 9159, and NIH Grant HD062588 (S. C. K. Lee). In addition, the authors thank Jenny Lee and Stephen Capella for assistance with data collection and Dr. Stuart Binder-Macleod for support in study design and interpretation of data.

References

[1] M. Bax, M. Goldstein, P. Rosenbaun et al., "Proposed definition and classification of cerebral palsy, April 2005," *Developmental Medicine and Child Neurology*, vol. 47, no. 8, pp. 571–576, 2005.

[2] D. W. Davis, "Review of cerebral palsy, Part I: description, incidence, and etiology," *Neonatal Network*, vol. 16, no. 3, pp. 7–12, 1997.

[3] D. L. Damiano, T. L. Martellotta, J. M. Quinlivan, and M. F. Abel, "Deficits in eccentric versus concentric torque in children with spastic cerebral palsy," *Medicine and Science in Sports and Exercise*, vol. 33, no. 1, pp. 117–122, 2001.

[4] M. E. Wiley and D. L. Damiano, "Lower-extremity strength profiles in spastic cerebral palsy," *Developmental Medicine and Child Neurology*, vol. 40, no. 2, pp. 100–107, 1998.

[5] S. K. Stackhouse, S. A. Binder-Macleod, and S. C. K. Lee, "Voluntary muscle activation, contractile properties, and fatigability in children with and without cerebral palsy," *Muscle and Nerve*, vol. 31, no. 5, pp. 594–601, 2005.

[6] L. T. Taft, "Cerebral palsy," *Pediatric Reviews*, vol. 16, no. 11, pp. 411–418, 1995.

[7] J. R. Engsberg, S. A. Ross, K. S. Olree, and T. S. Park, "Ankle spasticity and strength in children with spastic diplegic cerebral palsy," *Developmental Medicine and Child Neurology*, vol. 42, no. 1, pp. 42–47, 2000.

[8] S. A. Ross and J. R. Engsberg, "Relation between spasticity and strength in individuals with spastic diplegic cerebral palsy," *Developmental Medicine and Child Neurology*, vol. 44, no. 3, pp. 148–157, 2002.

[9] M. M. Hoffer, "Current concepts review. Management of the hip in cerebral palsy," *Journal of Bone and Joint Surgery A*, vol. 68, no. 4, pp. 629–631, 1986.

[10] C. J. Lin, L. Y. Guo, F. C. Su, Y. L. Chou, and R. J. Cherng, "Common abnormal kinetic patterns of the knee in gait in spastic diplegia of cerebral palsy," *Gait and Posture*, vol. 11, no. 3, pp. 224–232, 2000.

[11] A. L. Hof, "Changes in muscles and tendons due to neural motor disorders: implications for therapeutic intervention," *Neural Plasticity*, vol. 8, no. 1-2, pp. 71–81, 2001.

[12] J. H. Rimmer, "Physical fitness levels of persons with cerebral palsy," *Developmental Medicine and Child Neurology*, vol. 43, no. 3, pp. 208–212, 2001.

[13] M. N. Orlin, R. J. Palisano, L. A. Chiarello et al., "Participation in home, extracurricular, and community activities among children and young people with cerebral palsy," *Developmental Medicine and Child Neurology*, vol. 52, no. 2, pp. 160–166, 2010.

[14] K. J. Bell, S. Õunpuu, P. A. DeLuca, and M. J. Romness, "Natural progression of gait in children with cerebral palsy," *Journal of Pediatric Orthopaedics*, vol. 22, no. 5, pp. 677–682, 2002.

[15] L. A. Koman, B. P. Smith, and J. S. Shilt, "Cerebral palsy," *The Lancet*, vol. 363, no. 9421, pp. 1619–1631, 2004.

[16] J. H. Rimmer, B. Riley, E. Wang, A. Rauworth, and J. Jurkowski, "Physical activity participation among persons with disabilities: barriers and facilitators," *American Journal of Preventive Medicine*, vol. 26, no. 5, pp. 419–425, 2004.

[17] J. H. Rimmer, B. Riley, E. Wang, and A. Rauworth, "Accessibility of health clubs for people with mobility disabilities and visual impairments," *American Journal of Public Health*, vol. 95, no. 11, pp. 2022–2028, 2005.

[18] J. H. Rimmer, "The conspicuous absence of people with disabilities in public fitness and recreation facilities: lack of interest or lack of access?" *American Journal of Health Promotion*, vol. 19, no. 5, pp. 327–329, 2005.

[19] J. H. Rimmer, "Exercise and physical activity in persons aging with a physical disability," *Physical Medicine and Rehabilitation Clinics of North America*, vol. 16, no. 1, pp. 41–56, 2005.

[20] T. E. Johnston, A. E. Barr, and S. C. K. Lee, "Biomechanics of submaximal recumbent cycling in adolescents with and without cerebral palsy," *Physical Therapy*, vol. 87, no. 5, pp. 572–585, 2007.

[21] T. E. Johnston, L. A. Prosser, and S. C. K. Lee, "Differences in pedal forces during recumbent cycling in adolescents with and without cerebral palsy," *Clinical Biomechanics*, vol. 23, no. 2, pp. 248–251, 2008.

[22] T. E. Johnston, R. T. Lauer, and S. C. Lee, "The effects of a shank guide on cycling biomechanics of an adolescent with cerebral palsy: a single-case study," *Archives of Physical Medicine and Rehabilitation*, vol. 89, no. 10, pp. 2025–2030, 2008.

[23] T. E. Johnston, A. E. Barr, and S. C. K. Lee, "Biomechanics of recumbent cycling in adolescents with cerebral palsy with and without the use of a fixed shank guide," *Gait and Posture*, vol. 27, no. 4, pp. 539–546, 2008.

[24] C. G. A. McRae, T. E. Johnston, R. T. Lauer, A. M. Tokay, S. C. K. Lee, and K. J. Hunt, "Cycling for children with neuromuscular impairments using electrical stimulation-Development of tricycle-based systems," *Medical Engineering and Physics*, vol. 31, no. 6, pp. 650–659, 2009.

[25] A. M. Tokay, C. G. McRae, T. Johnston, and S. C. Lee, "The use of functional electrical stimulation assisted cycling in adolescents with cerebral palsy," *Biomedizinische Technik—Biomedical Engineering*, vol. 53, supplement 1, pp. 376–378, 2008.

[26] E. G. Fowler, L. M. Knutson, S. K. DeMuth et al., "Pediatric endurance and limb strengthening for children with cerebral palsy (PEDALS)—a randomized controlled trial protocol for a stationary cycling intervention," *BMC Pediatrics*, vol. 7, article 14, 2007.

[27] E. G. Fowler, L. M. Knutson, S. K. DeMuth et al., "Pediatric endurance and limb strengthening (PEDALS) for children with cerebral palsy using stationary cycling: a randomized controlled trial," *Physical Therapy*, vol. 90, no. 3, pp. 367–381, 2010.

[28] S. L. Kaplan, "Cycling patterns in children with and without cerebral palsy," *Developmental Medicine and Child Neurology*, vol. 37, no. 7, pp. 620–630, 1995.

[29] H. Williams and T. Pountney, "Effects of a static bicycling programme on the functional ability of young people with cerebral palsy who are non-ambulant," *Developmental Medicine and Child Neurology*, vol. 49, no. 7, pp. 522–527, 2007.

[30] T. E. Johnston, "Biomechanical considerations for cycling interventions in rehabilitation," *Physical Therapy*, vol. 87, no. 9, pp. 1243–1252, 2007.

[31] R. T. Lauer, T. E. Johnston, B. T. Smith, and S. C. K. Lee, "Lower extremity muscle activity during cycling in adolescents with and without cerebral palsy," *Clinical Biomechanics*, vol. 23, no. 4, pp. 442–449, 2008.

[32] L. D. Duffell, N. D. N. Donaldson, T. A. Perkins et al., "Long-term intensive electrically stimulated cycling by spinal cord-injured people: effect on muscle properties and their relation to power output," *Muscle and Nerve*, vol. 38, no. 4, pp. 1304–1311, 2008.

[33] D. J. Newham and N. D. N. Donaldson, "FES cycling," *Acta Neurochirurgica, Supplementum*, no. 97, pp. 395–402, 2007.

[34] G. M. Davis, N. A. Hamzaid, and C. Fornusek, "Cardiorespiratory, metabolic, and biomechanical responses during functional electrical stimulation leg exercise: health and fitness benefits," *Artificial Organs*, vol. 32, no. 8, pp. 625–629, 2008.

[35] T. E. Johnston, B. T. Smith, O. Oladeji, R. R. Betz, and R. T. Lauer, "Outcomes of a home cycling program using functional electrical stimulation or passive motion for children with spinal cord injury: a case series," *Journal of Spinal Cord Medicine*, vol. 31, no. 2, pp. 215–221, 2008.

[36] L. Griffin, M. J. Decker, J. Y. Hwang et al., "Functional electrical stimulation cycling improves body composition, metabolic and neural factors in persons with spinal cord injury," *Journal of Electromyography and Kinesiology*, vol. 19, no. 4, pp. 614–622, 2009.

[37] N. Hjeltnes, A. K. Aksnes, K. I. Birkeland, J. Johansen, A. Lannem, and H. Wallberg-Henriksson, "Improved body composition after 8 wk of electrically stimulated leg cycling in tetraplegic patients," *American Journal of Physiology*, vol. 273, no. 3, pp. R1072–R1079, 1997.

[38] J. S. Petrofsky, H. Heaton Jr., and C. A. Phillips, "Outdoor bicycle for exercise in paraplegics and quadriplegics," *Journal of Biomedical Engineering*, vol. 5, no. 4, pp. 292–296, 1983.

[39] P. Krause, J. Szecsi, and A. Straube, "Changes in spastic muscle tone increase in patients with spinal cord injury using functional electrical stimulation and passive leg movements," *Clinical Rehabilitation*, vol. 22, no. 7, pp. 627–634, 2008.

[40] G. C. B. Elder, J. Kirk, G. Stewart et al., "Contributing factors to muscle weakness in children with cerebral palsy," *Developmental Medicine and Child Neurology*, vol. 45, no. 8, pp. 542–550, 2003.

[41] T. E. Johnston and S. F. Wainwright, "Cycling with functional electrical stimulation in an Adult with spastic diplegic cerebral palsy," *Physical Therapy*, vol. 91, no. 6, pp. 970–982, 2011.

[42] E. Trevisi, S. Gualdi, C. C. De et al., "Cycling induced by functional electrical stimulation in children affected by cerebral palsy: case report," *European Journal of Physical and Rehabilitation Medicine*. In press. http://www.minervamedica.it/en/journals/europa-medicophysica/article.php?cod=R33Y9999N00A0114.

[43] R. Palisano, P. Rosenbaum, S. Walter, D. Russell, E. Wood, and B. Galuppi, "Development and reliability of a system to classify gross motor function in children with cerebral palsy," *Developmental Medicine and Child Neurology*, vol. 39, no. 4, pp. 214–223, 1997.

[44] T. E. Johnston, R. R. Betz, and R. T. Lauer, "Impact of cycling on hip subluxation in children with spinal cord injury," *Journal of Pediatric Orthopaedics*, vol. 29, no. 4, pp. 402–405, 2009.

[45] M. J. Buchfuhrer, J. E. Hansen, T. E. Robinson, D. Y. Sue, K. Wasserman, and B. J. Whipp, "Optimizing the exercise protocol for cardiopulmonary assessment," *Journal of Applied Physiology Respiratory Environmental and Exercise Physiology*, vol. 55, no. 5, pp. 1558–1564, 1983.

[46] K. J. Hunt, B. A. Saunders, C. Perret et al., "Energetics of paraplegic cycling: a new theoretical framework and efficiency characterisation for untrained subjects," *European Journal of Applied Physiology*, vol. 101, no. 3, pp. 277–285, 2007.

[47] S. Ferrante, A. Pedrocchi, G. Ferrigno, and F. Molteni, "Cycling induced by functional electrical stimulation improves the muscular strength and the motor control of individuals with post-acute stroke. Europa Medico-Physica-SIMFER," *European Journal of Physical and Rehabilitation Medicine*, vol. 44, no. 2, pp. 159–167, 2008.

Hindawi Publishing Corporation
International Journal of Pediatrics
Volume 2012, Article ID 583249, 11 pages
doi:10.1155/2012/583249

Research Article

Factors Influencing Physical Activity in Children and Youth with Special Health Care Needs: A Pilot Study

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Received 14 December 2011; Revised 3 February 2012; Accepted 13 February 2012

Academic Editor: Kristie F. Bjornson

Background. Evidence suggests that children and youth with special health care needs (CYSHCN) have decreased physical activity compared to peers. This study describes weight status and physical activity in CYSHCN and identifies factors associated with physical activity and community resources to promote physical activity. *Methods*. Parents ($n = 21$) and CYSHCN ($n = 23$) were recruited from a pediatric clinic. The most prevalent diagnoses were autism ($n = 7$, 30%) and cerebral palsy ($n = 3$, 13%). Interviews were conducted with parents for information on physical activity and community resources. Children's height and weight were measured to calculate body mass index (BMI). *Results*. The majority of CYSHCN ($n = 13$, 59%) were obese. CYSHCN did not meet recommended levels of 60 minutes of daily physical activity and engaged in more screen time than recommended. More children with cognitive/behavioral/emotional diagnoses were obese compared to children with physical/medical diagnoses. A majority of parents ($n = 16$, 73%) indicated their CYSHCN need more supervision to participate in physical activity in community programs. *Conclusion*. The majority of CYSHCN in this study were obese and sedentary. Resources to promote physical activity are needed for this population.

1. Introduction

A major emphasis in health care today is health promotion and disease prevention driven, in part, by the increased prevalence of childhood overweight and obesity and decreased physical activity levels among children [1]. Children and youth with special health care needs (CYSHCN) are at an increased risk for obesity and inactivity compared to their peers with typical development [2–4]. CYSHCN may have physical, cognitive, and/or emotional conditions that limit their abilities to be physically active, which may increase risk for overweight and obesity. CYSHCN are defined by the federal Maternal and Child Health Bureau as, "*those who have or are at increased risk for a chronic physical, developmental, behavioral, or emotional conditions and who also require health and related services of a type or amount beyond that required by children generally*" [5]. Health promotion strategies for CYSHCN may not be addressed in primary care or in rehabilitation services due to time constraints and competing chronic and/or acute medical needs. It is especially important to promote healthy weight in CYSHCN because chronic secondary conditions accompanying overweight and obesity may lead to health problems that limit independence [4].

Health consequences of obesity in childhood and adolescence include high blood pressure and high cholesterol, which are risk factors for cardiovascular disease (CVD) [6], increased risk of decreased glucose tolerance, insulin resistance and type 2 diabetes [7], breathing problems, such as sleep apnea, and asthma [8, 9] joint and musculoskeletal problems [8, 10], fatty liver disease, gallstones, and gastroesophageal reflux [7, 8], and risk of social and psychological problems, such as discrimination and poor self-esteem

[7, 11, 12]. Children who are obese have a high likelihood of being obese as adults [13–15] and may be at risk for serious health conditions such as heart disease, diabetes, and some cancers [16].

The current prevalence of obesity in children with typical development has increased from 5% to 17%, more than a threefold increase in the last 20–30 years [17]. Although overweight and obesity are known detriments to overall health, there is no national statistic of overweight or obesity specific to CYSHCN. The National Health and Nutrition Examination Survey (NHANES) database has been examined to determine overweight and obesity in children with developmental disorders and functional limitations [2]. Findings suggest that children with physical activity limitations were more than twice as likely to be overweight compared to children without these limitations. Responses from an online health promotion survey among adolescents with special health care needs indicated that 16.8% were obese and 19.3% were overweight compared to national database on typically developing peers where 13% were obese and 15.8% were overweight [18].

Physical inactivity is a known risk factor for overweight and obesity for all children [19] and studies suggest CYSHCN participate less in physical activity than their typically developing peers [20]. This is in part due to the impairments that CYSHCN experience because of their medical conditions/diagnoses and because of barriers to physical activity in the physical or built environment. Additional barriers to physical activity in the built environment include inaccessible playgrounds (nonadaptive equipment) and inaccessible school and work environments [20, 21]. Neighborhood characteristics such as crime and traffic patterns also pose barriers to outdoor physical activity as reported by parents of CYSHCN who were overweight or obese [18].

The social and/or family environment is important to facilitate healthy behaviors in children. Parent health behaviors establish norms and set routines that can influence a child's level of physical activity [22]. Parents monitor the health behaviors of their children and are an appropriate source for information on child health behaviors [22]. Moreover, parents of CYSHCN may be more invested in their child's health-related behaviors, activities, and services due to the chronicity and intensity of their child's health condition(s) [23]. However, the social and family environment may pose barriers to physical activity and healthy lifestyles. For example, in the social environment lack of necessary staff to provide a safe and supportive environment during organized physical recreation and highly competitive team sports may pose barriers that exclude CYSHCN from participating in active leisure [20]. The family environment may present barriers to physical activity for CYSHCN if parents do not have the time or financial resources for sporting equipment or membership fees. Parents may have limited social support to be sure their children get to participate in active recreation (i.e., a single-mother may be the head of the household) or families may live in poverty. These kinds of family factors present barriers to physical activity and are associated with higher levels of obesity in CYSHCN [24–26]. In planning and implementing this pilot study, the International Classification of Functioning Model (ICF) was used as the guiding conceptual framework. The ICF Model is an enablement model that uses a holistic perspective to focus on child's *abilities* given his or her health condition(s) [27]. This model consists of personal dimensions of health (body functions and body structure; activity; participation) and the contextual factors (physical and social) that may influence personal health outcomes. As illustrated in Figure 1, the ICF model was critical to frame the study and help identify personal and environmental factors that may influence physical activity.

Families seek health and medical advice from their children's primary care providers (PCPs) [28]. They often look to the PCPs for advice and resources to promote their children's health [28]. PCPs have indicated that they need more information and training to provide effective interventions and give appropriate guidance for patients and their families [29]. Therefore, it is key to learn from parents the resources that they use and those they need to promote physical activity and health in their children. It is important to develop a community resource database to support and inform clinical practice so that PCPs can direct families to available, accessible resources to promote healthy, active lifestyles for patients and their families.

It is important to note that participants in this pilot study were recruited from the primary care clinic in the Center for CYSHCN at St. Christopher's Hospital for Children (SCHC), a large pediatric tertiary care hospital in an urban community. Therefore, the participants were medically stable even though many had significant health and environmental challenges. Some of the environmental challenges these families and CYSHCN face are due to the community in which they live (the neighborhoods surrounding and served by SCHC). SCHC is located in Eastern North Philadelphia, in Pennsylvania's 1st Congressional District. This area is described as having the third highest childhood poverty rate in the nation (45% compared to the national average of 22%), the second highest percentage of children living in single parent families in the nation (67% compared to the national average of 34%), the second most food insecure district in the nation (49.6% of households between 2008–2010), and the poorest neighborhood in the Commonwealth of Pennsylvania [30, 31].

The primary purposes of this pilot study were to describe child factors (weight status, diagnosis), child activity levels (physical activity and sedentary behaviors), and parent factors (parent education, income, and employment) and to identify social and environmental facilitators and barriers to physical activity for CYSHCN. A secondary purpose was to explore relationships among child and parent factors and child activity levels. We hypothesized that parent factors would be correlated with childhood weight status categories and activity levels. Further, we hypothesized that childhood weight status categories would be correlated with physical activity and sedentary behaviors. Lastly, we hypothesized that childhood weight status categories would be correlated with medical diagnoses or conditions [2, 18].

The final purpose of this study was to inform PCPs about community facilitators and barriers to physical activity for

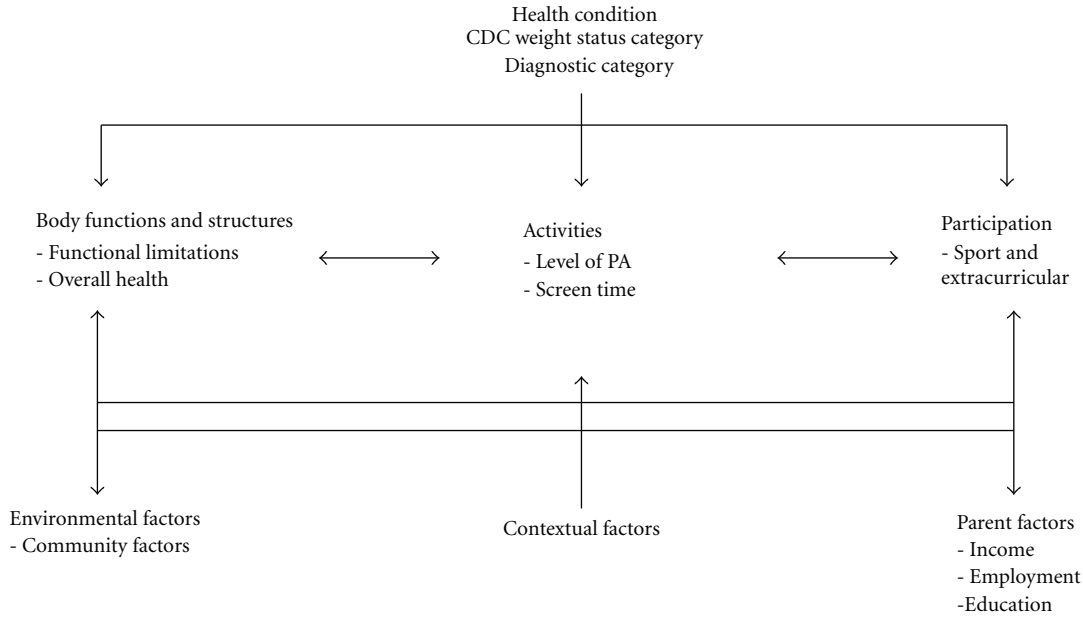

Figure 1: Modified ICF model.

CYSHCN and their families so they can provide appropriate guidance and resources and advocate with and for families for more community resources to promote active, healthy lifestyles.

2. Materials and Methods

2.1. Participants. The participants in this study were CYSHCN ($n = 23$) and their parents or legal guardians ($n = 21$), including mothers ($n = 17$), one father, one foster father, and two grandmothers. Two parents each had two CYSHCN. Inclusion criteria were that children were ages 3–18 years, had a diagnosed special health care need(s), were medically stable and ambulatory, and were patients in the primary care clinic at the Center for CYSHCN at SCHC. Both parents and children needed to be proficient in English. A sample of convenience was recruited by the medical director (pediatrician) and nurse manager and participants were enrolled by the study team. Child and parent demographics can be found in Table 1.

Parent-child dyads were chosen to participate in this pilot study based on the evidence that parents are role models for their children, they provide opportunities for their children to participate in physical activities, and they may be more invested in their children's activities and services due to the demands of their child's health condition(s) [22, 23].

Information on children's diagnoses indicated that the most frequent primary diagnosis was autism ($n = 7$, 30%) followed by cerebral palsy ($n = 3$, 13%), and asthma ($n = 2$, 9%). Eleven children (48%) each had a unique primary diagnosis, and the majority were genetic syndromes and neuromuscular conditions. Moreover, most children had one or more diagnoses in addition to their primary diagnosis, which is listed on Table 2. Due to the heterogeneity of the children's primary diagnoses, the research team created two diagnostic categories; physical/medical conditions ($n = 13$, 57%) and cognitive/emotional/behavioral conditions ($n = 10$, 43%) (see Table 2).

Most parents ($n = 14$, 67%) indicated that their children used between 1 and 8 pieces of equipment or adaptive devices (mean = 1.3). Six children (33%) used nebulizers or portable inhalers and three children (13%) were on gastrostomy tubes. Of the three children with cerebral palsy, one was classified as Gross Motor Function Classification System level II (GMFCS II) and two were classified as GMFCS level III [32].

At the time of this study, 74% ($n = 17$) of children were on medications and parents reported that their children took up to seven prescribed medications (mean = 3.5). Six children (33%) used inhaled steroids, which may be associated with weight gain [33]. Five of these children were in the physical/medical group.

Primary care providers (PCPs) often classify CYSHCN using the Complexity Index [34]. This tool uses a 10-point ordinal scale to rate the medical severity and social or family complexity of a child's condition [34]. CYSHCN in this study were assigned a rating by their PCP. Table 3 shows the distribution of complexity scores for children in this study. We present these ratings to describe the participants and to provide information on the contextual factors (personal and social environment) contributing to the severity of their conditions. Note that all CYSHCN have moderate to severe medical problems and 35% ($n = 8$) also have complicating social or family issues.

2.2. Measures. This is a cross-sectional exploratory, descriptive study in which we examined the relationship among child, family, and activity variables (see Figure 2). Additionally we conducted in-depth interviews with families to gather

Table 1: Parent/guardian and child demographics.

Variable	Child	Parent
Gender n (%) (Child $n = 23$, Parent, $n = 21$)		
Male	17 (74)	2 (10)
Female	6 (26)	19 (90)
Age mean (sd, range) (Child, $n = 23$, Parent, $n = 20$)	9.7 (4.64, 3–18)	38 (16.17, 24–58)
Race n (%) (Child, $n = 23$, Parent, $n = 20$)		
Asian	1 (4)	1 (5)
Black/African American	6 (26)	5 (25)
Native Hawaiian/Pacific Islander	1 (4)	1 (5)
White	5 (22)	5 (25)
Other	10 (43)	8 (40)
Annual household income ($n = 17$) *n* (%)		
< $15,000	NA	7 (41)
$15,000–29,999	NA	4 (24)
$30,000–$44,999	NA	4 (24)
$45,000–59,999	NA	1 (6)
$75,000–$99,999	NA	1 (6)
Child health factors mean (sd)		
Average BMI percentile ($n = 22$)	80 (31)	NA
Average number of days child achieves recommended level of physical activity (60 minutes of moderate-to-vigorous physical activity) ($n = 22$)	4.7 (1.9)	NA
"Screen time" (average no. of hours/typical weekday) ($n = 21$)	3.1 (2.3)	NA

Table 2: Primary diagnoses and diagnostic categories.

Primary diagnosis	Number of children	Diagnosis category
Autism	7	Cognitive/emotional/ behavioral
Cerebral palsy	3	Physical/medical
Asthma	2	Physical/medical
ADHD	1	Cognitive/emotional/ behavioral
Mood disorder	1	Cognitive/emotional/ behavioral
Severe intellectual disability	1	Cognitive/emotional/ behavioral
Charge syndrome	1	Physical/medical
Premature	1	Physical/medical
Beckwith-Wiedemann syndrome	1	Physical/medical
Noonan syndrome	1	Physical/medical
Spina bifida	1	Physical/medical
Seizures	1	Physical/medical
Brain tumor	1	Physical/medical
Spherocytosis	1	Physical/medical

qualitative data about community resources and barriers to physical activity for their CYSHCN. Each parent-child dyad participated in one data collection session, which included a series of questionnaires. Children were measured on height and weight to calculate body mass index (BMI).

Child Medical Information Questionnaire (CMIQ). This questionnaire was used in past research on physical activity and fitness programs for CYSHCN [35, 36]. Parents completed this questionnaire to provide information on their children's diagnoses or medical conditions, medications, medical technology, seizures, allergies, or problems with vision, hearing, or communication. Other information obtained from this questionnaire included the child's past surgical interventions, exercise restrictions, use of assistive devices, home modifications, and child's participation in physical activity programs or on sports teams.

Parent and Child Demographic and Health Behavior Questionnaires (PDHBQ and CDHBQ). These two questionnaires were developed for this study and were based on questionnaires used in past parent-child health behavior research on physical activity and sedentary behaviors [22]. The PDHBQ (36 items) and CDHBQ (31 items) used in the current study were revised to be more applicable to CYSHCN. Items on the PDHBQ included family structure, parent and child race and ethnicity, education and income levels, employment status, parent perceptions of child's overall weight and parent perceptions of their child's physical activity, and sedentary levels. Additionally, the PDHBQ had items for parents to rate availability and accessibility of physical activity resources in their neighborhood. The CDHBQ included items similar to

Table 3: Complexity Index Scores.

Complexity	Description	*n* (%)
0	Well child	0
0S	Well, no medical problems, but does have complicating social or family issues	0
1	One moderate medical problem involving one organ system	0
1S	One moderate medical problem involving one organ system with complicating social or family issues	0
2	One moderate or severe medical problem, involving one organ system with complications	5 (21.7)
2S	One moderate or severe medical problem, involving one organ system with complications and with complicating social or family issues	1 (4)
3	Two or more moderate or severe medical problems, involving two or more organ systems	9 (40)
3S	Two or more moderate or severe medical problems, involving two or more organ systems and with complicating social or family issues	5 (21.7)
4	Two or more moderate or severe medical problems, involving two or more organ systems with complications	1 (4)
4S	Two or more moderate or severe medical problems, involving two or more organ systems with complications and with complicating social or family issues	2 (8.6)

"S" indicates a child with a psychosocial issue or factor in the household (includes but not limited to single parent, foster care, and history of domestic violence) [27].

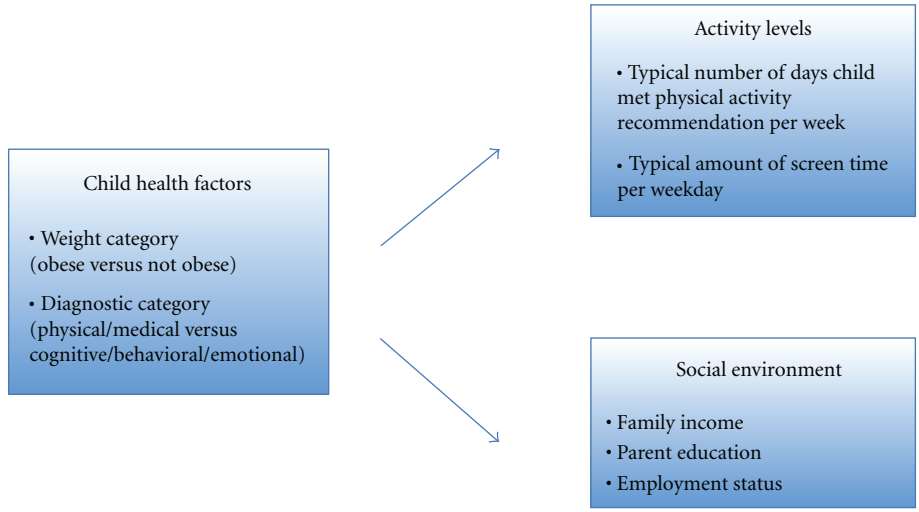

Figure 2: Items explored in correlation analysis.

the PDHBQ with additional items on perceptions of height and weight and ratings of overall health and desire to be healthier.

The health behavior items for both questionnaires were from the Youth Risk Behavior Surveillance Survey (YRBSS) for high-school and middle-school-aged students [37]. Some of the parent items were from the Behavioral Risk Factor Surveillance Survey (BRFSS) [38]. The specific items of interest for this study were the physical activity and sedentary behaviors items. We provided a definition of physical activity on the questionnaire to reduce ambiguity for the participants. The physical activity definition and PDHBQ items on physical activity and sedentary behavior are provided in Table 4.

Specific questions were asked of parents regarding what special assistance or adaptations children needed to be physically active. These adaptions may present as facilitators or barriers to a child's opportunities to participate in healthy activities. Using a 5-point rating scale, parents rated the degree to which an item presented as a barrier. Items included: child's mobility and health limitations, supervision and adaptive equipment in community programs, family finances to pay for adaptive equipment, and appropriate school-based physical education classes (see Table 5).

Lastly, to determine community resources and supports to promote health behaviors, parents were interviewed about availability and accessibility of physical activity resources in the families' community. Parents were also asked to identify

Table 4: PDHBQ items used in analysis.

Definitions	Items
(i) *Physical activity* is any activity that increases your child's heart rate and makes him or her get out of breath some of the time. (ii) *Physical activity* can be done in sports, playing with friends, or walking to school. (iii) Some examples of *physical activity* are running, brisk walking, rollerblading, biking, dancing, skateboarding, swimming, playing soccer, basketball, or football, and surfing.	Over a *typical or usual week*, on how many days is *your child physically active* for a total of at least 60 *minutes* per day? 0 day 1 day 2 days 3 days 4 days 5 days 6 days 7 days
Sedentary behavior was defined as time spent in "screen time." Screen time includes time spent watching the television, playing video games, and using the computer for nonhomework reasons.	How many hours is your child engaged in screen time on a school day? (a) My child does not watch TV, play video games, or use the computer on school days (b) Less than 1 hour per day (please specify number of minutes):—— (c) 1 hour per day (d) 2 hours per day (e) 3 hours per day (f) 4 hours per day (g) 5 hours per day

Table 5: Most common barriers to physical activity.

Item	Sometimes to always a barrier *n* (%)
(a) My child needs more supervision than is usually available in community programs ($n = 22$)	16 (73)
(b) My limited finances to pay for adaptive equipment for my child ($n = 20$)	12 (60)
(c) The lack of school-based physical education and/or activity programs that are appropriate for my child to participate in physical activity ($n = 19$)	9 (47)
(d) My child's mobility limitations or fragile health ($n = 22$)	10 (46)
(e) The lack of adaptive equipment in community programs to help my child participate ($n = 21$)	5 (24)

health-promoting resources that they perceived to be absent from their community. Specific questions asked in the PDHBQ were as follows:

> Please tell us what resources are available in your community to promote physical activity (e.g., *parks, fitness and recreation centers, summer camps, before or after school programs, public pools.*)
>
> Are the above resources accessible? *(Meaning—Are they easy to get to? Are they open at convenient times? Are the prices affordable?)*
>
> Are there any physical activity resources in community that you do not have, but would like to see?

Body Mass Index (BMI). BMI was calculated from each child's anthropometric measures (height and weight). Children's height was measured using a stadiometer (Road Rod 214 Portable Stadiometer, SECA, Hanover, MD) and weight was measured with a digital scale (UMO 26; Tanita, Arlington Heights, IL). All children could maintain upright posture for height measures although several required two measurers, one to ensure the head and trunk were aligned and to position the head piece and take the measure, the other to support lower extremities with proper foot and knee alignment.

2.3. Procedure

2.3.1. Recruitment and Enrollment. The PCP and nurse manager at SCHC approached families of children who met the inclusion criteria. Over the course of this one-year study, active recruitment and enrollment was conducted for eight months. A total of 45 parents gave permission to be contacted for participation in the study and 21 parents (47%) were enrolled. Approximately 250 phone calls were placed in an attempt to enroll the 45 parents. In many instances phone numbers were disconnected and parents were unable to be reached. Barriers for other parents who declined the invitation to participate were no time, difficulty accessing transportation, and lack of childcare. Forty-two appointments were scheduled to enroll the 21 participating parents. Reasons why scheduled appointments were missed included children being sick or hospitalized, parents forgetting, and poor weather conditions.

2.3.2. Data Collection Session. Upon arrival to the clinic at SCHC, parents and CYSHCN were escorted to a private

examination room in the clinic area. There was only one data collection session in the study. The session lasted approximately 90 minutes.

First, parents and children completed informed consent and assent forms, respectively. All questionnaires were administered by trained interviewers using the same sequence (CMIQ, PDHBQ, and CDHBQ). Most children required assistance from their parents to complete the CDHBQ. After completion of questionnaires, the child's anthropometric measures were taken. A standardized protocol was used to document height and weight and to calculate BMI [22].

Self-report measures were used in this study despite inherent biases in reporting because these are the usual measures to obtain demographic and descriptive information. Self-report measures are used most often to measures physical activity in CYSHCN with most evidence specific to children with cerebral palsy [39, 40]. Little evidence is available to support the validity or reliability of accelerometers or pedometers to measure physical activity in CYSHCN with available evidence specific to children and adolescent with cerebral palsy [41]. There are no studies that report using objective measures of sedentary behaviors for CYSHSN, so self-report is an appropriate measure [39, 40].

2.4. Data Analysis. A mixed methods design was implemented for descriptive and correlation analyses. The PASW-Statistic 18.0 [42] statistical package was used to generate frequencies and correlations. Parents responses on the PDHBQ were used in the analysis. Child responses on the CDHBQ were not analyzed because of the amount of missing and incomplete data due to most children's inability to complete the CDHBQ (i.e., cognition and/or attention limitations). In situations where children are too young or lack the cognitive ability to respond on their own behalf, parents have been found to reliably report their child's health information [43].

Descriptive analyses were generated for child factors (weight status and diagnostic categories) and PDHBQ items (parent ratings of child's physical activity level and sedentary behavior; parent demographics (education, income and employment); parent ratings on facilitators and barriers to physical activity).

Chi-square analysis, Fisher's Exact Tests, and Spearman correlations were used to test the hypotheses and measure correlations among child factors (weight status, diagnostic category) parent factors (income, education, and employment); child activity level (physical and sedentary behaviors). The alpha level was set at $P < 0.10$ to reduce Type II error. Given the study design and sample size, the larger P value will allow us to see potential trends in correlations [44, 45].

Qualitative analysis was conducted to examine parent responses to the open-ended physical activity resource questions. These questions were administered via in-depth interviews and open-ended questions. Responses were coded based on themes and categories and frequencies were documented to identify unique trends or patterns [46]. Figure 2 illustrates the items examined in the association analysis. Only statistically significant findings are reported in the results section.

3. Results

3.1. Descriptive Analysis

3.1.1. Body Composition, Physical Activity, and Child Health Factors. Many of the participants ($n = 12$, 55%) were obese (BMI ≥ 95th percentile). An additional 14% ($n = 3$) of participants were overweight (BMI ≥ 85th percentile). On average, parents reported that their children were physically active for 60 minutes or more on 4.68 (SD 2.0) days per week. None of the children in this study were reported to have participated on sports teams. Parents also reported that their children engaged in an average of 3.1 (SD 2.3) hours of screen time per day. Screen time was defined as television, video games, and computer use (for nonhomework activities) [47].

3.1.2. Parent Perception of Child's Overall Health. Parents used a 5-point Likert scale to rate their child's overall health and to rate how much their child's health or physical limitations were a barrier to physical activity. A majority (68%, $n = 15$) of parents rated their children in good, very good, or excellent overall health. A majority of parents (55%, $n = 12$) indicated that mobility limitations were rarely or never a barrier.

3.1.3. Environmental Facilitators and Barriers to Physical Activity. Parents were asked to rate the level by which specific social, organizational, and community resources or lack of resources presented as barriers to physical activity. Parents were asked to rate each item (presented in Table 5) on a 5-point Likert scale from "never" to "always" a barrier. A majority of parents perceived the level of supervision in community programs as a barrier. Additionally, a majority of parents agreed that limited finances to pay for adaptive equipment were also a barrier.

Parents were asked to identify available and accessible resources in their community to support physical activity behaviors for their children. Availability was defined as resources that exist in the community and accessibility was defined as, affordable, convenient hours of operation, easy to get to, and adaptable to the specific needs of their child. Parents were also asked what community resources were needed to support physical activity. When examining open-ended responses to these questions, parks were the most frequently identified physical activity resource in parents' communities. In addition, most parents perceived these parks to be an accessible resource to promote physical activity for their CYSHCN. On the other hand, pools are needed in many parents' communities. Twenty-one percent of parents said that they do not have a pool in their community, but would like one. Pools only accounted for five percent of the available resources cited by parents. A complete summary of responses is displayed in Table 6.

3.2. Factors Associated with Obesity in CYSHCN. Results of the chi-square analyses (Table 7) suggest that CYSHCN who were classified as having cognitive/behavioral/emotional conditions were more likely to be obese compared to

Table 6: Availability, accessibility and need of physical activity resources in the community.

Resource	Available	Accessible	Need
Parks	35%	80%	0%
*Other	18%	90%	NA
Recreation Centers	16%	90%	7%
Gyms	11%	67%	10%
YMCA	9%	100%	14%
Pools	5%	100%	21%
Playgrounds	5%	0%	10%
Sports	0%	0%	7%
Summer Camps	0%	0%	7%
**Needs	NA	NA	24%

*Other refers to available/accessible resources (such as church, bowling, variety club) that were only mentioned once.
**Needs refers to needed resources (such as horse backing riding, gymnastics and after school programs) that were only mentioned once.

Table 7: Chi-square analysis.

Diagnostic category ($n = 22$)	Obese n (%)	Not obese n (%)	Exact sig (2-sided)*
Physical/medical	5 (42)	7 (58)	$P = 0.10$
Cognitive/behavioral/ emotional	8 (80)	2 (20)	
Family income category ($n = 17$)	Obese n (%)	Not Obese n (%)	Exact Sig (2-sided)*
<$15,000 per year	2 (29)	5 (71)	$P = 0.06$
≥$15,000 per year	8 (80)	2 (20)	

*Fisher's Exact Test.

CYSHCN who were classified as having physical/medical conditions ($P = 0.10$). CYSHCN who belong to families of higher income were more likely to be obese compared to CYSHCN who belonged to families of lower income ($P = 0.06$).

Results of the Spearman's correlation suggest that BMI percentile was significantly, inversely related to the number of days in which a child achieves recommended levels of physical activity in a typical week ($r = -.43$, $P < 0.05$).

4. Discussion

A majority of CYSHCN in this study were obese. This finding is supported by previous studies demonstrating a high prevalence of overweight and obesity in CYSHCN [2, 18]. We hypothesized that parent factors would be correlated with child weight status category and activity levels. Additionally, we hypothesized that child weight status would be correlated with activity levels and diagnostic category.

There were three significant findings from the hypothesis testing. Children who were categorized with cognitive/emotional/behavioral conditions were significantly more likely to be obese than those categorized with physical/medical conditions. This finding is supported by previous studies of CYSHCN [18] and may highlight the need for more accessible facilities, family and recreation or fitness staff training, and adapted activities to provide safe, appropriate environments with sustained moderate-to-vigorous physical activity to result in improved health and weight status. Further research is necessary to understand health outcomes and long-term implications in this specific population of CYSHCN.

The second significant finding from hypothesis testing suggests that there was a significant relationship between increased parent income and increased obesity. This finding is counterintuitive but may be partially explained by the fact that most of the CYSHCN in the higher-income families had cognitive/emotional/behavioral conditions and children with these conditions were significantly more obese than children with physical/medical conditions.

Finally, the significant inverse correlation between obesity and physical activity is in keeping with the research evidence and suggests that inactivity in CYSHCN may lead to obesity [18]. On average, parents indicated that their children did not achieve the recommended level of daily moderate-to-vigorous physical activity (60 minutes every day) [48]. Data for typical high school students suggests that only 18.4% achieve the recommended daily levels of moderate-to-vigorous physical activity [49] but this is much better than our findings of 0%. Although CYSHCN did not meet the physical activity recommended threshold, parents responses were higher than expected and were not supported by the high obesity rates in the participants. A possible explanation is that the physical activity parents observed may not be at the necessary intensity (moderate to vigorous) to result in energy expenditure for CYSHCN to achieve and maintain healthy weight. In candid conversations during interview sessions, some parents spoke of their children not being able to fully participate in sports and recreation to the degree and competitive level of children with typical development. Also, as mentioned previously, we must consider the biases inherent in self-report data [50].

All CYSHCN in this study engage in screen time beyond the American Academy of Pediatrics recommendation (1-2 hours per day) [50]. In our study, CYSHCN participated in less screen time compared to typically developing children. Evidence suggests that preschoolers with typical development are exposed up to 4 hours of screen time per weekday, and older children and adolescents are exposed up to approximately 3 hours of television per day, not including time spent with videogames and computers [51–53]. Other studies have found that children with cognitive disorders, such as autism, spend a greater amount of their free time watching television or playing video games than their peers with typical development [54]. Children with cerebral palsy have been found to spend relatively the same amount of time engaged in screen time as their peers with typical development [55]. Findings from our study suggest that CYSHCN participate in less screen time than children with typical development and less screen time than reported in other studies with CYSHCN. This discrepancy may be due to parent report or that the item on screen time in the PDHBQ potentially caused confusion. We asked parents to

add up their children's screen time, which may have lead to inaccuracy in adding those numbers. There are limited studies investigating screen time in CYSHCN, this health behavior warrants future research.

In this study, parents identified multiple barriers to physical activity, with a lack of supervision in community programs being the most common concern. A majority of parents agreed that limited finances and inability to pay for adaptive equipment posed barriers to their children being able to participate in physical activities. Additionally parents identified multiple resources they needed in their community to promote physically activity for their children, including pools, gyms, and summer camps.

It is worth noting that no parents in this study had a playground in their community that was accessible to their CYSHCN. Since the average age of children in our study was 9.8 years and many children had cognitive delay, it is likely that by cognitive or developmental age, playgrounds may be useful to these families. Lack of playground resources speaks to potential problems with resource availability and/or appropriate adaptations for universal access for all children. National physical activity guidelines [48] should include recommendations on ways to adapt environments to promote active recreation in all children to make the guidelines more comprehensive and inclusive. Although these guidelines mention adults and CYSHCN, minimal recommendations are provided to address the health and physical activity needs of these populations [48]. Special attention and detail in national guidelines and clinical guidelines may help guide PCPs in their health promotion recommendations to CYSHCN. Likewise, citywide health promotion initiatives should include information and recommendations on universal access to include CYSHCN and their families so they can participate in and benefit from these initiatives as much as children with typical development and their families can.

5. Limitations

A main limitation in this study is the small sample size that resulted from difficulty with recruitment and enrollment. Data analysis was therefore limited to descriptive statistics and nonparametric correlation analyses. Subjects enrolled in the study were a heterogeneous population of CYSHCN making it impossible to examine specific conditions or diagnoses as related to weight status or physical activity levels. Likewise, the socioeconomic status of participant families varied but the majority of families were from the lower end of the socioeconomic status making it impossible to generalize across all income levels of families with CYSHCN.

As in many studies, there is the potential for selection bias, and the parent and child health questionnaires brought inherent self-report bias. Lastly, the 36-item PDBHQ and 31-item CDBHQ may have been too long and burdensome. Upon observation it appeared that parents and children might not have answered questions with the same interest level near the end of the interview. Through the course of data collection, it was determined that the CDBHQ was not appropriate for most CYSHCN in this study. If this study were to continue, only the PDBHQ would be used. A major revision would be necessary for the CDBHQ if it were to be used again.

6. Conclusion

To our knowledge, this is the first study conducted in primary care to examine parent perspectives and community resource needs to promote physical activity in CYSHCN. Also, this clinic-based study was conducted with a vulnerable population living in an underserved community. Additionally, this study contributes to the limited literature documenting overweight and obesity CYSHCN. Parents identified multiple resources in their communities to promote physical activity. PCPs should provide families with anticipatory guidance and recommendations to find those resources for health promotion and participation in healthy, active recreation. PCPs for CYSHCN who use the Complexity Index Scale to describe their patients should consider adding functional items to the scale to identify physical activity limitations and needs for their patients and families to promote healthy weight and active lifestyles. Physical and occupational therapists working in primary care clinics for CYSHCN should be included as part of the medical team to provide these children and families with resources, equipment ideas, and program ideas to promote healthy participation in physical activity and active recreation. Resources that were reported as a "need" in communities should be brought to the attention of local policy makers, PCPs, and advocates of CYSHCN. It is important that parents recognize the prevalence of overweight and obesity in CYSHCN and assist their children to engage in physical activity for health promotion and prevention of secondary conditions. Likewise, it is important that universally accessible community resources are available to CYSHCN to promote health behaviors.

Acknowledgments

The authors would like to thank the Philadelphia Department of Public Health, Department of Maternal, Child and Family Health, Division of Children with Special Health Care Needs for the 2010 Community Activity Grant that funded this project. Also, they would like to thank the medical and clinical staff at St. Christopher's Hospital for Children for allowing us to conduct this study in their Center for CYSHCN. Finally, and most importantly, we are very grateful to the parents and children who gave their time to participate in this study.

References

[1] T. M. McCambridge, D. T. Bernhardt, J. S. Brenner et al., "Active healthy living: prevention of childhood obesity through increased physical activity," *Pediatrics*, vol. 117, no. 5, pp. 1834–1842, 2006.

[2] L. G. Bandini, C. Curtin, C. Hamad, D. J. Tybor, and A. Must, "Prevalence of overweight in children with developmental disorders in the continuous national health and nutrition

examination survey (NHANES) 1999–2002," *Journal of Pediatrics*, vol. 146, no. 6, pp. 738–743, 2005.

[3] A. Y. Chen, S. E. Kim, A. J. Houtrow, and P. W. Newacheck, "Prevalence of obesity among children with chronic conditions," *Obesity*, vol. 18, no. 1, pp. 210–213, 2010.

[4] P. M. Minihan, S. N. Fitch, and A. Must, "What does the epidemic of childhood obesity mean for children with special health care needs?" *The Journal of Law, Medicine and Ethics*, vol. 35, no. 1, pp. 61–77, 2007.

[5] M. McPherson, P. Arango, H. Fox et al., "A new definition of children with special health care needs," *Pediatrics*, vol. 102, no. 1 I, pp. 137–140, 1998.

[6] D. S. Freedman, Z. Mei, S. R. Srinivasan, G. S. Berenson, and W. H. Dietz, "Cardiovascular risk factors and excess adiposity among overweight children and adolescents: the bogalusa heart study," *Journal of Pediatrics*, vol. 150, no. 1, pp. 12–17.e2, 2007.

[7] E. P. Whitlock, S. B. Williams, R. Gold, P. R. Smith, and S. A. Shipman, "Screening and interventions for childhood overweight: a summary of evidence for the US preventive services task force," *Pediatrics*, vol. 116, no. 1, pp. e125–e144, 2005.

[8] J. C. Han, D. A. Lawlor, and S. Y. Kimm, "Childhood obesity," *The Lancet*, vol. 375, no. 9727, pp. 1737–1748, 2010.

[9] E. R. Sutherland, "Obesity and asthma," *Immunology and Allergy Clinics of North America*, vol. 28, no. 3, pp. 589–602, 2008.

[10] E. D. Taylor, K. R. Theim, M. C. Mirch et al., "Orthopedic complications of overweight in children and adolescents," *Pediatrics*, vol. 117, no. 6, pp. 2167–2174, 2006.

[11] W. Dietz, "Health consequences of obesity in youth: childhood predictors of adult disease," *Pediatrics*, vol. 101, no. 3, pp. 518–525, 1998.

[12] M. B. Schwartz and R. Puhl, "Childhood obesity: a societal problem to solve," *Obesity Reviews*, vol. 4, no. 1, pp. 57–71, 2003.

[13] F. M. Biro and M. Wien, "Childhood obesity and adult morbidities," *American Journal of Clinical Nutrition*, vol. 91, no. 5, pp. 1499S–1505S, 2010.

[14] R. C. Whitaker, J. A. Wright, M. S. Pepe, K. D. Seidel, and W. H. Dietz, "Predicting obesity in young adulthood from childhood and parental obesity," *The New England Journal of Medicine*, vol. 337, no. 13, pp. 869–873, 1997.

[15] M. K. Serdula, D. Ivery, R. J. Coates, D. S. Freedman, D. F. Williamson, and T. Byers, "Do obese children become obese adults? A review of the literature," *Preventive Medicine*, vol. 22, no. 2, pp. 167–177, 1993.

[16] National Institutes of Health, *Clinical Guidelines on the Identification, Evaluation, and Treatment of Overweight and Obesity in Adults: the Evidence Report*, National Institutes of Health, U.S. Department of Health and Human Services, Bethesda, Md, USA, 1998.

[17] C. L. Ogden, M. D. Carroll, L. R. Curtin, M. M. Lamb, and K. M. Flegal, "Prevalence of high body mass index in US children and adolescents, 2007-2008," *Journal of the American Medical Association*, vol. 303, no. 3, pp. 242–249, 2010.

[18] J. H. Rimmer, E. Wang, K. Yamaki, and B. Davis, "Documenting disparities in obesity and disability," FOCUS Technical Brief 24, 2009.

[19] W. B. Strong, R. M. Malina, C. J. R. Blimkie et al., "Evidence based physical activity for school-age youth," *The Journal of Pediatrics*, vol. 146, no. 6, pp. 732–737, 2005.

[20] J. A. Rimmer and J. L. Rowland, "Physical activity for youth with disabilities: a critical need in an underserved population," *Developmental Neurorehabilitation*, vol. 11, no. 2, pp. 141–148, 2008.

[21] M. Law, T. Petrenchik, G. King, and P. Hurley, "Perceived environmental barriers to recreational, community, and school participation for children and youth with physical disabilities," *Archives of Physical Medicine and Rehabilitation*, vol. 88, no. 12, pp. 1636–1642, 2007.

[22] M. E. O'Neil, P. A. Shewokis, K. K. Falkenstein et al., "Psychosocial factors and health perceptions in parents and children who are overweight or obese," *Obesity*, vol. 18, no. 8, pp. 1558–1565, 2010.

[23] United States Department of Health and Human Services, "National survey of children with special health care needs chartbook 2005-2006," 2010, http://mchb.hrsa.gov/cshcn05/.

[24] D. Young-Hyman, L. J. Herman, D. L. Scott, and D. G. Schlundt, "Care giver perception of children's obesity-related health risk: a study of African American families," *Obesity Research*, vol. 8, no. 3, pp. 241–248, 2000.

[25] F. L. Wu, S. Yu, I. L. Wei, and T. J. Yin, "Weight-control behavior among obese children: association with family-related factors," *The Journal of Nursing Research*, vol. 11, no. 1, pp. 19–30, 2003.

[26] G. King, M. Law, S. King, P. Rosenbaum, M. K. Kertoy, and N. L. Young, "A conceptual model of the factors affecting the recreation and leisure participation of children with disabilities," *Physical and Occupational Therapy in Pediatrics*, vol. 23, no. 1, pp. 63–90, 2003.

[27] World Health Organization, "Toward a common language for functioning, disability and health: ICF The international classification of functioning, disability and health," 2002, http://www.who.int/classifications/icf/training/icfbeginnersguide.pdf.

[28] V. F. Keeton and C. Kennedy, "Update on physical activity including special needs populations," *Current Opinion in Pediatrics*, vol. 21, no. 2, pp. 262–268, 2009.

[29] S. E. Barlow, "Expert committee recommendations regarding the prevention, assessment, and treatment of child and adolescent overweight and obesity: summary report," *Pediatrics*, vol. 120, supplement 4, pp. S164–S192, 2007.

[30] The Annie E. Casey Foundation, "Kids count data center," 2009, http://datacenter.kidscount.org/.

[31] Food Research and Action Center, "Food hardship in America 2010: Households with and without children," 2011, http://frac.org/pdf/aug2011_food_hardship_report_children.pdf.

[32] R. J. Palisano, P. Rosenbaum, S. Walter, D. Russell, E. Wood, and B. Galuppi, "Development and reliability of a system to classify gross motor function in children with cerebral palsy," *Developmental Medicine and Child Neurology*, vol. 39, no. 4, pp. 214–223, 1997.

[33] K. H. Carlsen and J. Gerritsen, "Inhaled steroids in children: adrenal suppression and growth impairment," *European Respiratory Journal*, vol. 19, no. 6, pp. 985–988, 2002.

[34] Center for Medical Home Improvement, "Complexity index (Phoenix Pediatrics) David Hirsch, MD," 2001, http://www.medicalhomeinfo.org/downloads/pdfs/CareCoordinationToolkit06.pdf.

[35] M. A. Fragala-Pinkham, S. M. Haley, J. Rabin, and V. S. Kharasch, "A fitness program for children with disabilities," *Physical Therapy*, vol. 85, no. 11, pp. 1182–1200, 2005.

[36] M. A. Fragala-Pinkham, S. M. Haley, and S. Goodgold, "Evaluation of a community-based group fitness program for children with disabilities," *Pediatric Physical Therapy*, vol. 18, no. 2, pp. 159–167, 2006.

[37] Centers for Disease Control and Prevention, *Middle School Youth Risk Behavior Survey*, 2005YRB, Atlanta, Ga, USA, 2005.
[38] Centers for Disease Control and Prevention, "Behavior Risk Factor Surveillance System Questionnaire," 2005, http://www.cdc.gov/brfss/questionnaires/pdf-ques/2005brfss.pdf.
[39] K. M. Clanchy, S. M. Tweedy, and R. Boyd, "Measurement of habitual physical activity performance in adolescents with cerebral palsy: a systematic review," *Developmental Medicine and Child Neurology*, vol. 53, no. 6, pp. 499–505, 2011.
[40] C. M. Capio, C. H. P. Sit, B. Abernethy, and E. R. Rotor, "Physical activity measurement instruments for children with cerebral palsy: a systematic review," *Developmental Medicine and Child Neurology*, vol. 52, no. 10, pp. 908–916, 2010.
[41] K. M. Clanchy, S. M. Tweedy, R. N. Boyd, and S. G. Trost, "Validity of accelerometry in ambulatory children and adolescents with cerebral palsy," *European Journal of Applied Physiology*, vol. 111, no. 12, pp. 2951–2959, 2011.
[42] SPSS Inc., "PASW Statistics 18.0.," SPSS Inc., Chicago, Ill, USA, 2009, http://www-01.ibm.com/software/analytics/spss/products/statistics/.
[43] J. W. Varni, C. A. Limbers, and T. M. Burwinkle, "Parent proxy-report of their children's health-related quality of life: an analysis of 13,878 parents' reliability and validity across age subgroups using the PedsQL 4.0 generic core scales," *Health and Quality of Life Outcomes*, vol. 5, article 2, 2007.
[44] M. E. O'Neil, R. J. Palisano, and S. L. Westcott, "Relationship of therapists' attitudes, children's motor ability, and parenting stress to mothers' perceptions of therapists' behaviors during early intervention," *Physical Therapy*, vol. 81, no. 8, pp. 1412–1424, 2001.
[45] H. Motulsky, *Intuitive Biostatistics*, Oxford University Press, New York, NY, USA, 1995.
[46] J. W. Creswell, *Research Design: Qualitative, Quantitative, and Mixed Methods Approaches*, Thousand Oaks, Calif, USA, Sage, 2nd edition, 2003.
[47] National Governor's Association (NGA), "Successful State Strategies to Prevent Childhood Obesity," 2009, http://www.aap.org/obesity/community_advocacy/NGA.pdf#Limit Screen Time.
[48] United States Department of Health and Human Services, "physical activity guidelines for all Americans," 2008, http://www.health.gov/paguidelines/pdf/paguide.pdf.
[49] US Department of Health and Human Services, Center for Disease Control and Prevention. Youth Risk Behavior Surveillance Survey (YRBSS), 2009, http://www.cdc.gov/healthyyouth/yrbs/pdf/us_physical_trend_yrbs.pdf.
[50] S. A. Adams, C. E. Matthews, C. B. Ebbeling et al., "The effect of social desirability and social approval on self-reports of physical activity," *American Journal of Epidemiology*, vol. 161, no. 4, pp. 389–398, 2005.
[51] M. E. Bar-On, D. D. Broughton, S. Buttross et al., "Children, adolescents, and television," *Pediatrics*, vol. 107, no. 2, pp. 423–426, 2001.
[52] P. S. Tandon, C. Zhou, P. Lozano, and D. Christakis, "Preschoolers' total daily screentime at home and by type of child care," *The Journal of Pediatrics*, vol. 158, no. 2, pp. 297–300, 2011.
[53] V. C. Strasburger, D. A. Mulligan, T. R. Altmann et al., "Policy statement—children, adolescents, obesity, and the media," *Pediatrics*, vol. 128, no. 1, pp. 201–208, 2011.
[54] M. O. Mazurek, P. T. Shattuck, M. Wagner, and B. P. Cooper, "Prevalence and correlates of screen-based media use among youths with autism spectrum disorders," *Journal of Autism Development Disorders*. In press.
[55] C. A. Maher, M. T. Williams, T. Olds, and A. E. Lane, "Physical and sedentary activity in adolescents with cerebral palsy," *Developmental Medicine and Child Neurology*, vol. 49, no. 6, pp. 450–457, 2007.

Hindawi Publishing Corporation
International Journal of Pediatrics
Volume 2012, Article ID 460384, 9 pages
doi:10.1155/2012/460384

Research Article

Participation in Physical Activity for Children with Neurodevelopmental Disorders

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Received 1 November 2011; Accepted 13 February 2012

Academic Editor: Maria A. Fragala-Pinkham

The purpose of this study was to compare rates of participation for children (4–9 years of age) with neurodevelopmental disorders (NDDs) with and without externalizing behavior problems (EBPs) with children without disability and to examine mediators of the relation between disability and physical activity participation. Data for this study were drawn from Cycle 7 (2006-07) of the Canadian National Longitudinal Survey of Children and Youth (NLSCY). The frequency of children's participation in organized sports or physical activities varied depending on the child's health condition with children with NDDs and both NDDs and EBPs participating least in organized sports or physical activities followed by children with EBPs only. In contrast, there were no statistically significant differences by health group for children's participation in unorganized sports or physical activities. These differences remained even after controlling for the effects of other child and family sociodemographic characteristics, except for children with EBPs only. These findings highlight the importance of considering children's primary and other existing health conditions as well as family sociodemographic characteristics in order to better understand the factors that influence participation in organized physical activities for children with disabilities.

1. Introduction

The advantages of participation in physical activity for children are well established, including physical, psychosocial, and emotional benefits [1, 2]. For children with disabilities who may experience impairments in mobility, functioning, and overall well-being, participation in physical activity may be particularly valuable in that physical activity promotes physical development and health within a social context [3]. However, children with disabilities may be limited in terms of motor abilities and social skills, thus impacting their ability to participate in physical activities. Despite this, specific physical activity recommendations have been made by the Canadian Paedeatric Society [4] for certain chronic disease conditions including juvenile idiopathic arthritis, hemophilia, asthma, and cystic fibrosis, pointing to the recognition or importance of physical activity for children with chronic health conditions or disabilities. The purpose of the current study is to compare rates of participation for children with neurodevelopmental disorders with and without externalizing behavior problems with children without disability, and to examine mediators of the relation between disability and activity participation. Imms et al. [5] and King et al. [6] found disparate correlates of informal (unorganized) versus formal (organized) physical activity for children with disabilities, thus these two types of activities were considered separately.

Reviews of the physical activity and disability literature suggest that participation in physical activity is beneficial for children with disabilities for therapeutic reasons as well as general physical and social development [7]. Although disease state and individual capacity must be considered, the potential benefits of physical activity participation (including psychosocial, muscular strength, and cardiovascular capacity) may offset or even counteract disease progression, for example, for children with asthma or juvenile idiopathic arthritis [8]. Physical activity participation might also be advantageous in minimizing or counteracting secondary health (e.g., diabetes, obesity) and impairments (e.g., decreased strength, poor balance) for children with chronic health conditions [4, 9–11]. Finally, in addition to the physical benefits, physical activity participation has been associated with enhanced social identity, including perceptions of competence and similarity to peers, enhanced

self-worth, and strengthening social interaction and bonding [3].

An NDD is an impairment of the growth and development of the central nervous system, emerging in early development [12] and impacting various domains of functioning, including ambulation, information-processing, self-regulation, and communication [13]. Examples of NDDs include cerebral palsy, intellectual disability, and autism spectrum disorder. In Canada, 6% of children between the ages of 4 to 11 years were found to have an NDD [14]. Children with an NDD are also at greater risk of developing emotional and behavioral problems compared to their peers [15, 16]. Over half (55%) of Canadian children with an NDD were also found to exhibit one or more externalizing behavior problems (EBPs) [14]. EBPs refer to a group of behavior problems that are manifested in children's outward behavior [17] such as conduct disorder/physical aggression, indirect aggression, and hyperactivity-inattention problems.

While an abundance of information is available regarding rates of physical activity participation for children in general [1, 18, 19], less is known about rates of participation for children with disabilities specifically. Children with disabilities are less likely to engage in physical activity than their healthy peers [4, 10, 11, 20] and much of the literature on physical activity and disability has focused on children with cerebral palsy (CP) as it is the most common form of childhood disability [11, 21–23]. Bjornson and colleagues [21] found that children aged 8 to 13 years with CP took fewer steps per day (as measured by accelerometry) and were less active overall than typically developing peers. These differences were attenuated by gross motor ability, such that the highest functioning individuals (according to the Gross Motor Function Classification System, GMFCS [24]) were more similar to children with typical development [21].

Using data from a national Canadian disability survey, Kowalchuk and Crompton [25] found that 63% of Canadian children with disabilities (those who had difficulties with daily living activities or those for whom a physical or mental condition or health problem reduced activities) engaged in organized sport or physical activity, most of whom were doing so at least once per week. These results are higher than those reported by Longmuir and Bar-Or [26] who found that within their clinical sample of Canadian children, 47% of those with a chronic medical condition (e.g., kidney disease, hearing impairment) and 26% of those with a physical disability (including CP, brain injury, spina bifida, and muscular dystrophy) were physically active. Unfortunately, this study did not include a control/comparison group. Differences in these reported rates of participation are likely due to differences in the sample characteristics (e.g., the age of the study participants), the identification of disabilities, and broad definitions of physical activity. For instance, in Kowalchuk and Crompton's study, children were considered to be active (versus inactive) if they had participated in any organized sport or activity in the past 12 months, regardless of frequency, whereas Longmuir and Bar-Or created three categories of activity (active, moderately active, and sedentary) based on frequency, intensity, and duration of activities.

Children with comorbid conditions such as a physical disability and communication deficits may be at further risk of reduced physical activity participation [27]. This would suggest that research considers not only the complexity of the individual's medical or social impairment, but also any additional conditions which may impede participation in physical activity. While the general literature on EBPs and physical activity has not shown a link between EBPs such as aggression and sports participation [28, 29], to our knowledge, there are no existing studies that have investigated physical activity participation for children with NDDs with and without EBPs.

In addition to a lower amount of physical activity, children with disabilities tend to partake in different types of activities, particularly lower intensity activities, than their healthy peers [20, 30]. Despite recent advances in specific programs such as Special Olympics or Paralympic Games, children with physical disabilities may have fewer opportunities to engage in formal, organized activities, in particular competitive or elite sports contexts [3]. Some evidence has suggested that children with disabilities participate in predominantly informal, home-based activities [23, 31, 32] and are less inclined to participate in physical-based leisure activities as compared to social, recreational, or self-improvement type activities [22].

While much is known regarding the barriers and facilitating conditions of physical activity for children and youth in general [33], relatively few studies have examined barriers or facilitators to participation in physical activity for children with disabilities [23, 32, 34]. The identification of such factors may help inform recommendations for identifying resources and/or strategies for increasing opportunities and rates of participation for children with disabilities. For example, previous work on children with CP specifically has suggested that low parental education [11] and lower parental income [32] are associated with less participation in physical activity. Physical ability, often measured as gross motor function, has also been associated with participation such that better motor function has been associated with greater active-physical leisure time activity [5, 20, 23]. Similarly, Longmuir and Bar-Or [26] found that physical activity level varied dramatically by disability type, with those having more severe impairments being less likely to participate than those with less severe impairments. Moreover, Kowalchuk and Crompton [25] found that participation in physical activity was more likely if the child had a physical (70%) or nonphysical (69%) disability (e.g., learning disability), as compared to both physical and nonphysical (59%), and if the severity was mild (70%) as compared to very severe (45%). However, these differences in physical activity participation did not remain statistically significant once sociodemographic correlates were considered, suggesting that child or family characteristics (including child age, family income, and barriers to participation) were stronger determinants of participation than type or severity of disability. Identification of any comorbid conditions is thus an important consideration.

Other characteristics have also been examined as potential correlates of participation. Unlike findings in the typical

child development literature whereby boys are more likely to engage in physical activity than are girls [33, 35], an effect of gender has not typically been shown for participation in physical activity for children with disabilities [5, 6, 11, 20, 22, 26]. In one study, however, Law and colleagues [31] found that boys were more likely to participate in active physical activities than were girls (aged 6–14), as were children of dual-parent families as compared to single-parent families. Moreover, age is a potential determinant, with young (i.e., age 5 [11]) and older (age 17 [20]) children being less likely to be active than children in the mid-age range (ages 7 and 11, resp.). In the typical child development literature, access to recreational facilities has also been associated with physical activity participation [36].

Previous research on physical activity for children with disabilities has thus been limited by (i) a focus on children with CP, (ii) inconsistent definitions of physical activity, (iii) lack of control groups (i.e., comparisons to healthy children), and (iv) the omission of other comorbid conditions such as EBPs. The current study examines rates of participation in physical activity for children with NDDs living in Canada. In Canada, children's participation in physical activities often occurs outside of the school setting. The advantages of the present study include the use of a population-based set of data, a noncategorical definition of disability including NDDs with and without EBPs, comparisons with a control group of children without disability, and an examination of both organized and unorganized physical activity participation. Our study seeks to address two questions: (1) Are there differences in participation in organized and/or unorganized physical activity for children with NDDs, with and without EBPs, compared to children without disability?, and (2) What sociodemographic characteristics mediate differences between NDDs, EBPs, both NDDs and EBPs, and children without disability?

2. Materials and Methods

2.1. Data Source and Sample. Data for this study were drawn from Cycle 7 (2006-07) of the Canadian National Longitudinal Survey of Children and Youth (NLSCY), a longitudinal study of the physical and social development of Canadian children from birth into adulthood. The NLSCY, jointly conducted by Human Resources and Skills Development Canada (HRSDC) and Statistics Canada, started in 1994 and was repeated biennially. The first cohort of children who participated in the survey was between the ages of 0 to 11 years. The person most knowledgeable (PMK) of the child provided information on the participant child as well as information about him/herself, and his/her spouse or partner by completing a series of questionnaires in the household. The surveys were completed using computer-assisted interviewing (CAI) methods (either via personal interviewing in the household or telephone interviewing). In 90% of cases, the PMK was the biological mother of the child. In this study, the PMK will be referred to hereafter as the parent.

In 1994-1995, a total of 22,831 children were proportionally sampled from all areas of the country excluding children living in institutional settings and residing in Yukon, Nunavut, Northwest Territories, and First Nations reserves [37]. The cross-sectional sample for this study consisted of children who were between 4 and 9 years of age in Cycle 7 (2006-07); this was the most recent cycle in which all relevant variables were available.

2.2. Measures

2.2.1. Child Health Groups. The classification of child health consisted of four groups: (a) children with an NDD only (NDD), (b) children with an EBP only (EBP), (c) children with both an NDD and an EBP (BOTH), and (d) children who had neither a chronic condition (including an NDD or an EBP) nor an activity limitation (HEALTHY). The presence of an NDD in children was identified using a checklist of chronic conditions diagnosed by a health professional. Four parent-reported chronic conditions, including epilepsy, CP, intellectual disability, and a learning disability, were used for this definition, and 423 (3.4%) children were identified as having an NDD. This definition has been used in previous research [14, 38, 39]. It should be noted that approximately ten children were missing information required to identify the presence of an NDD and were, therefore, excluded from the sample. In addition, on the questionnaire, the term mental handicap was used to represent an intellectual disability.

The presence of an EBP was identified using a parent-reported child behavior rating scale, with items derived from the Child Behavior Checklist (CBCL [40]) and modified for Canadian children. Three EBP scales were used: hyperactivity-inattention (8 items, Cronbach's alpha = .87 [41]; for example, "How often would you say that [child's name] cannot sit still, is restless or hyperactive?"), physical aggression-conduct disorder (6 items, Cronbach's alpha = .89; for example, "How often would you say that [child] gets into many fights?"), and indirect aggression (5 items, Cronbach's alpha = .82; for example, "How often would you say that [child] when mad at someone, tries to get others to dislike that person?"). Each item was scored on a 3-point scale ranging from 0 (*never or not true)* to 2 (*often or very true*). Consistent with our previous work [14, 38], children were considered to have an EBP if at least one of their scores on the three EBP scales was two standard deviations above the mean or greater. A total of 1520 (12.3%) children were identified as having an EBP using these criteria. None of the children in the sample had missing information required to identify an EBP.

To identify children without disability (HEALTHY group), we used two exclusion criteria: (1) presence of another (non-NDD) chronic condition (a long-term condition diagnosed by a health professional, such as a heart condition, asthma, kidney disease, bronchitis, and diabetes), (2) limitation in activities (due to asthma or any other activi-ty limitations at home, at child care, at school, or other settings). There were 3266 children who had a long-term

condition and/or activity limitation and thus were excluded from the group of children without disability (HEALTHY group).

After the final classification of child health into four groups ($N = 9119$), there were 286 children in the NDD group, 1382 children in the EBP group, 137 children in the BOTH group, and 7314 children in the HEALTHY group. Approximately 25% of children in the NDD group were reported to have activity limitations at home and almost half of these children were reported to have limitations in "other" activities, including activities at child care or at school, and transportation or sports and games. Only 4.2% of children in the EBP group were found to have activity limitations at home and 8% of them had limitations in "other" activities. The percentages of children with activity limitations were higher in the BOTH group of children compared to the other groups. Approximately 43% of children who had both an NDD and EBP were found to have activity limitations at home and 63% of them were reported to have limitations in "other" settings and activities.

2.2.2. Sociodemographic Characteristics. Two other child characteristics were considered: child age (0 = preschool-age (4-5 years old), 1 = school-age (6–9 years old)), and sex (0 = male; 1 = female). In addition, four family characteristics were included in the analyses: single parent (0 = no; 1 = yes), parent's educational attainment (0 = less than or equal to high school education; 1 = greater than high school education), household income (0 = low income; 1 = moderate income; 2 = high income), and child living in a census metropolitan area (CMA) or census agglomeration (CA) using 2006 Census of Canada code (0 = no; 1 = yes). Living in a CMA or CA was used to identify areas expected to have higher access to services (i.e., higher populated areas).

The measure of household income was based on a comparison between parents' report of an estimate of their household income and the low income cutoff (LICO) score established by Statistics Canada. The LICO indicates an income level at which a family will likely spend a greater portion of its income on basic needs such as food, clothing, and shelter than does an average family of similar size [42]. An income-to-LICO ratio of less than 1 is generally defined as living in low income. In this study, a household income-to-LICO ratio equal to or greater than 1 but less than 2 was defined as moderate income, while an income-to-LICO ratio equal to or greater than 2 was defined as high income.

According to information on 2006 Standard Geographical Classification (SGC) developed by Statistics Canada [43], a CMA is a standard geographical entity consisting of an urban core, including several adjacent urban and rural areas that have a high degree of social and economic integration with that urban core. A CMA must have a population of at least 100,000 with an urban core of 50,000. To form a CA, the urban core must have a population of at least 10,000. It should be noted that if the population of the urban core of a CA declines below 10,000, the CA is retired. However, once an area is defined as a CMA, it is retained as a CMA even if its population declines below 100,000.

2.2.3. Outcome Variables. Two outcome variables were considered: children's participation in *organized* sports or physical activities and children's participation in *unorganized* sports or physical activities in the last year. The variable on organized sports or physical activities was based on two items from the survey, which asked parents to indicate how often the child had taken part in (a) sports with a coach or instructor (except dance, gymnastics or martial arts) in the past 12 months, and (b) lessons or instruction in other organized physical activities with a coach or instructor such as dance, gymnastics or martial arts in the past 12 months. Response options for both items range from 1 (*most days*) to 5 (*almost never*). Similar to previous research [44], a composite item was created to indicate children's participation in organized sports or physical activities in the last year, which was dichotomized (0 = about once a month or almost never; 1 = about once a week or more) due to skewness in responses. The second outcome variable was also a parent-reported item that assessed how often the child had taken part in unorganized sports or physical activities without a coach or instructor. The response scale was similar to the organized sports or physical activities outcome variable, and for the same reason (i.e., skewness), the responses were dichotomized to represent children's participation in unorganized sports or physical activities at least once a week versus about once a month or almost never.

2.3. Data Analysis. First, we calculated descriptive statistics (i.e., percentages) for each variable. Second, we conducted chi-square tests to examine group differences with the reference group being children without disability (i.e., children in the HEALTHY group), including group differences in participation in organized and/or unorganized physical activity for children with NDDs, with and without EBPs, compared to children without disability. The next set of analyses included a series of logistic regressions. In Model 1, we examined the associations between child health group and participation in organized sports or physical activities. In Model 2, we examined these associations after controlling for the effects of other child characteristics (i.e., age and sex). In Model 3, we included family sociodemographic characteristics in addition to the child characteristics and examined whether these factors played a role in explaining the relationships between child health group and participation in organized sports or physical activities. Moreover, we conducted contrast analyses to determine whether there were significant differences in beta coefficients among the child health groups. Finally, we performed additional logistic regression analyses, where we examined the effects of each variable separately, to determine what sociodemographic characteristics mediate differences between NDDs, EBPs, both NDDs and EBPs, and children without disability. All analyses were weighted and bootstrapped to account for complex survey design [45].

3. Results

Descriptive statistics for the total sample and for each child health group are presented in Table 1. Children in the NDD,

Table 1: Descriptive statistics of all study variables.

Characteristics (%)	Overall $N = 9119$	NDD $n = 286$	EBP $n = 1382$	BOTH $n = 137$	HEALTHY $n = 7314$
Child is school-aged	67	83*	69	89*	66
Child is male	49	65*	53*	73*	48
Child lives with a single parent	14	24*	19*	24*	13
Parent's educational attainment (>high school)	72	61*	68*	54*	74
Household income					
Low income	15	24*	20*	28*	14
Moderate income	35	37	38*	27	34
High income	50	39*	42*	45	52
Child lives in a CMA or CA	81	82	80	83	81
Child participates in organized sports or physical activities at least once a week	70	55*	66*	50*	71
Child participates in unorganized sports or physical activities at least once a week	71	73	72	63	71

*The superscript indicates group differences, with the reference group being HEALTHY at the $P < .05$ level.
CMA: census metropolitan area. CA: census agglomeration.
Source: National Longitudinal Survey of Children and Youth 2006-2007, Statistics Canada.

Table 2: Summary of results from logistic regression analyses predicting children's participation in organized sports or physical activities ($N = 9119$).

Variables	Model 1		Model 2		Model 3	
	OR	95% C.I.	OR	95% C.I.	OR	95% C.I.
NDD	.49***	.34–.71	.42***	.29–.60	.50***	.34–.72
EBP	.79*	.66–.95	.76**	.64–.92	.90	.74–1.10
BOTH	.40***	.25–.63	.32***	.20–.51	.39***	.22–.67
HEALTHY	Ref		Ref		Ref	
Child is school-aged			2.09***	1.80–2.43	2.31***	1.96–2.73
Child is boy			1.06	.92–1.21	1.07	.92–1.24
Child lives with a single parent					1.03	.84–1.27
Parent's educational attainment (>high school)					2.48***	2.12–2.89
Household income						
Low income					.48***	.39–.60
Moderate income					Ref	
High income					2.32***	1.97–2.74
Child lives in a CMA or CA					1.39***	1.19–1.63

Ref: the reference group. CMA: census metropolitan area. CA: census agglomeration.
Note. All reported values are odd ratios (OR) with 95% confidence intervals (CI).
*$P < .05$. **$P < .01$. ***$P < .001$.
Source: National Longitudinal Survey of Children and Youth 2006-2007, Statistics Canada.

EBP, and BOTH groups were more likely to be boys, live with a single parent, have a parent with less than or equal to high school education, and live in a low income household. Moreover, children in the NDD and BOTH groups were more likely to be school-age children as compared to children in the HEALTHY group. Consistent with our previous work [14, 38], these findings suggested some sociodemographic differences among the child health groups.

Regarding participation in sports or physical activity, children in the NDD, EBP, and BOTH groups were significantly less likely to participate in organized sports or physical activities than children in the HEALTHY group (see Table 1). This finding was particularly striking for the BOTH group; whereas more than 70% of children in the HEALTHY group participated in organized sports or physical activities at least once a week, only half of children in the

BOTH group, and slightly more than half (55%) of the children in the NDD group participated in organized sports or physical activities at least once a week. This was followed by children in the EBP group (66%). In contrast, there were no statistically significant differences by health group for children's participation in unorganized sports or physical activities. It should be noted that because we did not find any statistically significant differences in children's participation in unorganized sports or physical activities, this variable was not analyzed further.

The results from the logistic regression analyses are summarized in Table 2. Consistent with our comparison analyses, the results from Model 1 indicated that children in the NDD, EBP, and BOTH groups were less likely to participate in organized sports or physical activities as compared to children in the HEALTHY group. The findings from the contrast analyses suggested that the independent effect of being in the NDD and BOTH groups was similar (Wald $F = .50, P > .05$), while the independent effect of being in the EBP group was significantly lower than that for children in the NDD (Wald $F = 5.69, P < .05$) and BOTH (Wald $F = 7.80, P < .05$) groups. These findings suggest that the association between children's participation in organized sports or physical activities is stronger for children in the NDD and BOTH group than the EBP group, with these children reporting significantly less participation in organized sports or physical activities as compared to children in the HEALTHY group.

The findings from Model 2 suggested that the associations between child health group and participation in organized sports or physical activities in Model 1 remained statistically significant even after controlling for the effects of child age and sex. As expected, child age, but not sex, was significantly associated with participation in organized sports or physical activities. That is, school-age children had twice the odds (OR $= 2.09, P < .001$) of participating in organized sports or physical activities as compared to preschool-age children. This finding suggests that the child's health condition (NDD, EBP, BOTH) is associated with organized sports or physical activities over and above the effect of child age.

The results from Model 3 indicated that children in the NDD and BOTH groups were less likely to participate in organized sports or physical activities at least once a week over and above the effects of other child and family sociodemographic characteristics, including parental education, household income, and the child living in a CMA or CA. Interestingly, child EBP condition was no longer significantly associated with participation in organized sports or physical activities (OR $= .90, P > .05$) when controlling for sociodemographic characteristics. Furthermore, school-age children (OR $= 2.31, P < .001$), children who have a parent with greater than high school education (OR $= 2.48, P < .001$), who live in a high income family (OR $= 2.32, P < .001$), and in a CMA or CA (OR $= 1.39, P < .001$) were more likely to participate in organized sports or physical activities at least once a week.

We conducted additional logistic regression analyses, where each variable was examined separately, to determine which factors mediated the relationship between child health condition and participation. The findings indicated that parent's educational attainment and household income played a role in mediating the association between child EBP condition and participation in organized sports or physical activities.

Overall, these findings indicate that by comparison children in the NDD and BOTH groups were less likely to participate in organized sports or physical activities even after controlling for the effects of other child and family sociodemographic characteristics. In contrast, the likelihood of participation in organized sports or physical activities for children in the EBP group was no longer statistically different than children in the HEALTHY group after accounting for the differences in other child and family sociodemographic characteristics. A similar pattern to the first set of contrast analyses was found. Specifically, the independent effect of being in the NDD and BOTH groups was similar (Wald $F = .61, P > .05$), while the independent effect of being in the EBP group was significantly lower than that for children in the NDD (Wald $F = 7.96, P < .01$) and BOTH (Wald $F = 8.80, P < .01$) groups. These findings suggest that, even after controlling for the effects of other child and family sociodemographic characteristics, the association between children's participation in organized sports or physical activities is stronger for children in the NDD and BOTH group, with these children reporting significantly less participation in organized sports or physical activities as compared to children in the HEALTHY group.

4. Discussion

This study used a population-based Canadian survey to examine the physical activity participation of children with NDDs with and without EBPs as compared to children without disability. The results revealed that children with both NDDs and EBPs, possibly representing more "complex" health challenges, were the least likely to participate in organized physical activity, followed by children with NDDs (although the NDD group did not differ significantly from the BOTH group) and then those with EBPs as compared to children without disability.

It is possible that children with varying degrees of disability have fewer opportunities or less time to participate in organized physical activity [3], or that children with complex disabilities are less likely to be capable or feel competent in organized activities. However, the fact that children in the comorbid NDD and EBP group as well as the NDD group participated less than children without disability supports previous research that has found a link between severity of condition and activity participation [25, 27]. Severity of the condition (including the presence of a comorbid condition) may be particularly relevant in organized activity participation which requires formal motor skills and expertise, for example, the ability to manipulate a ball in many organized sports. Disability type has also previously been associated with children's perceptions of fitness, with those children with physical disabilities less likely to perceive

themselves as fit as their peers than those with a chronic medical condition [26]. Perceptions of fitness may then translate into different selection of activities, including reduced participation in organized physical activities.

However, differences in physical activity participation were demonstrated for organized but not unorganized physical activities. For unorganized physical activities, the majority of each group participated and no differences in participation rates were found in comparison to children without disability. This is in line with previous research suggesting that children with CP are more likely to engage in informal, rather than formal, physical activities [23, 31, 32]. The findings might be explained by the fact that informal activities may be more easily adapted and rules more flexible to allow for increased participation so that children with disabilities are at less of a disadvantage than is the case for formal, organized, and perhaps competitive, physical activities. Certainly, environmental barriers such as lack of adequate space and adapted activities which accommodate children's needs may be less of a concern with informal (or unorganized) types of activities [32]. There are certainly differences that need to be considered concerning availability of unorganized physical activity. For example, in Canada, children tend to participate in privately organized programming (e.g., sports clubs and organizations); however, in the United States of America (USA), many children participate in privately and federally-funded after-school programming [46].

The findings from the present study also point to other important factors that have an impact on the physical activity participation of children with disabilities, namely, of family sociodemographics. Although parent sociodemographic factors were associated with physical activity participation, they did not mediate associations of NDDs with or without EBPs and physical activity, perhaps pointing to the importance of other characteristics such as severity and motor functioning. However, parental sociodemographic characteristics did have an impact on the likelihood of participation for children with EBPs. The likelihood of participation in organized physical activities for children with EBPs who were of school age, lived in a CMA or CA, and had parents with higher levels of education and income, was no different than for children without disability. It is beyond the scope of the present study to speculate on the reasons for this but factors such as living in a CMA or CA and parental socio-economic resources may allow for differences in opportunities, programs, and environments conducive to the participation of children with EPBs.

However, for children with an NDD and those with both an NDD and an EBP, parent sociodemographic characteristics did not fully mediate the child health effects on the probability of participation in organized physical activity. These findings suggest that lower levels of participation for children with an NDD and both an NDD and EBP were not explained by sociodemographic characteristics measured in the current study. Findings from other international studies have suggested possible explanatory factors regarding participation in physical activity for children with disabilities. For example, gross motor function was shown to be a significant predictor of overall physical activity for Australian youth (aged 11–17) with CP, with those with higher levels of gross motor impairment being less likely to participate [20]. In addition, other research has suggested that child and family preferences for activity, family factors (such as cohesion), and environmental conditions (including babysitting or transportation) may be directly or indirectly associated with physical activity participation for children with disabilities [6, 32, 34]. These measures were not included in the present study as they were not available in the NLSCY and thus could not be considered. Yet, it is important to realize that factors relating not only the child but also to the family and the environment are associated with participation in physical activities for children with disabilities.

Strengths of this study include the use of a population-based sample of children which allowed us to include children with NDDs both with and without EBPs as well as an age-matched comparison group of children who had no chronic conditions or activity limitations. Previous research has been limited by small, sometimes condition-specific samples [5], with a particular focus on children with CP. We were also able to report rates of participation separately for organized as well as unorganized activities. Despite these strengths, limitations of the study include limited information on the severity of the child's condition and motor functioning, availability of appropriate activities (e.g., adapted activities), and measures of the physical environment which may impact opportunities for participation [6, 32, 34]. Finally, although a national study, sample sizes were too small to examine other questions such as the interaction effects (e.g., between gender and child health group) on physical activity participation.

We were also somewhat limited by the measure of physical activity included in the NLSCY survey. Although parent or self-reported items are frequently used in national surveys collecting physical activity information, there are limitations associated with this type of data such as reporting bias and respondent definitions of organized and unorganized activities. In addition, the parent-reported measure of physical activity included in the NLSCY does not include information about intensity, with whom the activity was performed, or preferences for activity (as does the *Children's Assessment of Participation and Enjoyment* (CAPE)), which is used in many other studies of physical activity in children or youth with disabilities [5, 6].

5. Conclusions

These findings have implications for practice and research, both within Canada and internationally. Specifically, our results highlight the importance of considering children's primary and other existing health conditions as well as family sociodemographic characteristics in order to better understand the factors that influence participation in organized physical activities. Our results also suggest that the largest differences in participation for children with health conditions is for organized and not unorganized physical activities. Policies aimed at increasing participation rates of

children with health conditions could focus on facilitating and increasing participation in organized physical activity and for some children such as those experiencing EBPs, ensuring that cost and availability are not barriers to access. While results from the current study suggest that Canadian children with an NDD and those with an NDD and an EBP are less likely to engage in organized physical activity than are their peers without disability, they are no less likely to engage in unorganized physical activity.

Acknowledgments

All authors contributed equally to this work. Thus, the order of authorship was determined alphabetically. This study was partially funded by a grant from the Canadian Institutes for Health Research (CIHR; TWC-94790). The CIHR does not impose any restrictions on free access to or publication of the research data. There was no conflict of interest. The authors would like to gratefully acknowledge contributions of Drs. Lucyna M. Lach, Peter L. Rosenbaum, and Jamie C. Brehaut.

References

[1] I. Janssen and A. G. LeBlanc, "Systematic review of the health benefits of physical activity and fitness in school-aged children and youth," *International Journal of Behavioral Nutrition and Physical Activity*, vol. 7, no. 40, 2010.

[2] A. F. Feldman and J. L. Matjasko, "Profiles and portfolios of adolescent school-based extracurricular activity participation," *Journal of Adolescence*, vol. 30, no. 2, pp. 313–332, 2007.

[3] D. E. Taub and K. R. Greer, "Physical activity as a normalizing experience for school-age children with physical disabilities. Implications for legitimation of social identity and enhancement of social ties," *Journal of Sport and Social Issues*, vol. 24, no. 4, pp. 395–414, 2000.

[4] J. Philpott, K. Houghton, and A. Luke, "Physical activity recommendations for children with specific chronic health conditions: Juvenile idiopathic arthritis, hemophilia, asthma and cystic fibrosis," *Paediatrics and Child Health*, vol. 15, no. 4, pp. 213–225, 2010.

[5] C. Imms, S. Reilly, J. Carlin, and K. J. Dodd, "Characteristics influencing participation of Australian children with cerebral palsy," *Disability and Rehabilitation*, vol. 31, no. 26, pp. 2204–2215, 2009.

[6] G. King, M. Law, S. Hanna et al., "Predictors of the leisure and recreation participation of children with physical disabilities: a structural equation modeling analysis," *Children's Health Care*, vol. 35, no. 3, pp. 209–234, 2006.

[7] C. C. Johnson, "The benefits of physical activity for youth with developmental disabilities: a systematic review," *American Journal of Health Promotion*, vol. 23, no. 3, pp. 157–167, 2009.

[8] P. J. Morris, "Physical activity recommendations for children and adolescents with chronic disease," *Current Sports Medicine Reports*, vol. 7, no. 6, pp. 353–358, 2008.

[9] D. R. Shapiro and J. J. Martin, "Multidimensional physical self-concept of athletes with physical disabilities," *Adapted Physical Activity Quarterly*, vol. 27, no. 4, pp. 294–307, 2010.

[10] J. E. Neter, D. F. Schokker, E. de Jong, C. M. Renders, J. C. Seidell, and T. L. S. Visscher, "The prevalence of overweight and obesity and its determinants in children with and without disabilities," *Journal of Pediatrics*, vol. 158, no. 5, pp. 735–739, 2011.

[11] J. N. Zwier, P. E. M. Van Schie, J. G. Becher, D. W. Smits, J. W. Gorter, and A. J. Dallmeijer, "Physical activity in young children with cerebral palsy," *Disability and Rehabilitation*, vol. 32, no. 18, pp. 1501–1508, 2010.

[12] H. Tager-Flusberg, *Neurodevelopmental Disorders*, Massachusetts Institute of Technology, Cambridge, Mass, USA, 1999.

[13] J. E. Farmer and K. K. Deidrick, "Introduction to childhood disability," in *Treating Neurodevelopmental Disabilities: Clinical Research and Practice*, J. E. Farmer, J. Donders, and S. Warschausky, Eds., pp. 3–20, The Guilford Press, New York, NY, USA, 2006.

[14] L. M. Lach, D. E. Kohen, R. E. Garner et al., "The health and psychosocial functioning of caregivers of children with neurodevelopmental disorders," *Disability and Rehabilitation*, vol. 31, no. 8, pp. 607–618, 2009.

[15] W. P. Witt, A. W. Riley, and M. J. Coiro, "Childhood functional status, family stressors, and psychosocial adjustment among school-aged children with disabilities in the United States," *Archives of Pediatrics and Adolescent Medicine*, vol. 157, no. 7, pp. 687–695, 2003.

[16] M. Hysing, I. Elgen, C. Gillberg, and A. J. Lundervold, "Emotional and behavioural problems in subgroups of children with chronic illness: results from a large-scale population study," *Child: Care, Health and Development*, vol. 35, no. 4, pp. 527–533, 2009.

[17] T. M. Achenbach and C. S. Edelbrock, "The child behavior profile:II. boys aged 12-16 and girls aged 6-11 and 12-16," *Journal of Consulting and Clinical Psychology*, vol. 47, no. 2, pp. 223–233, 1979.

[18] R. C. Colley, D. Garriguet, I. Janssen, C. L. Craig, J. Clarke, and M. S. Tremblay, "Physical activity of Canadian children and youth: accelerometer results from the 2007 to 2009 Canadian Health Measures Survey," *Health Reports*, vol. 22, no. 1, pp. 15–23, 2011.

[19] J. A. Fredricks and J. S. Eccles, "Extracurricular involvement and adolescent adjustment: Impact of duration, number of activities, and breadth of participation," *Applied Developmental Science*, vol. 10, no. 3, pp. 132–146, 2006.

[20] C. A. Maher, M. T. Williams, T. Olds, and A. E. Lane, "Physical and sedentary activity in adolescents with cerebral palsy," *Developmental Medicine and Child Neurology*, vol. 49, no. 6, pp. 450–457, 2007.

[21] K. F. Bjornson, B. Belza, D. Kartin, R. Logsdon, and J. F. McLaughlin, "Ambulatory physical activity performance in youth with cerebral palsy and youth who are developing typically," *Physical Therapy*, vol. 87, no. 3, pp. 248–257, 2007.

[22] R. J. Palisano, L. J. Kang, L. A. Chiarello, M. Orlin, D. Oeffinger, and J. Maggs, "Social and community participation of children and youth with cerebral palsy is associated with age and gross motor function classification," *Physical Therapy*, vol. 89, no. 12, pp. 1304–1314, 2009.

[23] A. Majnemer, M. Shevell, M. Law et al., "Participation and enjoyment of leisure activities in school-aged children with cerebral palsy," *Developmental Medicine and Child Neurology*, vol. 50, no. 10, pp. 751–758, 2008.

[24] R. J. Palisano, P. I. Rosenbaum, S. Walter et al., *Gross Motor Function Classification System*, Neurodevelopmental Clinical Research Unit, McMaster University, Hamilton, Canada, 1995.

[25] K. Kowalchuk and S. Crompton, "Social participation of children with disabilities," *Canadian Social Trends*, no. 88, pp. 63–72, 2009.

[26] P. E. Longmuir and O. Bar-Or, "Factors influencing the physical activity levels of youths with physical and sensory disabilities," *Adapted Physical Activity Quarterly*, vol. 17, no. 1, pp. 40–53, 2000.

[27] P. Raghavendra, R. Virgo, C. Olsson, T. Connell, and A. E. Lane, "Activity participation of children with complex communication needs, physical disabilities and typically-developing peers," *Developmental Neurorehabilitation*, vol. 14, no. 3, pp. 145–155, 2011.

[28] L. C. Findlay and R. J. Coplan, "Come out and play: shyness in childhood and the benefits of organized sports participation," *Canadian Journal of Behavioural Science*, vol. 40, no. 3, pp. 153–161, 2008.

[29] J. P. McHale, P. G. Vinden, L. Bush, D. Richer, D. Shaw, and B. Smith, "Patterns of personal and social adjustment among sport-involved and noninvolved urban middle-school children," *Sociology of Sport Journal*, vol. 22, no. 2, pp. 119–136, 2005.

[30] M. Brown and W. A. Gorden, "Impact of impairment on activity patterns of children," *Archives of Physical Medicine and Rehabilitation*, vol. 68, no. 12, pp. 828–832, 1987.

[31] M. Law, G. King, S. King et al., "Patterns of participation in recreational and leisure activities among children with complex physical disabilities," *Developmental Medicine and Child Neurology*, vol. 48, no. 5, pp. 337–342, 2006.

[32] K. Shikako-Thomas, A. Majnemer, M. Law, and L. Lach, "Determinants of participation in leisure activities in children and youth with cerebral palsy: systematic review," *Physical and Occupational Therapy in Pediatrics*, vol. 28, no. 2, pp. 155–169, 2008.

[33] J. F. Sallis, J. J. Prochaska, and W. C. Taylor, "A review of correlates of physical activity of children and adolescents," *Medicine and Science in Sports and Exercise*, vol. 32, no. 5, pp. 963–975, 2000.

[34] G. King, M. Law, S. King, P. Rosenbaum, M. K. Kertoy, and N. L. Young, "A conceptual model of the factors affecting the recreation and leisure participation of children with disabilities," *Physical and Occupational Therapy in Pediatrics*, vol. 23, no. 1, pp. 63–90, 2003.

[35] U. Ekelund, L. B. Sardinha, S. A. Anderssen et al., "Associations between objectively assessed physical activity and indicators of body fatness in 9- to 10-y-old European children: a population-based study from 4 distinct regions in Europe (the European Youth Heart Study)," *American Journal of Clinical Nutrition*, vol. 80, no. 3, pp. 584–590, 2004.

[36] G. J. Norman, S. K. Nutter, S. Ryan, J. F. Sallis, K. J. Calfas, and K. Patrick, "Community design and access to recreational facilities as correlates of adolescent physical activity and body-mass index," *Journal of Physical Activity and Health*, vol. 3, pp. S118–S126, 2006.

[37] Statistics Canada and Human Resources Development Canada, "National Longitudinal Survey of Children and Youth: Overview of survey instruments for 1994-1995, data collection 1," Minister of Industry, Ottawa, Canada, 1995, http://www.statcan.gc.ca/pub/89f0078x/89f0078x2004001-eng.pdf.

[38] R. E. Garner, R. G. Arim, D. E. Kohen et al., "Parenting children with neurodevelopmental disorders and/or behaviour problems," *Child: Care, Health and Development*. In press.

[39] R. G. Arim, R. E. Garner, J. C. Brehaut et al., "Contextual influences of parenting behaviours for children with neurodevelopmental disorders: results from a Canadian National Survey," *Disability and Rehabilitation*. In press.

[40] T. M. Achenbach and C. S. Edelbrock, "Behavioral problems and competencies reported by parents of normal and disturbed children aged four through sixteen," *Monographs of the Society for Research in Child Development*, vol. 46, no. 1, pp. 1–82, 1981.

[41] L. J. Cronbach, "Coefficient alpha and the internal structure of tests," *Psychometrika*, vol. 16, no. 3, pp. 297–334, 1951.

[42] C. Cotton, "Income research paper series: recent developments in the low income cutoffs," Minister of Industry, Ottawa, Canada, 2001, http://www.statcan.gc.ca/pub/75f0002m/75f0002m2001003-eng.pdf.

[43] Statistics Canada, "The 2006 Standard Geographical Classification. Volume I, The Classification," (Catalogue 12-571), 2006.

[44] L. C. Findlay, R. E. Garner, and D. E. Kohen, "Children's organized physical activity patterns from childhood into adolescence," *Journal of Physical Activity and Health*, vol. 6, no. 6, pp. 708–715, 2009.

[45] K. F. Rust and J. N. K. Rao, "Variance estimation for complex surveys using replication techniques," *Statistical Methods in Medical Research*, vol. 5, no. 3, pp. 283–310, 1996.

[46] J. L. Mahoney, A. L. Harris, and J. S. Eccles, "Organized activity participation, positive youth development, and the overscheduling hypothesis," *Social Policy Report*, vol. 20, no. 4, pp. 1–31, 2006.