SEDENTARY BEHAVIOUR AND PHYSICAL ACTIVITY IN INDIVIDUALS WITH CEREBRAL PALSY

EXPLORING THE OPERATIONALIZATION OF SEDENTARY BEHAVIOUR AND IDENTIFYING IMPORTANT HEALTH OUTCOMES OF PHYSICAL ACTIVITY IN INDIVIDUALS WITH CEREBRAL PALSY

BY:

Julia (Shi-Peng) Xiong, B.H.Sc.

A Thesis

Submitted to the School of Graduate Studies

In Partial Fulfillment of the Requirements

for the Degree of Master of Science

McMaster University

McMaster University
MASTER OF SCIENCE (2020)
Hamilton, Ontario (Rehabilitation Science)

TITLE: Operationalizing Sedentary Behaviour and Identifying Important Health Outcomes for Physical Activity in Individuals with Cerebral Palsy

AUTHOR: Julia (Shi-Peng) Xiong, B.H.Sc. (McMaster University)

SUPERVISOR: Dr. Jan Willem Gorter

NUMBER OF PAGES: xv, 139

LAY ABSTRACT

Engaging in physical activity (PA) and reducing sedentary behaviour (SB) decreases the chance of developing negative health outcomes, such as cardiovascular disease. Individuals with cerebral palsy (CP) across all ages spend less time active and more time sedentary than those without CP and therefore have an increased risk of health issues over the life course. We want to enhance the physical and mental health of individuals with CP through interventions and clinical care pathways that aim to increase PA and reduce SB in these individuals. Before we can do this, there are two research gaps that I will address with my thesis. First, there is not a standard definition for SB in individuals with CP, so researchers may not be reporting or measuring the same concept when studying SB. In a scoping review, I investigated the recent literature and learned that the way SB has been defined for individuals with CP generally matched those without CP. Second, we do not know which health outcomes are the most critical and important for PA promotion in individuals with CP. Without this information, it is difficult to critically appraise the literature and synthesize the effects of intervention studies when developing guidelines to manage PA in this population. I conducted a survey at a conference with clinicians and researchers. I learned from these experts that both mental and physical health outcomes are important to consider in people with CP. My work provides a summary of knowledge on SB in people with CP to help future research in reaching agreement upon a common definition for SB in this population. I have also developed a list of important and critical outcomes, which could be built upon in future discussion with stakeholders including people with CP.

ABSTRACT

Cerebral palsy (CP) is a motor disorder that causes activity limitations in life. Both children and adults with CP perform, overall, reduced physical activity (PA) levels with increased sedentary behaviour (SB), which predisposes them to negative health outcomes, such as cardiovascular disease. SB is considered a separate construct from PA because of its distinct negative health outcomes. While SB is defined in the typically developing population through a postural component of sitting, reclining or lying and energy expenditure component of ≤1.5 metabolic equivalents of task (METs), there is currently no consensus on an operationalized definition of SB for the CP population. Furthermore, there are currently no established outcomes to inform guidelines promoting PA in this population. I will address these two research gaps in my thesis, as I sought to investigate the operationalization of SB and to identify important health outcomes of PA, through expert opinions, in individuals with CP. The first chapter in my thesis is a scoping review of the operationalization of SB in individuals with CP in the recent literature. I found that i) the definition for SB in the typically developing population of ≤1.5 METs generally applies to sitting and lying in individuals with CP, and that ii) postures of sitting and lying are reported as operationalizations of SB in this population. The second chapter was a conference-based survey to identify critical and important health outcomes for PA in individuals with CP. Clinicians and researchers (n=55) who attended a workshop on PA in CP rated a list of nine pre-determined health outcomes on a 9 point Likert-type scale from the Grading of Recommendations Assessment, Development and Evaluation (GRADE) Approach, a rating scale for the importance of outcomes to inform clinical practice guideline development. The scale ranges numerically from 1 to 9 (7 to 9 – critical; 4 to 6 – important; 1 to 3 – of limited

importance) to distinguish between importance categories. The experts identified both the two included psychological aspects of health (i.e. anxiety and depression) as being critical. While some of the included physical health outcomes were rated as critical (i.e., sleep; nutrition, cardiorespiratory endurance), others were rated as important (i.e., body size, body composition, blood lipids and glucose, blood pressure). This research suggests that the operationalization of SB in individuals with CP is similar to that of the typically developing population, and that both the physical and mental aspects of health must be considered as outcomes in future PA management. A future step should be to discuss these health outcomes with patient input. The goal of using the GRADE Approach scale to rate health outcomes in this study is to eventually inform development of clinical practice guidelines for PA management in the CP population.

ACKNOWLEDGEMENTS

Research truly takes a team and it is my pleasure to have the opportunity to thank all those who have made the journey possible.

I would like to start off by thanking my supervisor and committee in their entirety. These past two years have been a series of learning opportunities and I am grateful to have such a supportive group of experts and mentors smooth, facilitate and encourage the process.

I would like to thank Dr. Gorter for your guidance, which greatly helped me understand the research process and encouraged me to become a more independent researcher. You have gone the extra mile to support me by providing opportunities to further my ideas and for that, I am extremely grateful. I learned tremendously from you as you demonstrated, through your actions, what excellent collaboration and communication in research looks like.

To my supervisory committee members, Dr. Kho, Dr. Timmons, and Dr. Verschuren, I am truly grateful for your support throughout this entire journey. I would like to thank Dr. Kho for encouraging me to think about good research practice, for your care in my work and my success, and for the large amount of time you spent discussing and editing my work. Thank you for believing in my potential through the constructive and insightful feedback you provided. Dr. Timmons, thank you for lending your vast knowledge of the field of PA, and for your clear and helpful feedback on my writing. Dr. Verschuren, thank you for your enthusiasm, positive and encouraging nature, and the field-specific lens you provided on my thesis. Your comments provided me great motivation in improving my work.

Thank you to Dr. Sarah Reedman, for helping in my scoping review. It was a pleasure working with and learning from you. Your passion for research in individuals with cerebral palsy was evident and incredibly energizing for me in the writing process.

I would like to give a special thanks to my student mentor, Dr. Patrick McPhee, who was instrumental in helping guide me in the research process. You have spent a countless amount of your own time reviewing my work. Thank you for your unique insights as an experienced researcher and for your ability to relate to what graduate students are going through.

Thank you to my colleagues, in particular Jael, Linda, and Sandra for being such a supportive group.

Last, but certainly not least, I would like to thank the staff in the Rehabilitation Science department. You have always offered support and guidance and have made my journey that much more enriching and enjoyable.

TABLE OF CONTENTS

DISCUSSION

LIST OF FIGURES AND TABLES

LIST OF SUPPLEMENTARY FIGURES AND TABLES

LIST OF APPENDICES

LIST OF ABBREVIATIONS

AACPDM	American Academy of Cerebral Palsy and Developmental Medicine
AM	Activity Monitor
BMI	Body Mass Index
CHERRIES	Checklist for Reporting Results of Internet E-Surveys
CP	Cerebral Palsy
EMG	Electromyography
FISSA	Fatigue Impact and Severity Self-Assessment
GMFCS	Gross Motor Function Classification System
HDL-C	High-Density Lipoprotein Cholesterol
HPA	Habitual Physical Activity
HZ	Height-for-Age Z score
IAACD	International Alliance of Academies for Childhood Disability
ICC	Intraclass Correlation
IQR	Interquartile Range
LPA	Light Physical Activity
MET	Metabolic Equivalent of Task
MVPA	Moderate-to-Vigorous Physical Activity
NR	Not Reported
PA	Physical Activity
PRISMA	Preferred Reporting Items for Systematic Reviews and Meta-Analyses
RMS	Root-Mean-Square
SB	Sedentary Behaviour
TDP	Typically Developing Peers

VA	Vertical Axis
VM	Vector Magnitude
WHO	World Health Organization

DECLARATION OF ACADEMIC ACHIEVEMENT

Student contribution to work:
This thesis is presented as a sandwich thesis and consists of a general introduction, two manuscripts (Chapters 1 and 2), an overall discussion and an overall conclusion. I, Julia Xiong, made significant original contributions to all co-authored studies in this thesis and am the first author for all of the included manuscripts.

Chapter 1
Operationalization, Measurement and Health Indicators of Sedentary Behaviour in Individuals with Cerebral Palsy: A Scoping Review

Contributions
Conceived and designed the research: Julia Xiong (JX), Michelle Kho (MEK), Jan Willem Gorter (JWG)
Screened title and abstract, and full-text articles: JX, Sarah Reedman (SR)
Extracted the data: JX (100% of the articles), SR (20% of the articles)
Drafted the manuscript: JX
Critical revision of the manuscript: JX, JWG, SR, MEK, Olaf Verschuren (OV), Brian Timmons (BWT)

Chapter 2
Identifying Critical and Important Health Outcomes for Physical Activity Promotion in Cerebral Palsy Using Perspectives from Clinicians and Researchers: A Conference-Based Survey

Contributions
Conceived and designed the research: JX, JWG, Patrick McPhee (PGM)
Acquired the data: JX
Analyzed and interpreted the data: JX
Drafted the manuscript: JX
Critical revision of the manuscript: JX, JWG, MEK, OV, BWT, PGM

INTRODUCTION

1.1 PREAMBLE

Individuals with cerebral palsy (CP) are at an increased risk of developing negative health outcomes [1]; this could be attributed to the joint and muscle pain, mobility problems, loss of function, and fatigue associated with the disorder, which lead to decreased physical activity (PA) levels and increased sedentary behaviour (SB) [2,3]. CP is not just a childhood disorder. A symposium on the life expectancy of individuals with CP indicates that their life expectancy is generally thought to match that of the typically developing population [4]. Both children and adults with CP experience health concerns such as being more prone to pain [5,6], and experiencing more fatigue [7,8], indicating that a lifelong approach to care is needed.

The purpose of this introduction is to provide information, relevant to the development of my thesis, on current knowledge of SB and PA in the typically developing population and individuals with CP across all age levels. I wish to provide research conducted in both populations to highlight some of the knowledge gaps in the field of physical behaviour in individuals with CP and illustrate the role of my thesis in addressing these gaps. An overview of the causes and secondary conditions of CP will be provided. I will then provide information on the burden of PA and SB in the typically developing population and in individuals with CP.

1.2 DEFINITION, PREVALENCE AND CAUSES OF CEREBRAL PALSY

CP is a group of non-progressive neurological disorder in motor and postural control. It affects affect approximately 2-3 per 1000 live births, making it leading cause of neurodevelopmental disability in North America [9,10]. The condition occurs due to permanent and non-progressive damage to developing brain, which can occur before, during or after birth

[11]. The typical areas in the brain damaged in the disorder are those that control movement and maintain posture and balance [12]. Since the problems associated with the motor disorder affects an individual across their lifespan, the overall goal of treatment is to improve quality of life and participation in life situations [13]. While there have been limited studies of life expectancy in this population, most children with CP can survive well into adulthood, even the most fragile children [14]. A study of a population register shows that 80% of those with mild impairments survive past age 58, which is similar to that for the general population [10]. For more severe impairments, mortality has shifted from childhood to early adulthood [10].

CP is frequently accompanied by impaired cognition, communication, and sensory perception, behavioural abnormalities, seizure disorders, musculoskeletal problems or a combination of these secondary problems [15]. The type and severity of the motor disorder is diagnosed from clinical signs of impaired motor control and functioning [16]. The impact of the motor impairment varies from a minimal effect on movement to the complete absence of the ability to move in a purposeful way [17]. CP is a description of clinical symptoms and does not indicate cause, pathology, or prognosis [17].

1.3 TYPES OF CEREBRAL PALSY AND CLASSIFICATIONS

The varying physical abilities of people with CP are categorized by the validated and clinically meaningful Gross Motor Function Classification System (GMFCS), a 5-level system which describes a range of functional ability. The impairments range from walking with some difficulties (GMFCS level I), to increasing levels of activity limitations (GMFCS level II), to requiring assistive mobility devices (GMFCS levels III), to severely limited walking even with assistive mobility devices (GMFCS level IV), and finally to a total loss of any form of independent mobility, even with specialized equipment (GMFCS level V) [18,19].

CP can also be described by the parts of body affected and by the way in which it affects movement. Bilateral CP indicates that both sides of the body are affected. There are two types of bilateral CP: 1) Quadriplegia indicates that all limbs and the trunk are affected[20]; 2) Diplegia indicates that both legs are affected [20]. Both arms may be affected but this occurs to a lesser extent [20]. Unilateral CP means that only one side of the body, the left or right side, is affected [20]. A form of unilateral CP is hemiplegia, in which one arm and one leg is affected [20].

CP can also be described by the type of motor impairment. Spastic CP is the most common, affecting 70-80% of those with CP [21]. The severity depends on how many of muscles are affected by stiffness, in which the overactive muscles override the less active ones [22]. In dyskinetic CP, individuals have involuntary movements, which could manifest in twisting or clenching fingers, or random movements [22]. The muscles could fluctuate between being stiff to having low tone, making it hard to maintain posture [22]. Ataxic CP results from damage to the part of the brain that helps fine-tune movement and maintain balance, which results in shaky movement [22]. Mixed CP indicates the individual could have a variety of symptoms that commonly manifests in more than one type of CP [22].

1.4 CONSEQUENCES IN CEREBRAL PALSY

Compared to their typically developing peers, the majority of individuals with CP have decreased muscle strength, muscle spasticity and decreased joint range of motion [23,24]. These physical impairments can significantly impact tasks of daily living such as dressing and walking [25–27]. A cycle of deconditioning in CP likely exists, in which low PA and increased SB may lead to reduced fitness and mobility, which further decreases PA and results in an increasingly sedentary lifestyle [18,19,28–30].

1. 5 PHYSICAL ACTIVITY AND SEDENTARY BEHAVIOUR IN THE TYPICALLY DEVELOPING POPULATION

The movement continuum is the continuity of behaviours based on their intensities [31]. Categorizations of activity along the movement continuum are termed “sleep”, “sedentary”, “light PA” (LPA), “moderate PA” and “vigorous PA” to describe increasing intensities of movement [31]. As the behaviours on the continuum interact in their impact on health, consideration of the range of behaviours is important in healthy living [31, 32].

The emerging field of research indicates that SB is characterized by both a postural component of sitting or reclining, and a low energy expenditure component [33]. On the other hand, in sports literature, SB often refers to an absence of some threshold of MVPA, in which participants are commonly characterized as sedentary when they are not meeting PA guidelines. The current consensus is to use “inactive” to describe the latter definition [33]. PA leads to a myriad of health benefits including a reduced risk of a heart attack, lower blood cholesterol, some cancers and all-cause mortality [34,35]. Physical inactivity has been identified by the World Health Organization (WHO) to be the fourth leading risk factor for global mortality [36]. Currently, there are several guidelines specifying suggested PA and SB times in the typically developing population. These include the guidelines set out by the WHO [37], the Canadian PA guidelines for adults [38], and the Canadian Society of Exercise Physiology (CSEP)’s 24-Hour Movement guidelines [39,40], a set of comprehensive guidelines for children that integrate recommendations for sleep, SB and PA. The 24-hour movement guidelines and Canadian PA guidelines suggest at least 60 minutes of MVPA for children and adolescents per day and at least 150 minutes of MVPA for adults per week [38,41]. The WHO guidelines suggest 150 minutes of moderate-intensity or 75 minutes of vigorous intensity aerobic activity per week for adults and

60 minutes of MVPA per day for children [37]. Globally, 1 is 4 adults are not meeting these guidelines [37].

For both children and adults in the typically developing population, the standard operationalized definition of SB has both a postural component of sitting, reclining or lying and an intensity component with an energy expenditure of ≤1.5 metabolic equivalents of task (METs) [33]. A MET is a ratio of the energy consumption during postures or activities against a reference resting basal metabolic rate (i.e., 3.5 $mL \cdot kg^{-1} \cdot min^{-1}$ or 1 $kcal \cdot kg^{-1} \cdot h^{-1}$) [42]. Moderate activity in the typically developing population is characterized by an energy expenditure of 3.0 to 6.0 METs and includes activities such as walking at brisk pace or cycling on level terrain or with few hills [43]. Vigorous activity in the typically developing population is characterized by an energy expenditure of greater than 6.0 METs and includes activities such as jogging, running or bicycling on steep, uphill terrain [43].

Evidence suggests that SB and PA are two different constructs and present separate and distinct risk factors for chronic diseases, including cancer, cardiovascular disease and diabetes [44]. This means that even if an individual is performing the recommended MVPA, they could still be spending the rest of their time being sedentary. The implication is that individuals can perform the recommended level of MVPA and reap the resulting health benefits such as reduced incidences of chronic pain, fatigue and osteoporosis, yet still be affected by the negative health outcomes associated with SB such as those affecting the cardiovascular and metabolic systems [31-33]. High levels of PA may be able to account for prolonged SB; a large meta-synthesis of more than one million adults has shown that high amounts of PA can attenuate or even eliminate the risk of all-cause mortality attributed to prolonged SB [45]. Prolonged sedentary time and infrequent breaks interrupting it have stronger associations with negative markers of metabolic risk, in particular waist circumference, body mass index (BMI), plasma glucose and triglycerides compared to increased breaks to sedentary bouts [35].

A standard operationalized definition for SB is provided by the Sedentary Behaviour Research Network (SBRN) [33]. The SBRN is the only organization for researchers to study topics surrounding SB, with a focus on disseminating this research to the academic community and the general public [46]. The formal call for the adoption of the standardized operationalized definition of SB was in 2012 through a letter to the editor set forth by the SBRN, which calls for journal editors to require all accepted manuscripts from thereon to use the definition [33]; The consensus of the operationalized definition of SB had the agreement of an international consortium of researchers [33]. The Terminology Consensus Project was a later initiative by the SBRN in efforts to provide standardized terminology for SB and related terms, including stationary behaviour, SB, standing, screen time, non-screen-based sedentary time, sitting, reclining, lying, and SB pattern [46].

The guidelines on SB set out by the WHO includes a recommendation of at most 60 minutes in SB per day for children under 5 [37]. The 24-Hour movement guidelines suggest no more than 2 hours of SB per day for children [47]. As of March 2020, the WHO is working on draft guidelines to include recommendations for both PA and SB for children, adolescents, adults and older adults [48]. CSEP is also currently developing 24-Hour Movement guidelines, which incorporates recommendations for SB, for adults and older adults[49]. The fact that there is no SB guideline development for some age groups even in typically developing population up until recently is partly attributed to lack of SB research. These developments highlight the paucity of evidence in SB research up until recently, an increase which could be attributed to the SBRN adoption of a standard operationalized definition of the SB construct [33], providing researchers with more guidance in studying this field.

1.6 SEDENTARY BEHAVIOUR IN CEREBRAL PALSY

Individuals with CP spend 76-99% of their waking time seated [50–54]. The patterns of SB differ for individuals with CP; children with CP not only spend more time in SB, but also have a reduced frequency of breaks from sedentary time compared to their typically developing peers [52]. The mobility limitations in people with CP may prevent them from interrupting sedentary bouts, which may predispose these individuals to greater cardiovascular and metabolic risks [35].

For individuals with CP who experience challenges with MVPA, efforts to replace SB with LPA throughout the day may be more feasible [55]. This may especially be the case for individuals with more severe CP in GMFCS levels IV and V due to their severe motor impairments. In addition, sitting time, one of the postures that is considered SB, increases with GMFCS level [50,53]. While the 24-Hour Movement guidelines indicates that the recommendations may be appropriate for children and youth with a disability or medical condition, including CP, the guidelines state, "a health professional should be consulted for additional guidance" [41]. The developers of the Canadian 24-hour movement guidelines published a study which explored the perceptions of inclusivity of these guidelines for children and youth with disabilities through interviewing parents of these children [56]. Parents indicated that the guidelines were not inclusive to their children's abilities and needs [56]. This suggests a gap for more inclusive guidelines for individuals with disabilities, including CP. While SB guidelines, and intervention studies to inform the evidence profile of a set of more inclusive guidelines, specific to the CP population, could be a goal, there is a paucity of research on these interventions [57]. A recent scoping review, which evaluates interventions for decreasing SB in children with disabilities, including CP, found limited evidence on interventions aimed to reduced SB in children with CP (n=2) [57]. Rather, the focus has traditionally been on increasing PA in this population [58]. The two intervention studies to reduce SB in children with CP specifically, found through this scoping review, did not support effectiveness of strategies to reduce SB in children with CP [59,60]. The reason for the limited research in the field could be attributed to the lack of operationalized definition and measurement challenges of SB for individuals with CP.

The scoping review on SB in children with physical disabilities highlighted some of these measurement challenges [57]. The review found that research should include the validation of accelerometers for measurement of SB in children who use wheelchairs, since accelerometers may not capture extraneous limb movements; it further stated that SB have been validated primarily with ambulatory children with CP, i.e., those with GMFCS levels I–III [61–63]. The cut points, location, wear time, and valid wear time varied across studies of children with physical disabilities [57]. For example, accelerometer cut points for SB ranged from 41 to 820 counts per minute [57]. These cut-points include thresholds validated in the typically developing population of <41 counts per minute and <100 counts per minute and those used for specific populations and age groups, including children with CP in different GMFCS levels [57]; Some researchers use the right hip or lower back, regardless of the side of the body affected by the disability, while others placed the accelerometer on the least affected side [57]. Wear time and criteria for valid wear time also varied, with instructions ranging from 3 to 8 days and valid wear time ranging from 1 to 5 days [57]. As there is considerable variability in measurement of SB for children with physical disabilities, including CP [57], there is a need to map the extent of measurement of SB for people with CP across all ages.

There is currently no operationalized definition of SB for individuals with CP. It is unknown whether current definitions of SB for the typically developing population [33] applies to individuals with CP, who have neuromuscular impairments and variable mobility restrictions [58,64]. These impairments and restrictions likely result in different energy expenditure and muscle activities between different levels of severity, even within similar postures [58,64]. Furthermore, variable energy expenditure could also be attributed to significant spasticity and involuntary movements in some individuals with CP [65,66]. These individuals may require increased effort to maintain postural control in independent sitting compared with children without disabilities [65,67,68], impacting the energy expenditure in typically sedentary postures. It has been hypothesized that the negative health effects of SB are due to a lack of contraction of skeletal muscles that characterizes the behaviour, which consequently contributes to decreased clearance of plasma triglycerides and oral glucose for plasma [35]. Individuals with CP display atypical muscle tone (ie, spasticity, hypotonia, athetosis, ataxia, or a combination) and challenges with muscle co-contraction, balance, and coordination, meaning these individuals may require different levels of muscle contraction in typically SB, such as sitting [64]. Furthermore, some individuals with CP require support while sitting, which may reduce the muscle contractions required in these postures [69]. These factors point towards the need to develop the definition of SB, through various components, including posture, energy expenditure, oxygen consumption and muscle activation, specifically for individuals with CP.

1.7 PHYSICAL ACTIVITY IN CEREBRAL PALSY

While globally, inactivity has been on the rise, these trends are exacerbated in individuals with CP. In fact, a systematic reviews of six studies shows that across all levels of motor function in children up to age 18, individuals with CP participate in 13% to 53% less habitual PA than their typically developing peers [70]. Lower level of PA renders individuals with CP especially susceptible to the negative health effects of inactivity [71]. These trends are concerning because reduced PA in the typically developing population is associated with a higher risk of developing multimorbidity, including cardiovascular disease [72].

Engaging in PA is challenging for individuals with CP for various reasons such as condition-related factors, including difficulties with movement [11]; personal factors, including lack of information about the importance of PA [73]; and environmental factors, including lack of appropriate equipment and access to fitness venues [74]. Nevertheless, PA in this population is crucial; in a consensus statement reporting prospective studies examining the correlation between health and PA among people with disabilities, activity level was found to be highly associated with years of survival in those with physical disabilities [75]. A narrative review showed that adolescents with CP exhibit a gradual decline in strength and fitness throughout adulthood [23]. These changes are hypothesized to result in decreased walking ability, despite ambulation acquired in adolescence [23]. These findings point towards the need for a lifespan approach to PA promotion.

PA promotion in the CP population may require different management plans than their typically developing peers. When discussing future directions of the Canadian Physical Activity Guidelines, Tremblay et al. note efforts are currently underway to develop guidelines for those living with chronic conditions, although not specific to any one chronic condition [47]. Given the heterogeneity in CP, PA promotion for the population as a whole, across ages and across severity levels, is likely a challenge for clinicians. Hence the call was made for a clinical care pathway on PA promotion in individuals with CP by the American Academy for Cerebral Palsy and Developmental Medicine (AACPDM) to guide clinicians in providing management strategies carefully tailored, timed and integrated into health services.

1.8 THE KNOWLEDGE GAP

While a letter to the editor by the SBRN provided a standard operationalized definition of SB in the typically developing population [33], no such standard definition exists for SB in individuals with CP. To begin designing interventions to decrease SB in individuals with CP, it is important to first operationalize SB in this population. As part of the operationalization process, the construct of SB is translated into a measurable variable. SB can be operationalized in several ways, such as by the sitting posture or the amount of time spent with low energy expenditure [33]. Even when the construct of SB is operationally defined to measurable variables, there could be large variability in the types of tools to measure SB. In one study that explored the effects of different operationalizations of SB in individuals with chronic stroke, it was found that SB results differed significantly based on measurement of different operationalizations of SB from combined postural and intensity data, to just postural data or just intensity data [76]. As the results of SB may vary based on the operationalization of SB in CP, it is crucial to begin the process of developing operationalized definitions of SB in this population. In this way, researchers and clinicians in the field of CP can discuss and capture the construct in a standard manner across studies (i.e., operationalized definition, measurement and reporting) for comparison of SB in the CP population. These findings can in turn help expand the currently limited research in this field to inform future guidelines to reduce SB in this population.

The negative health effects of physical inactivity point towards the need for PA guidelines with specific recommendations for individuals with CP [71]. The development of recent clinical practice guidelines, including the Canadian 24-hour movement guidelines, have followed the current standard for guideline development, the Appraisal of Guidelines for Research Evaluation (AGREE) II instrument, with the quality of evidence assessed using the Grading of Recommendations Assessment, Development, and Evaluation (GRADE) Approach [39,40]. The GRADE Approach is considered the standard for assessing the quality of evidence used in guideline development because it has marked improvements over previous system for rating the quality of evidence. The advantages of the GRADE Approach include an international collaborative effort in its development, explicit criteria for upgrading or downgrading the quality of evidence. and separation of the quality of evidence and the strength of recommendations [77]. The steps in the Approach include preliminary classification of health outcomes by experts and patients as important, critical or not important, before reviewing evidence [78].

In following the Approach, the evidence profiles used in the development of the 24-Hour Movement guidelines were driven by health indicators chosen and rated for their importance, as related to PA, by experts in movement behaviours [79,80]. For children and youth, the eleven critical or important health indicators are: body composition, cardiometabolic biomarkers, physical fitness, behavioural conduct/pro-social behaviour, cognition/academic achievement, quality of life/well-being, injuries, bone health, motor skill development, psychological distress, self-esteem [79]. For the toddlers, the eight critical or important health indicators are: adiposity, motor development, psychosocial health, cognitive development, fitness, bone and skeletal health, cardiometabolic health, and risks/harm [80]. There is currently no information available on the importance of health outcomes, relevant to PA, for individuals with CP. This information would impact the evidence profile for developing clinical practice guidelines in PA management for this population.

Individuals with CP may have different health concerns that are associated with or relevant to PA compared to those in the typically developing population; for example, two cross sectional studies with 111 children with CP and 7348 adults with CP show that individuals with CP have higher prevalences of mental illnesses, including depression and anxiety, compared to the typically developing population [81,82]. A literature review shows that children with CP often have feeding problems, which include oral motor problems, swallowing difficulties and airway protection problems, difficulties with proper positioning, requiring assistance with feeding, and prolonged feeding times [83]. The ability to swallow food and food processing problems affect 30 to 40% of children with CP and are the main contributors to inadequate food intake to meet metabolic demands for the population [83]. In a cross-sectional study looking at sleep quality in a group of 41 children with CP and 91 typically developing controls, children with CP are more likely to have problems with the initiation and maintenance of sleep, sleep-wake transition disorders and excessive sleepiness and arousal disorders compared to their typically developing peers [84]. These concerns, which are unique to or more prevalent in individuals with CP, indicate that for them, PA guideline development may require an evidence profile that examines different health outcomes compared to that of the typically developing population.

Just because an individual is meeting PA guideline recommendations, this does not prevent them from spending the rest of their day being sedentary. This suggests that the benefits conferred by PA could be negated by prolonged periods of SB and that these two constructs need to be explored separately [85]. In this thesis, I have begun to address some of the gaps identified.

I do so by exploring two broad questions regarding firstly, the operationalization of SB and, secondly, PA health outcomes that are important in individuals with CP across all ages. Each question is answered separately, and the results are described as a whole in the final discussion.

1.9 STUDY OBJECTIVES AND HYPOTHESES

The overarching objective of this thesis is to explore and better understand SB and identify the important and critical health outcomes for PA from a list of pre-determined health outcomes in individuals with CP. Together, this information can support the idea of a holistic approach to activity promotion by studying the range of physical behaviours across the movement continuum.

The objective of Chapter 1 is to map the existing research evidence, through a scoping review, on the operationalization of SB in individuals with CP. Future research would include establishing a consensus on a standard definition of SB in individuals with CP, similar to the initiative by the SBRN in typically developing individuals [46].

The objective of Chapter 2 is to conduct a conference-based survey to identify important and critical health outcomes relevant to PA, from a list of pre-determined outcomes, for individuals with CP. The rating of importance of these outcomes is based on the opinions of clinicians and researchers. The survey could help decide the health outcomes to include in the evidence profile for future guideline development in PA promotion for this population.

1.10 REFERENCES

1. Slot W, Roebroeck M, Nieuwenhuijsen C, Bergen M, Stam H, Burdorf A, et al. Cardiovascular disease risk in adults with spastic bilateral cerebral palsy. J Rehabil Med. 2013;45(9):866–72.

2. Carter S, Hartman Y, Holder S, Thijssen DH, Hopkins ND. Sedentary Behavior and Cardiovascular Disease Risk: Mediating Mechanisms. Exerc Sport Sci Rev. 2017 Apr;45(2):80–6.

3. Nystoriak MA, Bhatnagar A. Cardiovascular Effects and Benefits of Exercise. Front Cardiovasc Med. 2018 Sep 28;5:135.

4. Hutton JL. Outcome in cerebral palsy: life-expectancy. Paediatr Child Health. 2008 Sep;18(9):419–22.

5. Hirsh AT, Kratz AL, Engel JM, Jensen MP. Survey Results of Pain Treatments in Adults with Cerebral Palsy: Am J Phys Med Rehabil. 2011 Mar;90(3):207–16.

6. Tremblay MS, Colley RC, Saunders TJ, Healy GN, Owen N. Physiological and health implications of a sedentary lifestyle. Appl Physiol Nutr Metab. 2010 Dec;35(6):725–40.

7. Jahnsen R, Villien L, Stanghelle JK, Holm I. Fatigue in adults with cerebral palsy in Norway compared with the general population. Dev Med Child Neurol [Internet]. 2003 May [cited 2020 Jul 30];45(05). Available from: http://doi.wiley.com/10.1017/S0012162203000562

8. Balemans ACJ, van Wely L, Becher JG, Dallmeijer AJ. Longitudinal Relationship Among Physical Fitness, Walking-Related Physical Activity, and Fatigue in Children With Cerebral Palsy. Phys Ther. 2015 Jul 1;95(7):996–1005.

9. Kirby RS, Wingate MS, Van Naarden Braun K, Doernberg NS, Arneson CL, Benedict RE, et al. Prevalence and functioning of children with cerebral palsy in four areas of the United States in 2006: A report from the Autism and Developmental Disabilities Monitoring Network. Res Dev Disabil. 2011 Mar;32(2):462–9.

10. Paneth N, Hong T, Korzeniewski S. The Descriptive Epidemiology of Cerebral Palsy. Clin Perinatol. 2006 Jun;33(2):251–67.

11. Rosenbaum P, Paneth N, Leviton A, Goldstein M, Bax M, Damiano D, et al. A report: the definition and classification of cerebral palsy April 2006. Dev Med Child Neurol Suppl. 2007 Feb;109:8–14.

12. Niiler TA. Assessing Dynamic Balance in Children with Cerebral Palsy. In: Miller F, Bachrach S, Lennon N, O'Neil M, editors. Cerebral Palsy [Internet]. Cham: Springer International Publishing; 2018. p. 1–32. Available from: https://doi.org/10.1007/978-3-319-50592-3_49-1

13. Sewell MD, Eastwood DM, Wimalasundera N. Managing common symptoms of cerebral palsy in children. BMJ Online. 2014;349:g5474.

14. L Jr H. Pediatric Cerebral Palsy Life Expectancy: Has Survival Improved Over Time? Pediatr Ther [Internet]. 2013 [cited 2019 Apr 4];03(01). Available from: https://www.omicsonline.org/pediatric-cerebral-palsy-life-expectancy-has-survival-improved-over-time-2161-0665.1000146.php?aid=14350

15. Aisen ML, Kerkovich D, Mast J, Mulroy S, Wren TA, Kay RM, et al. Cerebral palsy: clinical care and neurological rehabilitation. Lancet Neurol. 2011 Sep;10(9):844–52.

16. Velde, Morgan, Novak, Tantsis, Badawi. Early Diagnosis and Classification of Cerebral Palsy: An Historical Perspective and Barriers to an Early Diagnosis. J Clin Med. 2019 Oct 3;8(10):1599.

17. Stanley FJ, Blair E, Alberman ED. Cerebral palsies: epidemiology and causal pathways. London : New York: Mac Keith ; Cambridge University Press; 2000. 251 p. (Clinics in developmental medicine).

18. Palisano RJ, Rosenbaum P, Bartlett D, Livingston MH. Content validity of the expanded and revised Gross Motor Function Classification System. Dev Med Child Neurol. 2008 Oct;50(10):744–50.

19. Palisano R, Rosenbaum P, Walter S, Russell D, Wood E, Galuppi B. Development and reliability of a system to classify gross motor function in children with cerebral palsy. Dev Med Child Neurol. 2008 Sep 29;39(4):214–23.

20. Cans C. Surveillance of cerebral palsy in Europe: a collaboration of cerebral palsy surveys and registers. Dev Med Child Neurol. 2007 Feb 13;42(12):816–24.

21. Gladstone M. A review of the incidence and prevalence, types and aetiology of childhood cerebral palsy in resource-poor settings. Ann Trop Paediatr. 2010;30(3):181–96.

22. Sankar C, Mundkur N. Cerebral palsy-definition, classification, etiology and early diagnosis. Indian J Pediatr. 2005 Oct;72(10):865–8.

23. Verschuren O, Smorenburg ARP, Luiking Y, Bell K, Barber L, Peterson MD. Determinants of muscle preservation in individuals with cerebral palsy across the lifespan: a narrative review of the literature: Muscle preservation in individuals with cerebral palsy. J Cachexia Sarcopenia Muscle. 2018 Jun;9(3):453–64.

24. Darrah J, Wiart L, Gorter JW, Law M. Stability of Serial Range-of-Motion Measurements of the Lower Extremities in Children With Cerebral Palsy: Can We Do Better? Phys Ther. 2014 Jul 1;94(7):987–95.

25. Johnston MV. Plasticity in the developing brain: implications for rehabilitation. Dev Disabil Res Rev. 2009;15(2):94–101.

26. Van BR, Miller MD, Russo RN, Murchland S, Crotty M. Activities of daily living in children with hemiplegic cerebral palsy: a cross-sectional evaluation using the Assessment of Motor and Process Skills. Dev Med Child Neurol. 2007 Feb 13;48(9):723–7.

27. Verschuren O, Peterson MD, Balemans ACJ, Hurvitz EA. Exercise and physical activity recommendations for people with cerebral palsy. Dev Med Child Neurol. 2016 Aug;58(8):798–808.

28. Hombergen SP, Huisstede BM, Streur MF, Stam HJ, Slaman J, Bussmann JB, et al. Impact of Cerebral Palsy on Health-Related Physical Fitness in Adults: Systematic Review. Arch Phys Med Rehabil. 2012 May;93(5):871–81.

29. Opheim A, Jahnsen R, Olsson E, Stanghelle JK. Walking function, pain, and fatigue in adults with cerebral palsy: a 7-year follow-up study. Dev Med Child Neurol. 2009 May;51(5):381–8.

30. Van Wely L, Balemans AC, Becher JG, Dallmeijer AJ. The effectiveness of a physical activity stimulation programme for children with cerebral palsy on social participation, self-perception and quality of life: a randomized controlled trial. Clin Rehabil. 2014 Oct;28(10):972–82.

31. Canadian 24-Hour Movement Guidelines - Glossary [Internet]. [cited 2020 Jul 30]. Available from: https://www.csep.ca/en/guidelines/glossary-2017

32. Chaput J-P, Carson V, Gray C, Tremblay M. Importance of All Movement Behaviors in a 24 Hour Period for Overall Health. IJERPH. 2014 Dec 4;11(12):12575–81.

33. Sedentary Behaviour Research Network. Letter to the Editor: Standardized use of the terms “sedentary” and “sedentary behaviours.” Appl Physiol Nutr Metab. 2012 Jun;37(3):540–2.

34. Petersen C, Bauman A, Grønbæk M, Helge J, Thygesen L, Tolstrup JS. Total sitting time and risk of myocardial infarction, coronary heart disease and all-cause mortality in a prospective cohort of Danish adults. Int J Behav Nutr Phys Act. 2014;11(1):13.

35. Healy GN, Dunstan DW, Salmon J, Cerin E, Shaw JE, Zimmet PZ, et al. Breaks in Sedentary Time: Beneficial associations with metabolic risk. Diabetes Care. 2008 Apr 1;31(4):661–6.

36. Mathers C, Stevens G, Mascarenhas M. Global health risks: mortality and burden of disease attributable to selected major risks. World Health Organization; 2009.

37. World Health Organization. Global recommendations on physical activity for health. [Internet]. 2010 [cited 2020 Jul 5]. Available from: http://www.ncbi.nlm.nih.gov/books/NBK305057/

38. Canadian Physical Activity Guidelines for Adults (18-64 years) [Internet]. CSEP | SCPE. [cited 2020 Jul 30]. Available from: https://csepguidelines.ca

39. Tremblay MS, Chaput J-P, Adamo KB, Aubert S, Barnes JD, Choquette L, et al. Canadian 24-Hour Movement Guidelines for the Early Years (0–4 years): An Integration of Physical Activity, Sedentary Behaviour, and Sleep. BMC Public Health. 2017 Nov;17(S5):874.

40. Tremblay MS, Carson V, Chaput J-P, Connor Gorber S, Dinh T, Duggan M, et al. Canadian 24-Hour Movement Guidelines for Children and Youth: An Integration of Physical Activity, Sedentary Behaviour, and Sleep. Appl Physiol Nutr Metab. 2016 Jun;41(6 (Suppl. 3)):S311–27.

41. Canadian 24-Hour Movement Guidelines : CSEP | SCPE [Internet]. [cited 2020 Jul 5]. Available from: https://csepguidelines.ca/

42. Boccalandro F. Exercise Stress Testing. In: Cardiology Secrets [Internet]. Elsevier; 2010 [cited 2020 Jun 28]. p. 54–9. Available from: https://linkinghub.elsevier.com/retrieve/pii/B9780323045254000071

43. Ainsworth BE, Haskell WL, Whitt MC, Irwin ML, Swartz AM, Strath SJ, et al. Compendium of Physical Activities: an update of activity codes and MET intensities: Med Sci Sports Exerc. 2000 Sep;32(Supplement):S498–516.

44. Saunders TJ, Chaput J-P, Tremblay MS. Sedentary Behaviour as an Emerging Risk Factor for Cardiometabolic Diseases in Children and Youth. Can J Diabetes. 2014 Feb;38(1):53–61.

45. Ekelund U, Steene-Johannessen J, Brown WJ, Fagerland MW, Owen N, Powell KE, et al. Does physical activity attenuate, or even eliminate, the detrimental association of sitting time with mortality? A harmonised meta-analysis of data from more than 1 million men and women. The Lancet. 2016 Sep;388(10051):1302–10.

46. on behalf of SBRN Terminology Consensus Project Participants, Tremblay MS, Aubert S, Barnes JD, Saunders TJ, Carson V, et al. Sedentary Behavior Research Network (SBRN) – Terminology Consensus Project process and outcome. Int J Behav Nutr Phys Act. 2017 Dec;14(1):75.

47. CSEP. [cited 2020 Jul 5]. Available from: https://csep.ca/news.asp?a=view&id=156

48. Public consultation on the draft WHO Guidelines on physical activity and sedentary behaviour for children and adolescents, adults and older adults 2020 [Internet]. [cited 2020 Jul 5]. Available from: https://www.who.int/news-room/articles-detail/public-consultation-on-the-draft-who-guidelines-on-physical-activity-and-sedentary-behaviour-for-children-and-adolescents-adults-and-older-adults-2020

49. Kaleigh Maclaren. Development for the world's first 24-Hour Movement Guidelines for Adults and Older Adults underway [Internet]. [cited 2020 Jul 30]. Available from: https://csep.ca/news.asp?a=view&id=215&pageToView=1

50. Claridge EA, Mcphee PG, Timmons BW, Martin Ginis KA, Macdonald MJ, Gorter JW. Quantification of Physical Activity and Sedentary Time in Adults with Cerebral Palsy: Med Sci Sports Exerc. 2015 Aug;47(8):1719–26.

51. Nooijen CF, Slaman J, Stam HJ, Roebroeck ME, Berg-Emons R, Learn2Move Research Group. Inactive and sedentary lifestyles amongst ambulatory adolescents and young adults with cerebral palsy. J NeuroEngineering Rehabil. 2014;11(1):49.

52. Obeid J, Balemans ACJ, Noorduyn SG, Gorter JW, Timmons BW. Objectively Measured Sedentary Time in Youth With Cerebral Palsy Compared With Age-, Sex-, and Season-Matched Youth Who Are Developing Typically: An Explorative Study. Phys Ther. 2014 Aug 1;94(8):1163–7.

53. Shkedy Rabani A, Harries N, Namoora I, Al-Jarrah MD, Karniel A, Bar-Haim S. Duration and patterns of habitual physical activity in adolescents and young adults with cerebral palsy. Dev Med Child Neurol. 2014 Jul;56(7):673–80.

54. Ryan JM, Crowley VE, Hensey O, Broderick JM, McGahey A, Gormley J. Habitual physical activity and cardiometabolic risk factors in adults with cerebral palsy. Res Dev Disabil. 2014 Sep;35(9):1995–2002.

55. Taylor NF. Sedentary behaviour in adolescents and young adults with cerebral palsy. Dev Med Child Neurol. 2014 Jul;56(7):609–10.

56. Handler L, Tennant EM, Faulkner G, Latimer-Cheung AE. Perceptions of Inclusivity: The Canadian 24-Hour Movement Guidelines for Children and Youth. Adapt Phys Act Q. 2019 Jan;36(1):1–18.

57. Ganz F, Hammam N, Pritchard L. Sedentary behavior and children with physical disabilities: a scoping review. Disabil Rehabil. 2020 Mar 2;1–13.

58. Verschuren O, Darrah J, Novak I, Ketelaar M, Wiart L. Health-Enhancing Physical Activity in Children With Cerebral Palsy: More of the Same Is Not Enough. Phys Ther. 2014 Feb 1;94(2):297–305.

59. Lauruschkus K, Hallström I, Westbom L, Tornberg Å, Nordmark E. Participation in physical activities for children with cerebral palsy: feasibility and effectiveness of physical activity on prescription. Arch Physiother. 2017 Dec;7(1):13.

60. Maher CA, Williams MT, Olds T, Lane AE. An internet-based physical activity intervention for adolescents with cerebral palsy: a randomized controlled trial: Internet-based Physical Activity Program for Adolescents with CP. Dev Med Child Neurol. 2010 Jan 28;52(5):448–55.

61. Ryan J, Walsh M, Gormley J. Ability of RT3 Accelerometer Cut Points to Detect Physical Activity Intensity in Ambulatory Children With Cerebral Palsy. Adapt Phys Act Q. 2014 Oct;31(4):310–24.

62. Trost SG, Fragala-Pinkham M, Lennon N, O'Neil ME. Decision Trees for Detection of Activity Intensity in Youth with Cerebral Palsy: Med Sci Sports Exerc. 2016 May;48(5):958–66.

63. Oftedal S, Bell KL, Davies PSW, Ware RS, Boyd RN. Validation of Accelerometer Cut Points in Toddlers with and without Cerebral Palsy: Med Sci Sports Exerc. 2014 Sep;46(9):1808–15.

64. Innes J, Darrah J. Sedentary Behavior: Implications for Children With Cerebral Palsy. Pediatr Phys Ther. 2013;25(4):402–8.

65. Bell KL, Davies PS. Energy expenditure and physical activity of ambulatory children with cerebral palsy and of typically developing children. Am J Clin Nutr. 2010 Aug 1;92(2):313–9.

66. Johnson RK, Hildreth HG, Contompasis SH, Goran MI. Total energy expenditure in adults with cerebral palsy as assessed by doubly labeled water. J Am Diet Assoc. 1997 Sep;97(9):966–70.

67. Walker JL, Bell KL, Boyd RN, Davies PS. Energy requirements in preschool-age children with cerebral palsy. Am J Clin Nutr. 2012 Dec 1;96(6):1309–15.

68. Carlberg EB, Hadders-Algra M. Postural dysfunction in children with cerebral palsy: some implications for therapeutic guidance. Neural Plast. 2005;12(2–3):221–72.

69. Balemans ACJ, Houdijk H, Koelewijn GR, Piek M, Tubbing F, Visser-Meily A, et al. A Comparison of the Physiology of Sedentary Behavior and Light Physical Activity in Adults With and Without a Physical Disability. J Phys Act Health. 2019 Oct 1;16(10):894–901.

70. Carlon SL, Taylor NF, Dodd KJ, Shields N. Differences in habitual physical activity levels of young people with cerebral palsy and their typically developing peers: a systematic review. Disabil Rehabil. 2013 Apr;35(8):647–55.

71. Bjornson KF, Belza B, Kartin D, Logsdon R, McLaughlin J, Thompson EA. The Relationship of Physical Activity to Health Status and Quality of Life in Cerebral Palsy: Pediatr Phys Ther. 2008;20(3):247–53.

72. Mcphee PG, Claridge EA, Noorduyn SG, Gorter JW. Cardiovascular disease and related risk factors in adults with cerebral palsy: a systematic review. Dev Med Child Neurol [Internet]. 2018 Sep 17 [cited 2019 Apr 9]; Available from: http://doi.wiley.com/10.1111/dmcn.14028

73. Buffart LM, Westendorp T, Van Den Berg-Emons RJ, Stam HJ, Roebroeck ME. Perceived barriers to and facilitators of physical activity in young adults with childhood-onset physical disabilities. J Rehabil Med. 2009;41(11):881–5.

74. Rimmer JH, Riley B, Wang E, Rauworth A, Jurkowski J. Physical activity participation among persons with disabilities: barriers and facilitators. Am J Prev Med. 2004;26(5):419–25.

75. Cooper RA, Quatrano LA. Research on physical activity and health among people with disabilities: a consensus statement. J Rehabil Res Dev. 1999;36(2).

76. Fanchamps MHJ, de Kam D, Sneekes EM, Stam HJ, Weerdesteyn V, Bussmann JBJ. Effect of different operationalizations of sedentary behavior in people with chronic stroke. Disabil Rehabil. 2018 Nov 26;1–7.

77. Guyatt GH, Oxman AD, Vist GE, Kunz R, Falck-Ytter Y, Alonso-Coello P, et al. GRADE: an emerging consensus on rating quality of evidence and strength of recommendations. BMJ. 2008;336(7650):924–6.

78. Schünemann H. The GRADE handbook. Cochrane Collaboration; 2013.

79. Poitras VJ, Gray CE, Borghese MM, Carson V, Chaput J-P, Janssen I, et al. Systematic review of the relationships between objectively measured physical activity and health indicators in school-aged children and youth. Appl Physiol Nutr Metab. 2016 Jun;41(6 (Suppl. 3)):S197–239.

80. Carson V, Lee E-Y, Hewitt L, Jennings C, Hunter S, Kuzik N, et al. Systematic review of the relationships between physical activity and health indicators in the early years (0-4 years). BMC Public Health. 2017 Nov;17(S5):854.

81. Whitney DG, Warschausky SA, Ng S, Hurvitz EA, Kamdar NS, Peterson MD. Prevalence of Mental Health Disorders Among Adults With Cerebral Palsy: A Cross-sectional Analysis. Ann Intern Med. 2019 Sep 3;171(5):328.

82. Whitney DG, Warschausky SA, Peterson MD. Mental health disorders and physical risk factors in children with cerebral palsy: a cross-sectional study. Dev Med Child Neurol. 2019 May;61(5):579–85.

83. Rempel G. The Importance of Good Nutrition in Children with Cerebral Palsy. Phys Med Rehabil Clin N Am. 2015 Feb;26(1):39–56.

84. Sandella DE, O'Brien LM, Shank LK, Warschausky SA. Sleep and quality of life in children with cerebral palsy. Sleep Med. 2011 Mar;12(3):252–6.

85. Ryan JM, Forde C, Hussey JM, Gormley J. Comparison of Patterns of Physical Activity and Sedentary Behavior Between Children With Cerebral Palsy and Children With Typical Development. Phys Ther. 2015 Dec 1;95(12):1609–16.

CHAPTER 1: Operationalization, Measurement and Health Indicators of Sedentary Behaviour in Individuals with Cerebral Palsy: A Scoping Review

Abstract

Background: Too much time and infrequent bouts of interrupting sedentary behaviour (SB) has negative health effects independent of performing moderate-to-vigorous physical activity. For individuals with cerebral palsy who have different muscle tones and activity compared to the typically developing population, it is uncertain whether the definition of SB provided by the SB Research Network (SBRN) also applies. We conducted a scoping review to explore the definition and measurement of SB in individuals with CP.

Methods: We searched 5 databases from January 1, 2011 to December 2, 2019, inclusive for primary studies of experimental, qualitative, longitudinal or observational designs measuring SB or postures typically characterized as sedentary (i.e., sitting, reclining, lying). Two independent reviewers screened titles, abstracts, and full texts for inclusion. 20% of data extraction was performed by two reviewers, and 80% by one reviewer.

Results: We screened 951 unique citations, reviewed 97 full-text publications, selected 37 studies and found 2 studies from a hand-search of the references of included articles. In total, we included 39 studies, out of which 5 studies addressed more than one research question. The operationalization of SB was done by studies through the measurements of muscle activation, energy expenditure or oxygen consumption in typically sedentary postures (i.e., sitting, reclining, lying) (n=7), and through the thresholds and postures used by accelerometers and activity monitors to measure time spent in SB (n=22). Variations existed for the thresholds used by tools that measure SB, although the postures that constitute as SB were consistently operationalized as sitting and lying. Of the 6 studies that measured energy expenditure of sitting and lying, 5 studies found energy expenditure to be ≤1.5 METs. The studies that evaluated the validity or reliability

of tools for measuring SB or typically sedentary postures had variations in population, instrument and cut-points (n=15).

Conclusions: The definition of ≤1.5 METs applies to sitting and lying in individuals with CP. Sitting and lying are considered postures in measuring SB. There is large variability in the tools used to measure SB in individuals with CP. Consensus on the definition of SB and its reporting is needed.

Introduction

Excessive total time spent in sedentary behaviour (SB) and infrequent interruptions of sedentary bouts have negative health consequences independent of time spent performing moderate-to-vigorous physical activity (MVPA) in the typically developing population [1]. These results suggest that the benefits conferred by PA could be negated by prolonged periods of SB. Movement lies on a continuum with categories termed "sedentary", "light PA" (LPA), "moderate PA" and "vigorous PA" to describe increasing intensities of movement [2]. SB is often considered to be a separate construct from a lack of MVPA because of its distinct deleterious health outcomes, which includes type 2 diabetes, cardiovascular events and all-cause morbidity [3].

By definition, cerebral palsy (CP) is a group of permanent disorders of the development of movement and posture, causing activity limitations, that are attributed to non-progressive disturbances to the developing foetal or infant brain [4]. Individuals with CP are more likely to spent increased time in SB compared to the typically developing population, due to the physical limitations associated with the condition. Both children and adults with CP may spend 76-99% of their waking time sitting [5, 6], compared to typically developing children and adults who spend approximately 55% of their waking time seated [7]. In individuals with CP, sitting time is associated with ability level according to the gross motor function classification system (GMFCS) [6, 8].

'Operationalization' refers the process of defining how to quantify a concept which itself is not directly measurable. The commonly used and accepted definition for SB was established through a global consensus project by the SB Research Network (SBRN) in 2012. In this definition, SB is operationationally defined in the typically developing population through an energy expenditure component, "any waking behavior characterized by an energy expenditure of ≤1.5 metabolic equivalents (METs)", and a postural component "in a sitting, reclining or lying posture" [9]. MET is a ratio of the energy consumption during postures or activities against a reference resting basal metabolic rate (i.e., 3.5 $mL \cdot kg^{-1} \cdot min^{-1}$ or 1 $kcal \cdot kg^{-1} \cdot h^{-1}$). Due to a lack of large muscle recruitment, the energy expenditure during SB is between 1.0-1.5 METs [10]. It is unclear whether this operationalization has been used in or is applicable to individuals with CP. Because they have balance impairments and atypical muscle tone, and consequently different muscle activity, compared to the typically developing population [11], it cannot be assumed that the ≤1.5 METs threshold also applies for these individuals. It is important to explore the construct of SB in individuals with CP.

An understanding of the definition of SB, and its impact on individuals with CP is a first step towards improving their health outcomes. Furthermore, for those with CP who have limited gross motor function (i.e. GMFCS level IV and V), interventions aimed to decrease SB through increased LPA may be more feasible to implement compared to those aimed solely to promote MVPA due to their severe motor restrictions, but the health impact of those intervention remain to be determined. Before further studies of these interventions and of the health outcomes of SB can be developed, a common understanding and validated measures for this physical behaviour in individuals with CP are needed. The purpose of this review is to, firstly, 'map' the range and extent of research on the operationalization and measurement tools of SB and, secondly, explore studies that report on the relationships between health indicators and SB in children and adults with CP. A scoping review is suited towards the purpose of discovering the boundaries of research of these concepts and developing a broad overview of the topic of SB in individuals with CP [12].

A recent scoping review has explored patterns, measurement and interventions to reduce SB in children with physical disabilities, including CP [13]. The review demonstrated that children with physical disabilities spend more time sedentary than their typically developing peers and that no interventions have been effective in reducing SB in the population. The review also reports the measurement methods for SB in these individuals, which were all accelerometer-type devices. However, the population in this scoping review included children up to age eighteen years and did not include adults with physical disability. The review also searched for studies that described measuring a behaviour termed as being "sedentary" rather than studies that also characterize activities that are constituted as SB (e.g., measuring the sitting posture through muscle activation). To address some of these remaining knowledge gaps, we identified the need to do a comprehensive search reporting on the operationalization of the term "sedentary" in individuals with CP and studies that measured energy expenditure and muscle activation while sitting, lying or reclining. This was because there is no consensus on the definition of SB in individuals with CP. Thus, researchers may have reported differently on this behaviour differently.

Our scoping review aimed to expand on the existing literature by exploring the operationalization of SB in individuals with CP across all ages and functional levels.

Furthermore, we aim to explore the relationship between SB and health indicators in people with CP.

This scoping review was guided by the following primary research questions:

(1) What is the documentation of SB in individuals with CP?

(a) What is the operationalization of SB or activities that constitute SB in this population?

(b) What tools (direct and indirect) are used to measure the operationalization of SB in individuals with CP, and are the reliability and the validity of these tools assessed in this population?

A secondary research question directed at included studies was:

(2) What is the relationship between SB and health indicators in people with CP?

Methods

We followed the Joanna Briggs Institute Reviewer's Manual for guidance on conducting the present systematic scoping review [14]. The manual draws upon and provides additional guidance on the scoping review framework outlined by Arksey and O'Malley [15] and Levac, Colquhoun and O'Brien [16]. The reporting of the scoping review followed the Preferred Reporting Items for Systematic Reviews and Meta-Analysis Extension for Scoping Reviews [17].

Rather than answering a set of more specific clinical questions as in a systematic review, such as on the effectiveness or feasibility of treatment or practice, the primary purpose of the questions in this review is to 'map' the range and extent of research on operationalizing and measuring SB in children and adults with CP [12]. A scoping review is better suited towards the purpose of discovering the boundaries of research on SB research. As there is potentially a wide range in the types of studies used to answer the research questions, a scoping review would be more appropriate than a systematic review because it generally incorporates evidence from a greater range of study designs and methodologies.

Eligibility Criteria

Inclusion/ Exclusion Criteria

To be included in the scoping review, the study had be a full original primary research article published in an English peer-reviewed journal that: (a) Study design - Has an experimental (randomized controlled trial and quasi-experimental trial), qualitative, longitudinal or observational (including cross-sectional, cohort, and case-control studies) design; (b) Population - includes a sample of at least 50% of individuals with CP of any age; (c) Phenomena of interest - (a) Reports an operationalization of SB or activities that constitute SB through, (i) Measuring a typically sedentary posture using the terms "sitting", "lying" or "reclining" through different methods (e.g., muscle activation), (ii) Using the term "sedentary", in the study's introduction/methods/results/discussion (e.g., describes anywhere in the study the construct measured was SB, even if it is used interchangeably with physical inactivity), to describe the behaviour or activity in which time spent (i.e., percent time, minutes or hours) is measured; (b)

Reports on the validity and/or reliability of a tool for measuring SB or typically sedentary postures; (d) Context - contain evidence from any context.

We excluded studies that: (a) only reported on measures generally obtained while in a sedentary posture (e.g., resting heart rate, resting energy expenditure) because the postures were often not described in detail and the purpose of these measures were not to characterize SB; (b) studies whereby the sample contained less than 50% of individuals with CP; (c) conference abstracts, protocols, books, grey literature or theses (not published/peer reviewed and/or do not provide the results of primary research); or (d) review articles (not a primary research design).

Search Strategy

We consulted a Health Sciences librarian, identified relevant databases, and developed and piloted the search strategy. A Health Sciences librarian also peer-reviewed the final search strategy. We searched the following databases: OVID Medline, CINAHL, Excerpta Medica database (EMBASE), OVD psycINFO and Physiotherapy Evidence Database (PEDro). Databases were reviewed from January 1, 2011 to December 2, 2019, inclusive. The start date of the time range was decided in order to capture recent articles that use the SBRN definition of SB, aligning with a few months before the SBRN Letter to the Editor calling for a standardized use of the terms "sedentary" and "sedentary behaviours" [9]. The rationale for the selection of the start date is that since the letter by the SBRN called to clarify the difference between "SB" and "physical inactivity". The measurement and reporting of SB were expected to be more uniform and adhere to a more rigorous standard. The search terms "sedentary behaviour" and "cerebral palsy" and their respective synonyms were searched. The search terms are provided in Appendix S2.

We imported all citations into Covidence (2015 Veritas Health Innovation, Melbourne, Australia). The title/abstract and then full text was screened for eligibility by two independent reviewers (JX, SR) using predetermined inclusion criteria (Appendix S1). Studies which did not clearly violate the inclusion/exclusion criteria at the stage of title/abstract screening moved onto the full-text screening stage. Discrepancies at each of these two screening stages were resolved by consensus discussion or through a third reviewer (JWG) if required before moving onto the next screening stage. Multiple reports of the same study were identified and linked before reporting.

The reference lists of included publications were hand searched for additional studies that were not identified by the initial search.

Data Collection

Using the Joanna Briggs Institute Reviewer's Manual [14], we extracted the following information from each article: study characteristics (author(s), design, sample size, objectives, country of origin), demographics (percent of patients with CP, age, sex, CP diagnoses), (a) operationalization of SB or SB activities through (i) measurement results of behaviours termed "sitting", "lying", "reclining" or "sedentary" through a physiological component (e.g., muscle activation or energy expenditure); (ii) the SB threshold, cut-point or postures of measurement tools for time spent in behaviours termed "sedentary"; (c) report of the psychometric properties of a tool for measuring behaviours termed "sitting", "lying", "reclining" or "sedentary"; (d) reports on a statistical assessment of a relationship between SB time and a health indicator. One reviewer (JX) piloted a data extraction table on ten randomly selected articles. The table and extracted data were then reviewed by two other authors (SR, JWG) and discussed to ensure completeness of the extracted data. Data extraction was independently conducted for 20% of included studies by both reviewers (JX, SR). The reviewers then convened to discuss and ensure agreement between the extracted data before one reviewer continued to extract data from the remaining 80% of included studies (JX). Any discrepancies between the two reviewers during the screening and data extraction process were resolved through a discussion in a meeting with a 3rd reviewer (JWG). As scoping reviews do not aim to produce a critically appraised and synthesized result to a particular question [14], a formal assessment of methodological quality of the included studies was not conducted.

Analysis involved categorizing the age of participants based on the age groups set out in the Canadian 24-Hour Movement Guidelines [18]. The results are classified under the aims of this review: (a) operationalization of SB through measurements of SB or a typically sedentary posture (e.g., through muscle activation or energy expenditure); (b) operationalization of SB through the SB threshold, cut-point or postures of measurement tools for time spent in SB by individuals with CP; (c) Report on the validity and/or reliability of tools for measuring SB, or one or more typically sedentary postures; (d) Reports on a statistical assessment of a relationship between SB time and a health indicator. For studies that reported statistically significant associations between SB and health indicators, thresholds were used to categorize the strength of these associations. The validity and reliability of measurement tools were categorized, based on thresholds, for the reported results of correlation coefficient or the area under the receiver operating characteristic curve. The thresholds, developed by Landis and Koch [19], were used to categorize any correlations or areas under the receiver operating characteristic curve under the following interpretations: 0- < 0.2 poor, 0.2- < 0.4 fair, 0.4- < 0.6 moderate, 0.6- < 0.8 substantial, and 0.8- < 1.0 almost perfect [19, 20]. For studies that did not report a correlation coefficient, we reported on the rating for validity and reliability as reported by the study. As the purpose of this review is not to examine the effectiveness of interventions, only the baseline data for intervention studies will be included. For the secondary aim, selected studies that report on the relationship between SB with at least one of the following eleven health indicators in individuals with CP were included. The detailed inclusion criteria for the health indicators is in Appendix S3. The health indicators were indicators are chosen based on literature, covering both prevalence and expert discussions of health indicators, important to individuals with CP. They were selected by one author based on a review of the literature (JX), and then checked by two experts for thoroughness (PM, JWG). Selection of health indicators was also based on a desire to cover both the physical and psychological aspects of health. The health indicators included are:

1) cardiometabolic disease risk factors: (a) cardiorespiratory fitness; (b) body size; (c) body composition; (d) sleep (e) nutrition; (f) blood pressure; (g) blood lipids; 2) depression and depression-related symptoms; 3) anxiety; 4) pain; 5) fatigue.

Common risk factors for multimorbidity risk, in particular, cardiometabolic disease, and the core outcomes (a-g) for assessing cardiometabolic disease risk factors, were identified by expert consensus through a Delphi procedure [21, 22]. Depression and anxiety are the two most common mental illnesses in the general population and have a two-to-three-fold greater likelihood of occurring in individuals with a long-term condition or disability [23]. Pain occurs in more 50% of individuals with CP from moderate to severe intensity and could be one of the most relevant factors influencing their physical activity and participation in daily activities [24, 25]. Similarly, fatigue has been identified by rehabilitation physicians, along with pain, as one of the most important CP-related impairments in adulthood that can interfere with activities of daily life [26]. Due to the elevated prevalence and reported agreement by experts on the importance of these eleven health outcomes, exploring their relationship with SB could elucidate the importance of SB specific to important and critical health concerns in individuals with CP.

Results

We identified 1644 potentially eligible citations and 951 unique citations after deduplication. Of these, 854 were removed after title and abstract screening. After completing full-text screening, 53 articles were excluded due to ineligible study design, research population, or not measuring SB as outlined in the research questions. An additional 2 studies were discovered upon a hand search of the citations in included studies. Thirty-nine unique studies from 46 publications met the inclusion criteria. Figure 1 is a flowchart of the study selection process. As studies rather than reports are the unit of interest, we attempted to link multiple reports of the same study or contact the corresponding authors. The demographic information for publications reporting on the same study was recorded for the largest sample size in which movement behaviour data from participants were extracted since some reports may have used a subset of data from a larger study.

Charting the data

Of the 39 included studies: 29 were cross-sectional [6, 8, 27-56, 59], 1 was quasi-experimental with cross-sectional results reported [57, 58]; 1 was longitudinal [60-62], 3 were randomized controlled trials [5, 63, 64], 2 were pre-post trial [65, 66], 1 was a quasi-randomized trial [67], 1 was a randomized wait-list controlled trial [68], and 1 study used a cross-sectional design to test tool validity and a longitudinal design to test reliability [69]. The study results were from a total of 1818 participants.

Figure 2 shows the number of publications, or the reports of studies, over the review's years of inclusion range. The number of publications increased from one per year between 2011-2013 [31, 32, 53] to 13 articles in 2014 [5, 6, 29, 30, 39, 42, 43, 47- 50]. Between 2015-2019, the number of studies ranged from three per year in 2018 to eight per year in 2019 [8, 27, 28, 33-38, 40, 41, 44-46, 51, 52, 54, 56-58, 60-69].

The included studies were conducted in the following countries: Australia (n=14) [27, 29, 30, 32, 35-37, 41, 44, 54, 56, 59, 60-63, 68] , Republic of Ireland (n=4) [43, 47-52], Netherlands (n=5) [5, 28, 55, 64, 67], Canada (n=4) [8, 33, 40, 42], USA (n=2) [34, 45], China (n=2) [31, 65], Middle East (n=2) [6, 57, 58], United Kingdom (n=2) [39, 53], Spain (n=1) [46], Germany (n=1) [38], Sweden (n=1) [66] and Switzerland (n=1) [69].

The participants in 33 of the 39 studies in this review included children under 18 years of age, while 20 studies included children over 18 years of age. A single study may contain a sample which overlaps across multiple age groups. The number of studies for each age group were as follows: early years (0–4 years of age) (n=7) [35-37, 39, 55, 59-62, 67] , children and adolescents (5–17 years of age) (n=32) [5, 6, 27, 29-32, 35-39, 41-46, 48-58, 60-69], adults (18-64 years of age) (n=20) [5, 6, 8, 27-30, 33, 35, 39, 40, 43, 45, 47, 49, 50, 56-58, 63, 64, 69] and older adults (65+ years of age) (n=5) [8, 28, 33, 49, 50, 61]. Three studies did not indicate an age range but provided a mean age [31, 38, 66]. The majority of adult studies were composed of young adults, with the upper age limit being in the twenties. The sample size for the CP group included in each study ranged from 11 to 222 (first quartile=20, third quartile=57, median=36, interquartile range= 37).

While all studies included individuals with CP classified as GMFCS I, II and/or III (n=39), fourteen studies also included participants classified as GMFCS IV and/or V [6, 28, 33, 35-37, 40, 44, 46, 55, 56, 59-62, 64, 66].

The 39 included studies covered three major topics to address the primary research question (Figure 3); studies that answered the secondary research question were selected from those that addressed the primary research question. Seven studies reported the operationalization of typically sedentary postures through measurements of muscle activation, energy expenditure or oxygen consumption [28, 32, 38, 45, 54-56] (Table 1). Twenty-two studies from 28 publications report on SB cut-points and postures used for direct measures of minutes, hours or percent time spent in SB [5, 6, 30, 31, 33, 36, 37, 40-42, 44, 46-48, 51, 52, 56-58, 60-68] (Table 2). Fifteen studies from sixteen publications evaluated the validity or reliability of measurement tools or algorithms that measure SB or typically sedentary postures [8, 27, 29, 32, 34, 35, 38, 39, 43, 45, 49, 50, 53, 54, 59, 69] (Table 1). The studies of validity or reliability validated against the energy expenditure in sitting or lying so there was overlap between four studies that report on the operationalization of sedentary postures through measurements of energy expenditure and studies that evaluated the validity or reliability of measurement tools or algorithms that measure SB or typically sedentary postures [32, 38, 45, 54]. There was also overlap between one study that both reported an operationalization of SB through measurements of energy expenditure in sedentary postures and through postures used by a measurement tool for time spent in SB [56]. Twelve publications from eight studies, out of the 39 studies identified for the primary aim, reported on the relationship between SB and health indicators in individuals with CP [30, 40, 46-49, 56-58, 60-62] (Table 3).

Operationalization of Activities that Constitute SB in Individuals with CP

Operationalization of SB through measurements of energy expenditure, muscle activation and oxygen consumption in typically sedentary postures

Seven studies, enrolling a total of 241 individuals with CP in GMFCS I-V, measured postures typically characterized as sedentary through muscle activation or energy expenditure [28, 32, 38, 45, 54-56] (Table 1). Of these studies, two studies measured muscle activation [28, 55] and two studies measured oxygen consumption [28, 45]. No studies measured the reclining posture. The results of the operationalization of these postures is found in Table S1.

Six studies reported measurements for the physiological aspects of the sitting posture [28, 38, 45, 54-56] and four studies reported measurements for the physiological aspects of the lying posture [32, 38, 45, 56]. The energy expenditure for sitting supported and unsupported in persons with CP do not differ significantly in MET values from healthy adult controls [28]. The MET values for the sitting and lying postures were ≤1.5METs across all GMFCS levels, I-IV [28] and I-V [55]. There was small variability between GMFCS levels. Energy expenditure for writing and lying across GMFCS I-III was ≤1.5METs [45]. The energy expenditure for handwriting for GMFCS I-II and for GMFCS III were both ≤1.5METs [54]. For one study, energy expenditure for SB ranged from 1.2 (1.1–1.3) METs for GMFCS I-III, although the postures were not specified [32]. One study that did not express energy expenditure as METs reported, during sitting for the non-hemiparetic and hemiparetic side, energy expenditures of 0.3 ± 0.5 kcal/min and 0.4 ± 0.6 kcal/min, respectively [38].

Five out of the six studies that measured the METs of SB postures found levels for energy expenditure that align with the SBRN threshold of ≤1.5 METs. The one study that found intensities for typically sedentary postures above this threshold reported on the peak energy expenditures and activities during the corresponding times for these peak energy expenditures, as reported by the parents of the participants [56]. The study found the peak energy expenditures to be 5.98 METs during lying down for one participant in GMFCS V, and 5.90 METs during sitting for one participant in GFMCS V. The measurement instrument used was the Actiheart VR accelerometer, which is an indirect measure of energy expenditure.

As MET is calculated by dividing resting VO_2 by VO_2 expended in a certain behaviour, three out of the five studies examining sedentary MET estimated resting metabolic rate using Schofield's equation [70], which provides a non-population specific estimate [32, 45, 54]. Two studies used the resting energy expenditures of each individual in the sample [28, 55].

The muscle activation for the sitting posture was measured by two studies [28, 55]. During sitting supported and unsupported, adults with CP did not differ significantly in electromyography (EMG) values from health adult controls [28]. Muscle activity during sitting did not differ significantly from lying down at rest [55].

The oxygen consumption for the sitting posture was measured by two studies [28, 45]. Oxygen consumption at supine rest across GMFCS I-III was not significantly lower than during sitting [45]. During lying, VO2 did not differ significantly between persons with CP in GMFCS I, GMFCS II, GMFCS III-IV, and healthy controls [28].

Characterization of SB through SB Thresholds, Cut-Points and Postures used by Measurement Tools

Twenty-two studies from 28 publications used cut-points and postures to measure minutes, hours, or percent time spent in SB [5, 6, 30, 31, 33, 36, 37, 40-42, 44, 46-48, 51, 52, 56-58, 60-68] (Table 3). Details of the measurement methods are found in Table S2-S4. The results of percent time, minutes or hours spent in SB by individuals with CP reported in these 22 studies are presented in Table S5. These studies enrolled a total of 1220 individuals with CP in GMFCS I-V. Of these studies, 14 used accelerometers, one used a heart rate monitor combined with an accelerometer, one study used a heart rate monitor, and six studies used activity monitors, in which SB was operationalized through cut-points or energy expenditure threshold.

Six studies used activity monitors to measure time spent in SB [5, 6, 30, 57, 58, 63, 64]. Two studies reported using the ActivPAL3 activity monitor [6, 57, 58], while two studies reported using the ActivPAL activity monitor [30, 63]. Two studies used the VitaMove ambulatory monitoring system [5, 64]. All studies measuring SB percent time, minutes or hours with activity monitors characterized SB activities as lying and sitting [5, 6, 30, 57, 58, 63, 64]. One study also considered wheelchair mobility as sedentary in addition to sitting and lying postures [6].

Fourteen studies used a triaxial accelerometer to measure SB time [31, 33, 36, 37, 40, 41, 42, 44, 47, 48, 51, 52, 60, 61, 62, 65-68]. Three studies measured SB using the ActiGraph GT3X [33, 40, 43]. Two studies used the SB cut-points validated in the typically developing population of <100 counts per minute in the vertical axis [40, 66], citing Freedson [71, 72]. One study cited Healy [1] and Matthews [7] for the SB cut-points of <100 counts per minute [33]. One study used sedentary cut-points validated in individuals with CP [44], citing Oftedal [54], which validates the cut-points in the same age group of 1-3 years for GMFCS level I to III and for GMFCS level IV to V.

Three studies measured SB using the ActiGraph GT3X+ [41, 67, 68]. Two studies cited the SB cut-point of <100 counts per minute in the vertical axis by Evenson [73] [67, 41]. One study used sedentary cut-points validated in individuals with CP [68], citing Trost [54], which validates the cut-points of < 8 counts per 15 s in the same GMFCS level I-III group.

Two studies measured SB using the ActiGraph GT3X and GT3X+ [36, 37, 60-62]. One study used SB cut-points validated in preschoolers of the typically developing population [36; 37], citing the Butte [73] vector magnitude threshold of 820 counts per minute. The other study used GMFCS specific sedentary cut-points validated in individuals with CP [60-62], derived from validation studies by Keawutan [35] and Oftedal [59] in the same age range between 2-5 years.

One study used the ActiTrainer triaxial accelerometer [66]. The study used the SB cut-points validated in the typically developing population of <100 counts per minute in the vertical axis, citing Freedson [72].

Two studies used the triaxial RT3 accelerometer [47, 48, 51, 52]. Both these studies used the SB cut-points validated in typically developing children and adolescents [47, 48, 51, 52]. One study cited the Vanhelst [75] vector magnitude threshold of <41 counts per minute for the RT3 accelerometer [48, 51, 52]. Another study cited Rowland [76] for a threshold of <100 counts per minute [47].

Three studies measured SB with uniaxial accelerometers, including one study using the ActiGraph GT1M [42] and two other studies using the Actigraph 7164 [31, 65]. The studies all cited the SB cut-point of <100 counts per minute in the vertical axis by Evenson [73].

One study used the ActiheartVR accelerometer [56], which converts accelerometer counts and heart rate into energy expenditure using a branched equation [77]. The study used the SB threshold for energy expenditure of ≤1.5 METs, citing Ward [78].

The Bodyguard measured SB time indirectly through energy expenditure derived from heart rate [46]. The study defined SB as < 20% of the maximal MET but did not cite the source of this cut-point.

Indirect methods of measuring SB consists of self-reported measures of the behaviour. The International Physical Activity Questionnaire was the only self-reported measurement tool of SB used [66]. The questionnaire asks participants to record the amount of time spent sitting under various settings, as well as time spent in a motor vehicle and has been validated in the typically developing population [79].

The reporting of time per day or percentage time spent in SB by these tools was done by 21 out of the 22 studies that measured sedentary time [5, 6, 30, 31, 33, 36, 37, 41, 42, 44, 47, 48, 51, 52, 56-58, 60-68], since one of these studies measured but did not report the sedentary time [46] (Table S5). Of the 21 studies, 8 studies reported SB solely through percentage time spent in SB [6, 31, 36, 37, 44, 60-62, 64, 65, 67], 8 studies solely through minutes or hours per day in SB [5, 30, 33, 40, 59, 63, 66, 68], and 5 studies through both percentage time in SB and through minutes or hours per day in SB [41, 47, 48, 51, 52, 56-58].

Waking hours was measured in different ways among these studies. While most studies reported time spent in SB as a percentage of waking time, one study reported it as a percentage of 24 hours [6] and another study assumed a day to be 12 hours [56].

In addition to reporting sedentary time, some studies also reported the pattern of SB, including frequency of breaks and length of bouts. Three studies reported the number of sedentary breaks [33, 44, 59]. A break was defined by only one of these two studies as any movement of 3 seconds or more that are of light, moderate, or vigorous intensity [59]. Five studies reported on the frequency of sedentary bouts; one study reported on bouts of 60 minutes and 120 minutes [57, 58]; one study on bouts ranging from 10 seconds to 30 minutes [5]; one study on bouts ranging from less than 1 minute to greater than 10 minutes [44]; one study on bouts ranging from more than 5 seconds to more than 30 minutes [64]; and one study on bouts exceeding 30 minutes [56].

Validity and Reliability of Tools Measuring Sedentary Postures or Sedentary Time

Fifteen studies from sixteen publications, enrolling a total of 513 individuals with CP in GMFCS I-V, evaluated the validity of measurement tools or algorithms that measure SB or typically sedentary postures [8, 27, 29, 32, 34, 35, 38, 39, 43, 45, 49, 50, 53, 54, 59, 69] (Table 1). Of these studies, two also reported test-retest reliability [29, 69]. One study reported inter-instrument reliability [45]. The results for the SB cut-points or SB activities evaluated for validity and reliability are found in Table S6. Two of the fifteen studies were conducted on individuals in GMFCS IV-V, who have greater mobility impairments [35, 59]. Both these studies were conducted in individuals 5 years of age or under. Two of the 15 studies included individuals with CP who were older than 20 years of age [8, 49, 50].

Of the two studies that reported test-retest reliability, one study found excellent agreement between 2 school days measured 4 weeks apart for the sitting and lying postures [69]. Another study measured the agreement of the ActivPAL after 6 days, for the sitting posture, and found substantial relative reliability although it was not specified if the two measurements were taken on a weekday or weekend [29]. A study that evaluated inter-instrument reliability found excellent agreement between the right and left hip placement for the ActiGraph, Sensewear and StepWatch for a range of activities of varying intensities, including sedentary behaviours [45].

One study examined construct validity by assessing the ability of the instrument to discriminate between school days and weekends for sitting and lying [69]. One study examined concurrent validity among three accelerometer-based instruments [45]. Thirteen studies examined criterion validity by agreement between device output and 'ground-truth' direct video observation or indirect calorimetry in standardized lab settings [8, 27, 29, 32, 34, 35, 38, 39, 43, 49, 50, 53, 54, 59]. One study validated against direct in-person observations [34]. For the studies that validated against indirect calorimetry, two specified using the SBRN energy expenditure threshold for SB [32, 45], whereas one study used a threshold of ≤2 METs for SB [49, 50], citing Garber [80].

For sitting and lying, one study showed fair to good agreement with indirect calorimetry for measurements taken on the same day by the ActiGraph GT3X, in children and young adults with CP [45]. Two studies reported that the ActiGraph GT3X and ActiGraph GT3X+ accelerometers [35] and ActiGraph GT1M accelerometer [59] significantly overestimated SB in toddlers and children with CP who were stationary with or without limb movement. Another study showed that, for sitting and lying, the ActiGraph GT3X showed excellent classification accuracy based on validated cut-points, in children with CP [54]. One study reported that the ActiGraph 7164 had excellent classification accuracy for a resting protocol of SB, in children with CP, without details of the sedentary postures measured [32]. The only non-instrument based measurement tool for SB, a machine learning algorithm, showed excellent classification accuracy for ActiGraph GTX+ results, for all device placement locations, for the sitting and lying postures, in children and young adults with CP [27]. No significant difference was found between indirect calorimetry and the RT3 accelerometer for the lying posture in individuals with CP over 6 years of age [49, 50].

The ActivPAL was reported to overestimate SB in children and young adults with CP by misclassifying sitting as standing [39, 43]. However, another study found that the instrument underestimated SB in children and young adults with CP for sitting [29]. Two studies showed excellent classification accuracy of the instrument's ability to detect sitting and lying in children, youth and adults with CP [8, 53].

One study showed good to fair agreement with indirect calorimetry for measurements of a range of activity intensities, including SB, in children and young adults with CP, by the Sensewear armband monitor [45]; the Sensewear armband monitor is an instrument with a triaxial accelerometer and heat-related channels to estimate energy expenditure. Another study found a significant difference between the Sensewear armband monitor and indirect calorimetry in individuals with CP over 6 years of age [49, 50]. There was no significant difference between indirect calorimetry and the Sensewear armband monitor placed on the hemiparetic and non-hemiparetic side when sitting in children with CP [38].

There was no significant difference between indirect calorimetry and the Intelligent Device Energy Expenditure and Activity, which collects raw data from five accelerometer sensors and converts it to energy expenditure, in individuals with CP over 6 years of age [49, 50].

One study indicated fair to good agreement with indirect calorimetry by the StepWatch, an instrument which uses a uniaxial accelerometer to record energy expenditure and step count, in children and young adults with CP [45]. One study showed fair to good construct validity for a set-up of five inertial measurement unit devices, which uses a series of accelerometers equipped on the body to detect walking episodes, in children and young adults with CP [69]. The results reported agreement of SB, consisting of sitting and lying, between two school days of the same week and low correlation between a school day and a weekend (intraclass correlation (ICC) < 0.50), indicative of the discriminative capacity of the instrument. The Pediatric SmartShoe, which uses a series of accelerometers in the shoes to detect various postures, showed excellent classification accuracy for the ability to detect sitting in different postures in daily life situations along a range of activity intensities, including SB, for children with CP [34].

In general, there was excellent classification accuracy and agreement between the SB measurement tools and the standards against which they were validated. There were moderate to excellent levels for all types of reliability reported.

Report on Health Indicators

Twelve publications from eight studies, out of the 39 studies identified for the primary aim, reported on the relationship between SB and health indicators in individuals with CP, with the majority reporting on associations of these indicators with SB [30, 40, 46-49, 56-58, 60-62] (Table 3). The findings were conducted in a total of 449 individuals with CP in GMFCS I-V. The statistical relationships between SB and these health indicators is reported in Table S7.

For cardiometabolic disease risk factors, statistically significant positive associations were noted between SB and blood pressure for individuals with CP across all ages in GMFCS I-III [48, 51, 52]. No significant correlations were noted between SB in individuals with CP and the following health indicators: waist circumference [40]; height [30]; height-for-age z score [60, 61, 62]; c-reactive protein levels [47]; metabolic syndrome [47] as defined according to the most recent joint interim statement [81];cardiorespiratory fitness [48, 51, 52]; and body fat percentage [56]. In another study, significant positive correlations of moderate strength were found between body fat percentage for young children with CP across all GMFCS levels, although the significant associations disappeared when adjusted for GMFCS level [60-62]. The study also found statistically significant but weak negative association between SB and fat-free mass controlled for height, but the significant associations disappeared when adjusted for GMFCS level [60-62]. Statistically significant positive correlations of fair strength were noted between SB and BMI for children in GMFCS level II-III [57, 58]. However, no associations between BMI and SB were found in another study for adults with CP across all GMFCS levels [22].

Children with CP and chronic pain across all GMFCS levels spent significantly less time sedentary than their typically developing peers (TDP) with chronic pain, whereas no differences were found in sedentary time between CP and TDP without chronic pain [46].

No significant correlations were noted between SB in adults with CP across all GMFCS levels and fatigue [40].

Discussion

In this scoping review, we identified 39 studies that addressed the primary research questions of operationalizing and measuring SB, out of which eight studies were used to address our secondary research question by reporting on health indicators. Although the included studies represented the population of individuals with CP across all ages and functional levels, the majority of studies were conducted in children. Of the studies that were conducted in adults, many were conducted for individuals in early adulthood (i.e., in the early twenties) rather than in older adults. The studies pertained to the measurement of typically sedentary postures, reports of sedentary thresholds, and evaluation of the validity/reliability of instruments and algorithms that measured SB.

The primary research question pertains to the operationalization of SB. The research question was addressed by identifying studies that measured percent time, minutes or hours in SB. The postures classified as sedentary through activity monitors were sitting and lying, which matched the postures classified as sedentary in the typically developing population. The primary question was also addressed by exploring the operationalization of SB through measurements of 3 variables; energy expenditure, oxygen consumption and muscle activation. As SB in this population is not defined through these variables, the studies explored reports measurements of the physiological aspects of typically sedentary postures. These results could inform the first steps towards operationalized definitions of SB using these three variables. The energy expenditure of typically sedentary postures in individuals with CP generally matched the threshold outlined in the typically developing population (≤1.5 METs) [9]. Only one study, which reported peak energy expenditures and the corresponding activities to the time of the peak energy expenditures, measured intensities higher than this threshold for sitting and lying [56]. However, the results in this study were found in only two individuals and may be inaccurate because the instrument used measured heart rate and accelerometry, which are used to infer energy expenditure and are not direct measures of energy expenditure. Furthermore, since the corresponding activities to the time of peak energy expenditure were reported by the parents of participants, the results are subject to social desirability bias or recall bias [82]. The overall results support the operationalization of the SBRN definition in individuals CP because the threshold for energy expenditure for typically sedentary postures matches the current SBRN definition. The findings of the energy expenditure in typically sedentary postures also suggest that researchers interested in reducing SB in people with CP need only to measure sitting or lying time and show a reduction in these postures in order to demonstrate change in SB through the postural and intensity components, to align with the SBRN definition of SB [9].

There is a large variability in the types of tools used to measure SB time, which consists primarily of accelerometers. These tools vary in the measurement epochs, thresholds for SB, and accelerometer axes, making it difficult to draw comparisons between studies of SB time and patterns in individuals with CP. In order to compare SB time between studies, there needs to be a greater consistency in reporting (e.g. report both minutes/day sedentary time and percent wear-time). Furthermore, there should be consistency in the operationalization of SB used through citing the SBRN definition and studies should explain explicitly what construct they are measuring (posture or energy or both) as an indicator of SB.

The majority of studies demonstrated good to excellent criterion validity for the commonly used SB cut-point of ≤100 count per minute [71-73, 77], against gold standards including direct observation or indirect calorimetry. These results indicate that even though the cut-point was originally validated in typically developing population, it has been demonstrated to be valid in individuals with CP at GMFCS level I-III. The gaps in the evaluation of validity and reliability of SB measurement tools are the lack of studies in GMFCS IV and V where the risk of misclassification of sedentary time is much higher. The reason is the inability of accelerometers to detect small changes in their sedentary time, because their movements (e.g., rolling, creeping, standing) have lower acceleration and would possibly be assigned to 'sedentary'. Only two validity studies were conducted in non-ambulatory individuals classified in GMFCS level IV-V, both of which studied young children and indicated a tendency of accelerometer cut-points to overestimate SB [35, 59]. However, the other study that reported an overestimation of SB was conducted in children classified in GMFCS level I [43], which indicates that more studies are needed to determine the effect of GMFCS level on the accurate estimation of SB. Of two validation studies conducted in middle aged to older adults, all participants were ambulatory [8, 49, 50]. The findings indicate a gap in literature testing the validity of thresholds to measure SB in the non-ambulatory population at GMFCS level IV and V and in older participants. Most of the studies that validated against indirect calorimetry followed the SBRN threshold of ≤1.5 METs. Only one study validated against the threshold of ≤2 METs for SB [50]. The 2.0 METs SB threshold was created by the American College of Sports Medicine for adults [80]. The threshold was determined through examining energy expenditure of activities considered as very light PA for cardiorespiratory endurance and resistance exercises rather than energy expenditure in sedentary postures. As such, the threshold should not be used for classifying SB.

A gap in the SB literature in individuals with CP is that all protocols for validation were conducted in lab environments. These study conditions produce almost perfect agreement with a gold standard. However, when the cut-point is applied in free-living situations, there is a large risk of misclassification because the situations in the validity study may not necessarily represent typical daily movements. The tendency for misclassification in daily settings has been demonstrated in the typically developing population. In the typically developing population, the ActiHeart was shown to significantly underestimate sedentary time compared to a reference instrument, the ActivPAL, under daily living conditions [84]. Similarly, the ability of the ActiGraph GT3X+ to detect standing compared with sitting/lying in older adults was no better than chance [85]. The misclassification may be especially prominent with higher cut-points since standing still results in low accelerometer counts [86]. These considerations from the typically developing population may be especially relevant for individuals in GMFCS level IV-V with more severe motor restrictions, where light PA, such as standing, may be more feasible to perform than MVPA due to their motor restrictions. The findings in the typically developing populations further highlights the need for the calibration and validation of cut-points that maximizes ecological validity to accurately measure SB time in individuals with CP especially in non-ambulatory individuals.

Another gap is the lack of reliability studies for the measurement of SB across all GMFCS levels, as only three studies in this review, conducted in GMFCS levels I-III, reported results for reliability [29, 45, 69]. One study considered for inclusion in the review stated that reliability could not be calculated for SB because activity counts were primarily 0 counts per epoch [87]. Validity connects to reliability because a tool needs to be able to consistently measure accurate levels of SB [88]. While the reliability of the instrument is determined in labs using mechanical apparatuses, the reliability of measured SB levels, or the consistency in measured time in the behaviour, requires the device to be worn by participants [89-91]. Several studies in typically developing children has demonstrated the number of wear days required, which is typically 7 consecutive days of measurement with ⩾4 days of valid accelerometer data for children and 3-5 days among adults [92, 93], indicating an impact of age on reliability. Based on this review and the studies cited by this review, reliability for individuals with CP has only been conducted on accelerometer-based devices. Some of the other measurement tools used in the operationalization of SB from this review, including the Cortex Metamax, for measuring energy expenditure [94-95], and surface EMG, for measuring muscle activity [97]. are shown to be valid and reliable only in the typically developing population. Most reliability studies that were cited were conducted in the typically developing population, for PA rather than for SB, and were used to justify the measurement period used by studies in this review. As reliability and validity are inextricably linked, further studies of reliability are needed for commonly used measurement tools in the operationalization of SB specific to this population.

The secondary research question was to identify studies of SB that reported on health indicators. The studies reviewed showed a statistically significant association with only two indicators reported in two studies: blood pressure [48] and BMI [57, 58]. One explanation for the lack of relationships found between the other health indicators studied and SB in this population is that sample sizes were generally <100 participants, as is typical for this population. If SB is very high across the studied group, which is apparent from the cross-sectional SB outcomes, then there is unlikely to be enough of a gradient in SB in a small sample to see significant associations. Some relationships between SB and health indicators in individuals with CP matched results seen in typically developing individuals. For example, in the typically developing children, significant positive associations were also seen with BMI [2, 98, 99], and blood pressure [100]. On the other hand, significant negative associations were found between cardiorespiratory fitness and SB in typically developing children and adolescents [101] whereas no associations were found in individuals with CP [52].

The limited information on the relationships of health indicators with SB in individuals with CP is evident as only six of the eleven health indicators that were sought in the review were studied (i.e., body size, body composition, blood pressure, fatigue, pain, cardiorespiratory fitness). As such, the effects of SB are not well known for children and adults with CP. However, health outcomes of SB seen in the typically developing population should not be extrapolated to individuals with CP. Part of the negative health outcomes of SB is due to a general lack of activation of the large muscle groups that contribute to weight bearing when performing them [102, 103]. Individuals with CP have increased muscle activation in postures characterized as sedentary [104]. As such, it cannot be assumed that the negative health outcomes of sedentary postures in the typically developing population also applies to individuals with CP and further studies should be conducted to gain a greater understanding of the effects of SB in individuals with CP.

Limitations

There are several possible limitations to this review with regards to the breadth of studies captured. While SB and physical inactivity have previously been differentiated [9], we encountered studies during our search that used SB and physical inactivity synonymously. The limitation prevents inclusion of studies that intend to report on SB but characterize the behaviour instead as “physical inactivity”.

This review was limited to studies that may have intended to measure SB but did not provide an operationalization of SB. Two included studies defined SB in an indirect way [12, 13]. These studies stated in the introduction or discussion sections that time spent “sitting/lying” are representative of “sedentary behaviours” but then stated only that “sitting and lying” were measured in the methods section, without further specifying that these postures constituted as SB. The studies highlight the limitation that there could have been other studies that intended to capture SB but were not included because they did not explicitly define the behaviour.

Another limitation of this study is that we restricted the start date of our search to January 2011, to align with the SBRN letter to the editor calling for a standardized definition of SB [9]. Earlier studies that may have aligned with the definition would have been excluded.

The selection of included health outcomes was based solely on literature of prevalence [23] and one study of experts deciding on multimorbidity risk factors [21, 22]. However, there may be other outcomes, considered critical or important to individuals with CP, with a relationship to SB which were not reported in this review (i.e., association between SB and gross motor function [30], motor capacity and capability [36, 37], growth velocity z-score [60-62], and five fundamental movement skills [31]).

Conclusion

The objective of this study was to operationalize SB and report on the tools that measure the operationalization of SB in individuals with CP across all ages and across all functional levels. We found 39 studies, which reported on SB postures, thresholds, and measurement tools, mainly in children and adolescents with CP. The definition for SB outlined by the SBRN of ≤1.5 METs generally applies to the typically sedentary postures, consisting of sitting and lying among individuals with CP. The tools that measure SB are highly variable in the way they are used. While the reported validity of SB measurement tools in individuals with CP is generally good to excellent, there is a tendency to overestimate SB. There were very few relationships between health indicators and SB that were reported in the population.

For future steps, a similar process to the Terminology Consensus Project carried out by the SBRN for the typically developing population should be conducted [83]. In this initiative, a literature review was completed to identify the current uses of the SBRN definition for SB. Key terms from the search were collated, and a draft list of terms and their definitions was created for consensus, through a survey, by a panel of international experts. A similar process should be conducted for expert consensus on the SBRN definition of SB in individuals with CP, and information that should be reported in future SB studies. Such a consensus project will promote for researchers to use a standardized definition and reporting of SB to, in turn, allow for comparisons of SB between studies.

Furthermore, the results of the scoping review could be used in future research examining changes in SB in individuals with CP. The results of this review demonstrated that the energy cost of sitting or lying could be inferred to be ≤1.5 METs, aligning with both the postural and intensity components in the SBRN's definition of SB. This implication could especially be important in population-based settings, in which it may be easier to measure posture through an accelerometer with an inclinometer, and inferring energy expenditure, rather than directly measuring energy expenditure. This option may also be easier than indirectly measuring energy expenditure through accelerometers, since energy expenditure obtained from accelerometers may be inaccurate, as shown in the typically developing population [105]. However, more research should first be conducted to determine the accuracy of SB measurement tools for older adults and in non-ambulatory individuals with CP.

Funding:

Dr. Gorter holds the Scotiabank Chair in Child Health Research at McMaster University. The funders have no role in the development of the review.

References

1. Healy GN, Matthews CE, Dunstan DW, Winkler EAH, Owen N. Sedentary time and cardio-metabolic biomarkers in US adults: NHANES 2003–06. European Heart Journal. 2011;32(5):590–7.
2. Tremblay MS, Colley RC, Saunders TJ, Healy GN, Owen N. Physiological and health implications of a sedentary lifestyle. Appl Physiol Nutr Metab. 2010;35(6):725–40.
3. Wilmot EG, Edwardson CL, Achana FA, Davies MJ, Gorely T, Gray LJ, et al. Sedentary time in adults and the association with diabetes, cardiovascular disease and death: systematic review and meta-analysis. Diabetologia. 2012;55(11):2895–905.
4. Rosenbaum P, Paneth N, Leviton A, Goldstein M, Bax M, Damiano D, et al. A report: the definition and classification of cerebral palsy April 2006. Dev Med Child Neurol Suppl. 2007;109:8–14.
5. Nooijen CF, Slaman J, Stam HJ, Roebroeck ME, Berg-Emons R, Learn2Move Research Group. Inactive and sedentary lifestyles amongst ambulatory adolescents and young adults with cerebral palsy. J NeuroEngineering Rehabil. 2014;11(1):49.
6. Shkedy Rabani A, Harries N, Namoora I, Al-Jarrah MD, Karniel A, Bar-Haim S. Duration and patterns of habitual physical activity in adolescents and young adults with cerebral palsy. Dev Med Child Neurol. 2014;56(7):673–80.
7. Matthews CE, Chen KY, Freedson PS, Buchowski MS, Beech BM, Pate RR, et al. Amount of Time Spent in Sedentary Behaviors in the United States, 2003-2004. American Journal of Epidemiology. 2008;167(7):875–81.
8. Claridge EA, van den Berg-Emons RJG, Horemans HLD, van der Slot WMA, van der Stam N, Tang A, et al. Detection of body postures and movements in ambulatory adults with cerebral palsy: a novel and valid measure of physical behaviour. J NeuroEngineering Rehabil. 2019;16(1):125.
9. Sedentary Behaviour Research Network. Letter to the Editor: Standardized use of the terms “sedentary” and “sedentary behaviours.” Appl Physiol Nutr Metab. 2012;37(3):540–2.
10. Pate RR, O’Neill JR, Lobelo F. The Evolving Definition of “Sedentary”: Exercise and Sport Sciences Reviews. 2008;36(4):173–8.
11. Bax MCO, Flodmark O, Tydeman C. Definition and classification of cerebral palsy. From syndrome toward disease. Dev Med Child Neurol Suppl. 2007;109:39–41.
12. Munn Z, Peters MDJ, Stern C, Tufanaru C, McArthur A, Aromataris E. Systematic review or scoping review? Guidance for authors when choosing between a systematic or scoping review approach. BMC Med Res Methodol. 2018;18(1):143.
13. Ganz F, Hammam N, Pritchard L. Sedentary behavior and children with physical disabilities: a scoping review. Disability and Rehabilitation. 2020;1–13.
14. Peters MDJ, Godfrey CM, Khalil H, McInerney P, Parker D, Soares CB. Guidance for conducting systematic scoping reviews: International Journal of Evidence-Based Healthcare. 2015;13(3):141–6.
15. Arksey H, O’Malley L. Scoping studies: towards a methodological framework. International Journal of Social Research Methodology. 2005;8(1):19–32.
16. Levac D, Colquhoun H, O’Brien KK. Scoping studies: advancing the methodology. Implementation Sci. 2010;5(1):69.

17. Tricco AC, Lillie E, Zarin W, O'Brien KK, Colquhoun H, Levac D, et al. PRISMA Extension for Scoping Reviews (PRISMA-ScR): Checklist and Explanation. Ann Intern Med. 2018;169(7):467.
18. Canadian Society for Exercise Physiology. Canadian 24-Hour Movement Guidelines [Internet]. CSEP. [cited 2020 June 20]. Available from: https://csepguidelines.ca/
19. Landis JR, Koch GG. The measurement of observer agreement for categorical data. Biometrics. 1977;33(1):159–74.
20. Zweig MH, Campbell G. Receiver-operating characteristic (ROC) plots: a fundamental evaluation tool in clinical medicine. Clin Chem. 1993;39(4):561–77.
21. Benner JL, McPhee PG, Gorter JW, Hurvitz EA, Peterson MD, Obeid J, et al. Focus on Risk Factors for Cardiometabolic Disease in Cerebral Palsy: Toward a Core Set of Outcome Measurement Instruments. Archives of Physical Medicine and Rehabilitation. 2019;100(12):2389–98.
22. McPhee PG, Benner JL, Balemans ACJ, Verschuren O, van den Berg-Emons RJG, Hurvitz EA, et al. Multimorbidity risk assessment in adolescents and adults with cerebral palsy: a protocol for establishing a core outcome set for clinical research and practice. Trials. 2019;20(1):176.
23. Smith KJ, Peterson MD, O'Connell NE, Victor C, Liverani S, Anokye N, et al. Risk of Depression and Anxiety in Adults With Cerebral Palsy. JAMA Neurol. 2019;76(3):294.
24. Alriksson-Schmidt A, Hägglund G. Pain in children and adolescents with cerebral palsy: a population-based registry study. Acta Paediatr. 2016;105(6):665–70.
25. Riquelme I, Cifre I, Montoya P. Age-Related Changes of Pain Experience in Cerebral Palsy and Healthy Individuals. Pain Med. 2011;12(4):535–45.
26. Hilberink S, Roebroeck M, Nieuwstraten W, Jalink L, Verheijden J, Stam H. Health issues in young adults with cerebral palsy: Towards a life-span perspective. Acta Derm Venereol. 2007;39(8):605–11.
27. Ahmadi M, O'Neil M, Fragala-Pinkham M, Lennon N, Trost S. Machine learning algorithms for activity recognition in ambulant children and adolescents with cerebral palsy. J NeuroEngineering Rehabil. 2018;15(1):105.
28. Balemans ACJ, Houdijk H, Koelewijn GR, Piek M, Tubbing F, Visser-Meily A, et al. A Comparison of the Physiology of Sedentary Behavior and Light Physical Activity in Adults With and Without a Physical Disability. Journal of Physical Activity and Health. 2019;16(10):894–901.
29. Bania T. Measuring Physical Activity in Young People with Cerebral Palsy: Validity and Reliability of the ActivPAL™ Monitor: Measuring PA in People with CP. Physiother Res Int. 2014;19(3):186–92.
30. Bania TA, Taylor NF, Baker RJ, Graham HK, Karimi L, Dodd KJ. Gross motor function is an important predictor of daily physical activity in young people with bilateral spastic cerebral palsy. Dev Med Child Neurol. 2014;56(12):1163–71.
31. Capio CM, Sit CHP, Abernethy B, Masters RSW. Fundamental movement skills and physical activity among children with and without cerebral palsy. Research in Developmental Disabilities. 2012;33(4):1235–41.
32. Clanchy KM, Tweedy SM, Boyd RN, Trost SG. Validity of accelerometry in ambulatory children and adolescents with cerebral palsy. Eur J Appl Physiol. 2011;111(12):2951–9.

33. Claridge EA, Mcphee PG, Timmons BW, Martin Ginis KA, Macdonald MJ, Gorter JW. Quantification of Physical Activity and Sedentary Time in Adults with Cerebral Palsy: Medicine & Science in Sports & Exercise. 2015;47(8):1719–26.
34. Hegde N, Zhang T, Uswatte G, Taub E, Barman J, McKay S, et al. The Pediatric SmartShoe: Wearable Sensor System for Ambulatory Monitoring of Physical Activity and Gait. IEEE Trans Neural Syst Rehabil Eng. 2018;26(2):477–86.
35. Keawutan P, Bell KL, Oftedal S, Davies PSW, Boyd RN. Validation of Accelerometer Cut-Points in Children With Cerebral Palsy Aged 4 to 5 Years: Pediatric Physical Therapy. 2016;28(4):427–34.
36. Keawutan P, Bell KL, Oftedal S, Davies PSW, Ware RS, Boyd RN. Habitual Physical Activity in Children With Cerebral Palsy Aged 4 to 5 Years Across All Functional Abilities: Pediatric Physical Therapy. 2017;29(1):8–14.
37. Keawutan P, Bell KL, Oftedal S, Davies PSW, Ware RS, Boyd RN. Relationship between habitual physical activity, motor capacity, and capability in children with cerebral palsy aged 4–5 years across all functional abilities. Disability and Health Journal. 2018;11(4):632–6.
38. Koehler K, Abel T, Wallmann-Sperlich B, Dreuscher A, Anneken V. Energy Expenditure in Adolescents With Cerebral Palsy: Comparison of the SenseWear Armband and Indirect Calorimetry. Journal of Physical Activity and Health. 2015;12(4):540–5.
39. McAloon MT, Hutchins S, Twiste M, Jones R, Forchtner S. Validation of the activPAL activity monitor in children with hemiplegic gait patterns resultant from cerebral palsy. Prosthet Orthot Int. 2014;38(5):393–9.
40. McPhee PG, Brunton LK, Timmons BW, Bentley T, Gorter JW. Fatigue and its relationship with physical activity, age, and body composition in adults with cerebral palsy. Dev Med Child Neurol. 2017;59(4):367–73.
41. Mitchell LE, Ziviani J, Boyd RN. Habitual Physical Activity of Independently Ambulant Children and Adolescents With Cerebral Palsy: Are They Doing Enough? Physical Therapy. 2015;95(2):202–11.
42. Obeid J, Balemans ACJ, Noorduyn SG, Gorter JW, Timmons BW. Objectively Measured Sedentary Time in Youth With Cerebral Palsy Compared With Age-, Sex-, and Season-Matched Youth Who Are Developing Typically: An Explorative Study. Physical Therapy. 2014;94(8):1163–7.
43. O' Donoghue D, Kennedy N. Validity of an activity monitor in young people with cerebral palsy gross motor function classification system level I. Physiol Meas. 2014 Nov 1;35(11):2307–18.
44. Oftedal S, Bell KL, Davies PSW, Ware RS, Boyd RN. Sedentary and Active Time in Toddlers with and without Cerebral Palsy: Medicine & Science in Sports & Exercise. 2015;47(10):2076–83.
45. O'Neil ME, Fragala-Pinkham M, Lennon N, George A, Forman J, Trost SG. Reliability and Validity of Objective Measures of Physical Activity in Youth With Cerebral Palsy Who Are Ambulatory. Physical Therapy. 2016;96(1):37–45.
46. Riquelme I, do Rosário RS, Vehmaskoski K, Natunen P, Montoya P. Influence of chronic pain in physical activity of children with cerebral palsy. NRE. 2018;43(2):113–23.
47. Ryan JM, Crowley VE, Hensey O, Broderick JM, McGahey A, Gormley J. Habitual physical activity and cardiometabolic risk factors in adults with cerebral palsy. Research in Developmental Disabilities. 2014;35(9):1995–2002.

48. Ryan JM, Hensey O, McLoughlin B, Lyons A, Gormley J. Reduced Moderate-to-Vigorous Physical Activity and Increased Sedentary Behavior Are Associated With Elevated Blood Pressure Values in Children With Cerebral Palsy. Physical Therapy. 2014 Aug 1;94(8):1144–53.
49. Ryan JM, Walsh M, Gormley J. A comparison of three accelerometry-based devices for estimating energy expenditure in adults and children with cerebral palsy. J NeuroEngineering Rehabil. 2014;11(1):116.
50. Ryan J, Walsh M, Gormley J. Ability of RT3 Accelerometer Cut Points to Detect Physical Activity Intensity in Ambulatory Children With Cerebral Palsy. Adapted Physical Activity Quarterly. 2014;31(4):310–24.
51. Ryan JM, Forde C, Hussey JM, Gormley J. Comparison of Patterns of Physical Activity and Sedentary Behavior Between Children With Cerebral Palsy and Children With Typical Development. Physical Therapy. 2015;95(12):1609–16.
52. Ryan JM, Hensey O, McLoughlin B, Lyons A, Gormley J. Associations of Sedentary Behaviour, Physical Activity, Blood Pressure and Anthropometric Measures with Cardiorespiratory Fitness in Children with Cerebral Palsy. Zhu S, editor. PLoS ONE. 2015;10(4):e0123267.
53. Tang KT, Richardson AM, Maxwell D, Spence WD, Stansfield BW. Evaluation of an Activity Monitor for the Objective Measurement of Free-Living Physical Activity in Children With Cerebral Palsy. Archives of Physical Medicine and Rehabilitation. 2013;94(12):2549–58.
54. Trost SG, Fragala-Pinkham M, Lennon N, O'Neil ME. Decision Trees for Detection of Activity Intensity in Youth with Cerebral Palsy: Medicine & Science in Sports & Exercise. 2016;48(5):958–66.
55. Verschuren O, Peterson MD, Leferink S, Darrah J. Muscle Activation and Energy-Requirements for Varying Postures in Children and Adolescents with Cerebral Palsy. The Journal of Pediatrics. 2014;165(5):1011–6.
56. Williams SA, McFadden LM, Blackmore AM, Davey P, Gibson N. Do adolescents with cerebral palsy meet recommendations for healthy weight and physical activity behaviours? Disability and Rehabilitation. 2019;1–6.
57. Aviram R, Harries N, Shkedy Rabani A, Amro A, Nammourah I, Al-Jarrah M, et al. Comparison of Habitual Physical Activity and Sedentary Behavior in Adolescents and Young Adults With and Without Cerebral Palsy. Pediatric Exercise Science. 2019;31(1):60–6.
58. Bar-Haim S, Aviram R, Shkedy Rabani A, Amro A, Nammourah I, Al-Jarrah M, et al. Effects of Exercise Interventions on Habitual Physical Activity and Sedentary Behavior in Adolescents With Cerebral Palsy. Pediatric Exercise Science. 2019;31(4):416–24.
59. Oftedal S, Bell KL, Davies PSW, Ware RS, Boyd RN. Validation of Accelerometer Cut Points in Toddlers with and without Cerebral Palsy: Medicine & Science in Sports & Exercise. 2014;46(9):1808–15.
60. Oftedal S, Davies PSW, Boyd RN, Stevenson RD, Ware RS, Keawutan P, et al. Longitudinal Growth, Diet, and Physical Activity in Young Children With Cerebral Palsy. PEDIATRICS. 2016;138(4):e20161321–e20161321.
61. Oftedal S, Davies PS, Boyd RN, Stevenson RD, Ware RS, Keawutan P, et al. Body composition, diet, and physical activity: a longitudinal cohort study in preschoolers with cerebral palsy. Am J Clin Nutr. 2017;105(2):369–78.
62. Keawutan P, Bell KL, Oftedal S, Ware RS, Stevenson RD, Davies PSW, et al. Longitudinal physical activity and sedentary behaviour in preschool-aged children with cerebral palsy across all functional levels. Dev Med Child Neurol. 2017;59(8):852–7.

63. Bania TA, Dodd KJ, Baker RJ, Graham HK, Taylor NF. The effects of progressive resistance training on daily physical activity in young people with cerebral palsy: a randomised controlled trial. Disability and Rehabilitation. 2016;38(7):620–6.
64. Slaman J, Roebroeck M, Dallmijer A, Twisk J, Stam H, van den Berg-Emons R, et al. Can a lifestyle intervention programme improve physical behaviour among adolescents and young adults with spastic cerebral palsy? A randomized controlled trial. Dev Med Child Neurol. 2015;57(2):159–66.
65. Capio CM, Sit CHP, Eguia KF, Abernethy B, Masters RSW. Fundamental movement skills training to promote physical activity in children with and without disability: A pilot study. Journal of Sport and Health Science. 2015;4(3):235–43.
66. Lauruschkus K, Hallström I, Westbom L, Tornberg Å, Nordmark E. Participation in physical activities for children with cerebral palsy: feasibility and effectiveness of physical activity on prescription. Arch Physiother. 2017;7(1):13.
67. Halma E, Bussmann JBJ, Berg-Emons HJG, Sneekes EM, Pangalila R, Schasfoort FC, et al. Relationship between changes in motor capacity and objectively measured motor performance in ambulatory children with spastic cerebral palsy. Child Care Health Dev. 2020;46(1):66–73.
68. Reedman SE, Boyd RN, Trost SG, Elliott C, Sakzewski L. Efficacy of Participation-Focused Therapy on Performance of Physical Activity Participation Goals and Habitual Physical Activity in Children With Cerebral Palsy: A Randomized Controlled Trial. Archives of Physical Medicine and Rehabilitation. 2019;100(4):676–86.
69. Gerber CN, Carcreff L, Paraschiv-Ionescu A, Armand S, Newman CJ. Reliability of single-day walking performance and physical activity measures using inertial sensors in children with cerebral palsy. Annals of Physical and Rehabilitation Medicine. 2019 Apr;S1877065719300338.
70. Schofield WN. Predicting basal metabolic rate, new standards and review of previous work. Hum Nutr Clin Nutr. 1985;39 Suppl 1:5–41.
71. Freedson PS, Melanson E, Sirard J. Calibration of the Computer Science and Applications, Inc. accelerometer: Medicine & Science in Sports & Exercise. 1998;30(5):777–81.
72. Freedson P, Pober D, Janz KF. Calibration of Accelerometer Output for Children: Medicine & Science in Sports & Exercise. 2005;37(Supplement):S523–30.
73. Evenson KR, Catellier DJ, Gill K, Ondrak KS, McMurray RG. Calibration of two objective measures of physical activity for children. Journal of Sports Sciences. 2008;26(14):1557–65.
74. Butte NF, Wong WW, Lee JS, Adolph AL, Puyau MR, Zakeri IF. Prediction of Energy Expenditure and Physical Activity in Preschoolers: Medicine & Science in Sports & Exercise. 2014;46(6):1216–26.
75. Vanhelst J, Béghin L, Rasoamanana P, Theunynck D, Meskini T, Iliescu C, et al. Calibration of the RT3 accelerometer for various patterns of physical activity in children and adolescents. Journal of Sports Sciences. 2010;28(4):381–7.
76. Rowlands AV, Thomas PWM, Eston RG, Topping R. Validation of the RT3 Triaxial Accelerometer for the Assessment of Physical Activity: Medicine & Science in Sports & Exercise. 2004;36(3):518–24.
77. Mcminn D, Acharya R, Rowe D, Gray S, Allan J. Measuring Activity Energy Expenditure: Accuracy of the GT3X+ and Actiheart Monitors. International Journal of Exercise Science. 2013;6:217–29.
78. Ward DS, Saunders RP, Pate RR. Physical activity interventions in children and adolescents. Champaign, IL: Human Kinetics; 2007. 270 p. (Physical activity intervention series).

79. Rosenberg DE, Bull FC, Marshall AL, Sallis JF, Bauman AE. Assessment of Sedentary Behavior With the International Physical Activity Questionnaire. Journal of Physical Activity and Health. 2008;5(s1):S30–44.
80. Garber CE, Blissmer B, Deschenes MR, Franklin BA, Lamonte MJ, Lee I-M, et al. Quantity and Quality of Exercise for Developing and Maintaining Cardiorespiratory, Musculoskeletal, and Neuromotor Fitness in Apparently Healthy Adults: Guidance for Prescribing Exercise. Medicine & Science in Sports & Exercise. 2011;43(7):1334–59.
81. Alberti KGMM, Eckel RH, Grundy SM, Zimmet PZ, Cleeman JI, Donato KA, et al. Harmonizing the Metabolic Syndrome: A Joint Interim Statement of the International Diabetes Federation Task Force on Epidemiology and Prevention; National Heart, Lung, and Blood Institute; American Heart Association; World Heart Federation; International Atherosclerosis Society; and International Association for the Study of Obesity. Circulation. 2009;120(16):1640–5.
82. Brenner PS, DeLamater JD. Social Desirability Bias in Self-reports of Physical Activity: Is an Exercise Identity the Culprit? Soc Indic Res. 2014;117(2):489–504.
83. Tremblay MS, Aubert S, Barnes JD, Saunders TJ, Carson V, et al. Sedentary Behavior Research Network (SBRN) – Terminology Consensus Project process and outcome. Int J Behav Nutr Phys Act. 2017;14(1):75.
84. Júdice PB, Santos DA, Hamilton MT, Sardinha LB, Silva AM. Validity of GT3X and Actiheart to estimate sedentary time and breaks using ActivPAL as the reference in free-living conditions. Gait & Posture. 2015;41(4):917–22.
85. Aguilar-Farías N, Brown WJ, Peeters GMEE (Geeske). ActiGraph GT3X+ cut-points for identifying sedentary behaviour in older adults in free-living environments. Journal of Science and Medicine in Sport. 2014;17(3):293–9.
86. Ridgers ND, Salmon J, Ridley K, O'Connell E, Arundell L, Timperio A. Agreement between activPAL and ActiGraph for assessing children's sedentary time. Int J Behav Nutr Phys Act. 2012;9(1):15.
87. Mitchell LE, Ziviani J, Boyd RN. Variability in Measuring Physical Activity in Children with Cerebral Palsy: Medicine & Science in Sports & Exercise. 2015;47(1):194–200.
88. Allen MJ, Yen WM. Introduction to measurement theory. Waveland Press; 2001.
89. Welk GJ, Schaben JA, Morrow Jr JR. Reliability of accelerometry-based activity monitors: a generalizability study. Medicine and science in sports and exercise. 2004;36(9):1637–45.
90. Mcclain JJ, Sisson SB, Tudor-Locke C. Actigraph accelerometer interinstrument reliability during free-living in adults. Medicine and science in sports and exercise. 2007;39(9):1509.
91. Esliger DW, Tremblay MS. Technical reliability assessment of three accelerometer models in a mechanical setup. Medicine and science in sports and exercise. 2006;38(12):2173–81.
92. for the ISCOLE Research Group, Barreira TV, Schuna JM, Tudor-Locke C, Chaput J-P, Church TS, et al. Reliability of accelerometer-determined physical activity and sedentary behavior in school-aged children: a 12-country study. Int J Obes Supp. 2015;5(S2):S29–35.
93. Trost SG, Mciver KL, Pate RR. Conducting Accelerometer-Based Activity Assessments in Field-Based Research: Medicine & Science in Sports & Exercise. 2005 Nov;37(Supplement):S531–43.
94. Brehm M-A, Harlaar J, Groepenhof H. Validation of the portable VmaxST system for oxygen-uptake measurement. Gait & Posture. 2004;20(1):67–73.
95. Medbø JI, Mamen A, Welde B, Heimburg E von, Stokke R. Examination of the Metamax I and II oxygen analysers during exercise studies in the laboratory. Scandinavian Journal of Clinical and Laboratory Investigation. 2002;62(8):585–98.

96. Meyer T, Georg T, Becker C, Kindermann W. Reliability of Gas Exchange Measurements from Two Different Spiroergometry Systems. Int J Sports Med. 2001;22(8):593–7.
97. Smoliga JM, Myers JB, Redfern MS, Lephart SM. Reliability and precision of EMG in leg, torso, and arm muscles during running. Journal of Electromyography and Kinesiology. 2010;20(1):e1–9.
98. Owen N, Healy GN, Matthews CE, Dunstan DW. Too Much Sitting: The Population Health Science of Sedentary Behavior. Exercise and Sport Sciences Reviews. 2010;38(3):105–13.
99. Proper KI, Singh AS, van Mechelen W, Chinapaw MJM. Sedentary Behaviors and Health Outcomes Among Adults. American Journal of Preventive Medicine. 2011;40(2):174–82.
100. Chaput J-P, Saunders TJ, Mathieu M-È, Henderson M, Tremblay MS, O'Loughlin J, et al. Combined associations between moderate to vigorous physical activity and sedentary behaviour with cardiometabolic risk factors in children. Appl Physiol Nutr Metab. 2013;38(5):477–83.
101. Santos R, Mota J, Okely AD, Pratt M, Moreira C, Coelho-e-Silva MJ, et al. The independent associations of sedentary behaviour and physical activity on cardiorespiratory fitness. Br J Sports Med. 2014;48(20):1508–12.
102. Hamilton MT, Hamilton DG, Zderic TW. Role of Low Energy Expenditure and Sitting in Obesity, Metabolic Syndrome, Type 2 Diabetes, and Cardiovascular Disease. Diabetes. 2007;56(11):2655–67.
103. Tikkanen O, Haakana P, Pesola AJ, Häkkinen K, Rantalainen T, Havu M, et al. Muscle Activity and Inactivity Periods during Normal Daily Life. Johannsen D, editor. PLoS ONE. 2013;8(1):e52228.
104. Innes J, Darrah J. Sedentary behavior: implications for children with cerebral palsy. Pediatric physical therapy : the official publication of the Section on Pediatrics of the American Physical Therapy Association. 2013;25(4):402–8.
105. Ndahimana D, Kim E-K. Measurement Methods for Physical Activity and Energy Expenditure: a Review. Clin Nutr Res. 2017;6(2):68.
106. Treuth MS, Schmitz K, Catellier DJ, McMurray RG, Murray DM, Almeida MJ, et al. Defining accelerometer thresholds for activity intensities in adolescent girls. Med Sci Sports Exerc. 2004;36(7):1259–66.
107. Puyau MR, Adolph AL, Vohra FA, Butte NF. Validation and Calibration of Physical Activity Monitors in Children. Obesity Research. 2002;10(3):150–7.
108. Brunton LK, Bartlett DJ. Construction and validation of the fatigue impact and severity self-assessment for youth and young adults with cerebral palsy. Developmental Neurorehabilitation. 2017;20(5):274–9.

Appendix S1. Search Terms in MEDLINE

-> Cerebral Palsy
1. Cerebral Palsy/
2. cerebral pals*.tw.
3. (Hemiplegi* or diplegi* or quadriplegi* or unilateral*) adj5 spastic*.tw.
4. (Hemiplegi* or diplegi* or quadriplegi* or unilateral*) adj3 ataxi*.tw.
5. little* disease.tw.

->Sedentary Behaviour
1. Sedentary Behavior/
2. sedentar*.tw.
3. Physical inactiv*.tw.
4. Stationary.tw.
5. lack adj2 activit*.tw.
6. low adj3 energy expend*.tw.
7. ((chair or stroller or seated or screen or television or tv or reading or media or computer or PC or car or automobile* or motor vehicle* or auto or bus or driving or indoor or in-door) adj3 (time* or duration*)).tw.
8. Screen?time.tw.
9. Screen time/
10. (screen or television or tv or media or computer or PC) adj3 ("use" or usage).tw.
11. (watch* or view*) adj2 (dvd* or video* or television or tv).tw.
12. (screen media or social media or video gam* or videogam* or computer gam* or electronic gam* or internet) adj5 (time or duration* or usage or "use").tw.
13. (Smartphone* or ipad or cellphone or mobile phone or cellular phone) adj2 (time or duration* or usage or "use"). tw.
14. ((screen) adj5 (behavio?r* or usage or "use")).tw.
15. Resting.tw.
16. Reclining.tw.
17. Lying.tw
18. Supine.tw.
19. Sitting.tw.
20. bed rest*.tw.
21. Bed rest/
22. Supine position/
23. Sitting position/
24. motor* transport.tw.
25. PC adj3 (usage or "use").tw.
26. (Web or internet) adj2 browsing.tw.

Limited to English language and articles from January 1, 2011

Appendix S2. Screening Form for Title/Abstract and for Full-text Articles

Title/Abstract Screening:

1. **Is the study in English?**

If no → EXCLUDE
If yes or unclear → go to next question

2. **Is the study published after and including Jan 2011?**

If no → EXCLUDE
If yes or unclear → go to next question

3. **Does the study have a reported sample of at least 50% of individuals with cerebral palsy?**

If no → EXCLUDE
If yes or unclear → go to next question

4. **Does the study measure SB or a posture typically characterized as sedentary (i.e., sitting, reclining, lying)?**

If no → EXCLUDE
If yes or unclear → go to next question

5. **Is this study experimental (randomized controlled and quasi-experimental), qualitative, longitudinal or observational (including cross-sectional, cohort, and case-control studies)?**

If no → EXCLUDE
If yes or unclear → go to full text screening

Full Text Screening:

1. Is the study in English?

If no ☐ EXCLUDE
If yes or unclear ☐ go to next question

2. Is the study published after and including Jan 2011?

If no ☐ EXCLUDE
If yes or unclear ☐ go to next question

3. Does the study have a reported sample of at least 50% of individuals with cerebral palsy?

If no ☐ EXCLUDE
If yes or unclear ☐ go to next question

4. Does study report on the following areas of SB:

INCLUDED:

- Measure a level of sedentary behaviour (i.e., percent time/minutes/hours spent in SB) and provide a definition for “sedentary behaviour” anywhere in the article

OR

- Analyze the validity, reliability and/or calibration of a tool for SB measurement (i.e., level of sedentary behaviour, sitting, reclining or lying)

OR

- Measure a posture typically characterized as sedentary [i.e., sitting/reclining/lying] (e.g., through muscle activation, energy expenditure)

EXCLUDED:

- Measure a variable generally obtained while in a sitting/lying/reclining posture (i.e., resting energy expenditure, resting heart rate)

If does not contain the criteria for inclusion □ EXCLUDE
If yes or unclear □ go to next question

5. Is this study experimental (randomized controlled and quasi-experimental), qualitative, longitudinal or observational (including cross-sectional, cohort, and case-control studies)?
If no □ EXCLUDE
If yes □ include in review

Appendix S3. Operationalization of Health Indicators Included

Identification of key terms for risk factors of cardiometabolic disease was found using search terms in a study reporting core outcomes for cardiometabolic risk factors [21, 22]:

1. Cardiometabolic disease risk factor

2. cardiorespiratory endurance (cardiorespiratory fitness, aerobic fitness, aerobic capacity, oxygen consumption, physical endurance, cardiovascular fitness, physical fitness, exercise capacity, VO2max, VO2peak)

3. body size (body height, body weight, body mass index (BMI))

4. body composition (body density measures, such as skinfold thickness measurements, bioelectrical impedance analysis or dual-energy x-ray absorptiometry body scans, body circumference)

4. Sleep (sleep, sleep disorders, drowsiness, nightmare, sleep debt, sleep pattern, sleep quality, sleep stage, sleep time, sleep waking cycle, sleep walking, sleep spindle, sopor, disturbances of sleep, sleep perturbation, sleep wake disorder, chronobiology disorder, fragmented sleep, hypnic headache, hypnagogic hallucination narcolepsy, paroniria, periodic limb movement disorder, sleep disordered breaking, sleep arousal disorder, dreams, dyssomnia, hyposomnia, hypersomnia, insomnia, parasomnia, sleeplessness, sleepiness, somnolence, tiredness, or sleep disorder, sleep disturbance, sleep paralysis)

5. Nutrition (diet, food intake behaviour/pattern, nutrition, feeding behavior, dietary intake, feeding/eating/drinking/alimentary/nutrition behavior/habit/pattern, diet, food intake/consumption/uptake, energy intake/consumption, calorie uptake/intake/consumption, nutrient uptake/intake/consumption, dietary uptake/intake/consumption, fat content, appetite, portion size, alcohol consumption, carbohydrate/fat/fluid/protein/vitamin/mineral intake, meal skipping)

6. blood pressure (Mean arterial blood pressure, systolic blood pressure, diastolic blood pressure, pulse pressure, blood pressure, hypertension encephalopathy, normotension, prehypertension, hypertension, hypotension, vascular resistance, intravascular pressure, arterial pressure, venous pressure, arterial pressure, intra-arterial pressure, arterial tension, blood tension, vascular pressure, arterial blood pressure, artery blood pressure, artery pressure, intraarterial blood pressure, intraarterial pressure, systemic arterial pressure, systemic artery pressure, aortic pressure, lung artery pressure, coronary artery pressure, mean arterial pressure, artery perfusion pressure, blood pressure fluctuation/regulation/variability, capillary pressure, atrial/atrium pressure/tension, ventricle/ventricular pressure/tension, orthostatic blood pressure, pulmonary artery occlusion pressure, pulse pressure, venous/vein/vena blood pressure, abnormal blood pressure depressed/ elevated blood pressure)

7. blood lipids (dyslipidemia, hyperlipidemia, hypolipemia, cholesterol, triacylglycerol, triglyceride, lipid profile)

Identification of key terms for depression, anxiety and related diseases was found from the Read code as outlined by Smith [24].

8. depression and depression-related symptoms
9. anxiety

Pain and fatigue were defined as any studies that used these terms.

10. Pain
11. Fatigue (i.e., physical tiredness, muscle soreness, exhaustion of your muscles and body [108])

Figure 1. PRISMA Flow Diagram Detailing the Study Selection Process

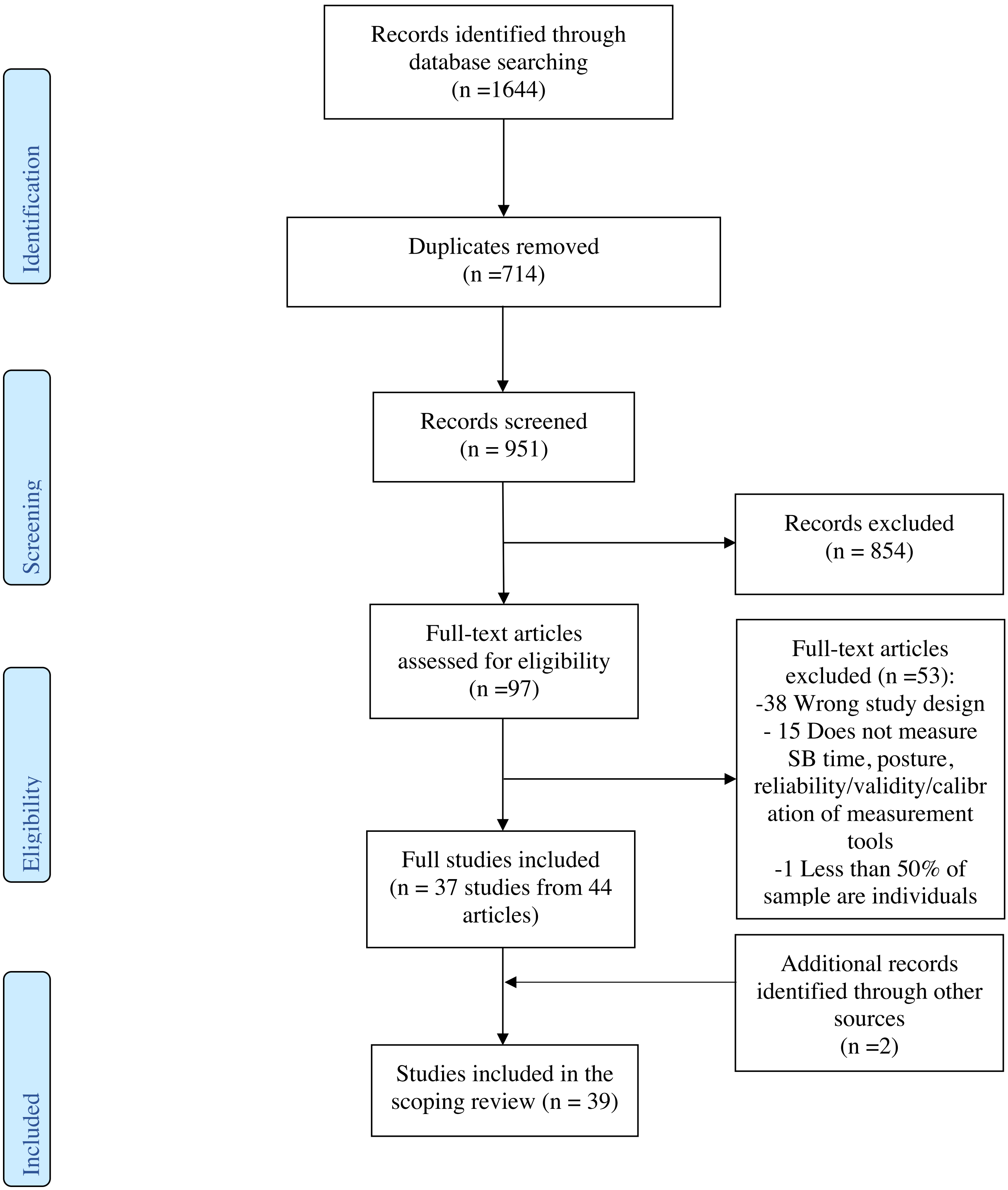


Figure 2. Distribution of the Number of Publications Across the Year Range Included

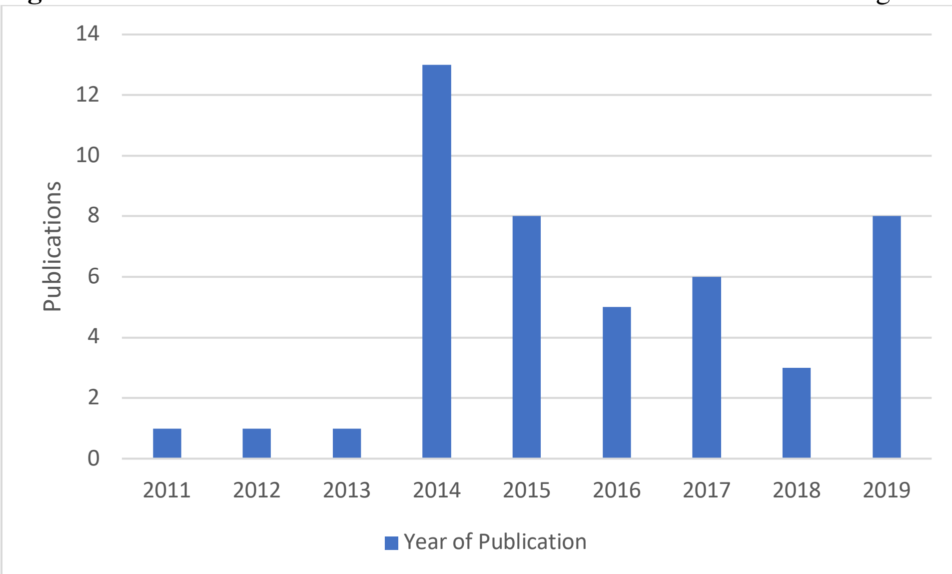
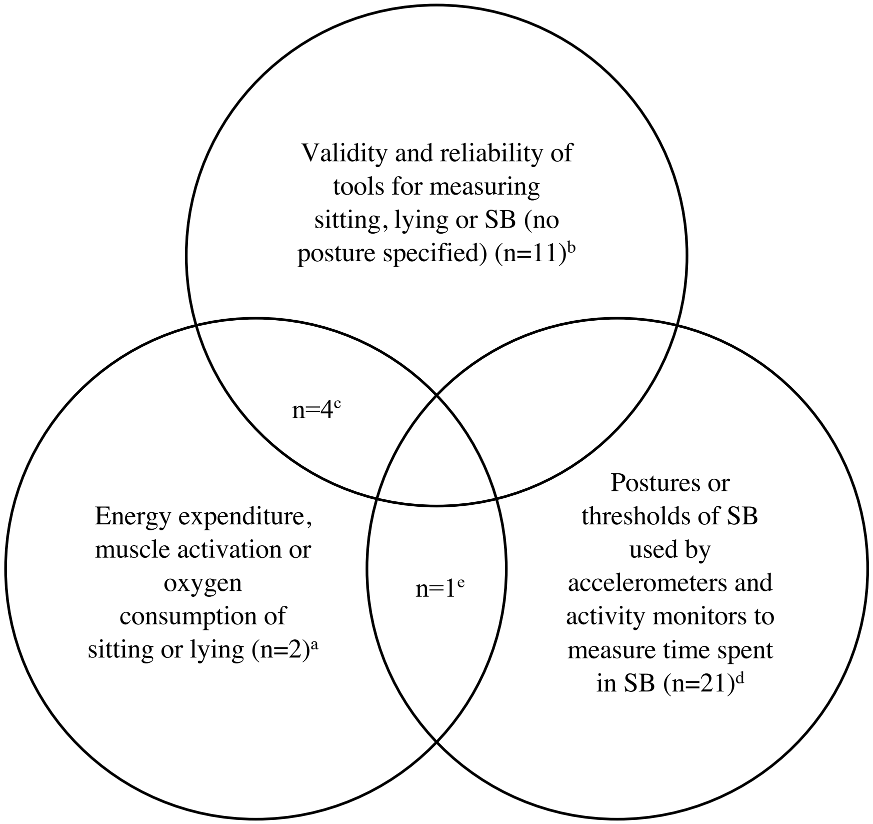


Figure 3. Thirty-nine included studies categorized based on the topics reported in the "Results" for the primary research question



[a] Balemans [28]; Verschuren [55]
[b] Bania [29]; Claridge [8]; Gerber [69]; Hegde [34]; Keawutan [35]; McAloon [39]; O' Donoghue [43]; Oftedal [59]; Ryan [49]; Ryan [50]; Tang [53]
[c] Clanchy [32]; Koehler [38]; O'Neil [45]; Trost [54]
[d] Ahmadi [27]; Aviram [57], Bar-Haim [58]; Bania [30]; Bania [63]; Capio [31]; Capio [65]; Claridge [33]; Halma [67]; Keawutan [37], Keawutan [36]; Keawutan [62], Oftedal [60], Oftedal [61]; Lauruschkus [66]; McPhee [40]; Mitchell [41]; Nooijen [5]; Obeid [42]; Oftedal [44]; Riquelme [46]; Reedman [68]; Ryan [51], Ryan [52], Ryan [48]; Ryan [47]; Shkedy [6]; Slaman [64]
[e] Williams [56]

Table 1. Studies Measuring Physiological Components of Sedentary Postures & the Validity and Reliability of Tools for Measuring SB in Individuals with CP

Author	Operationalization of sedentary postures through measurement of physiological components					Type of Tool Tested for Validity or Reliability		Validity or Reliability of Tools to Measure SB									
	Sitting	Lying	Energy expenditure	Oxygen consumption	Muscle activation	Machine Learning Algorithm	Accelerometer (i.e., activity monitors and tools containing accelerometers)	Validity						Reliability			
								Type of validity			Sitting	Lying	Other	Type of Reliability		Sitting	Lying
								Concurrent	Criterion	Construct				Test-retest	Inter-instrument		
Ahmadi [27]						X			X		X	X					
Balemans [28]	X	X	X	X	X		X										
Bania [29]							X		X		X			X		X	X
Clanchy [32]							X		X								
Claridge [8]							X		X		X						
Gerber [69]							X			X	X	X		X		X	X
Hegde [34]							X		X								
Keawutan [35]							X		X				Stationary with or without limb movement				
Koehler [38]			X				X		X		X						
McAloon [39]							X		X		X						
O' Donoghue [43]							X		X		X						
Oftedal [59]							X		X		X	X	Standing completely still				
O'Neil [45]	X	X	X	X			X	X			X	X		X			
Ryan [49]; Ryan [50]							X		X			X	< 2.0 MET				
Tang [53]							X		X		X	X					
Trost [54]							X		X		X	X					
Verschuren [55]	X		X		X		X										
Williams [56]	X	X	X				X										

Table 2. SB Operationalization by Measurement Tools for Sedentary Time (i.e., percent time/minutes/hours) in Individuals with CP

Author	Operationalization of SB used in Measurement		Direct Tools (accelerometer that are not activity monitors)							Operationalization of SB used in Measurement		Direct Tools (activity monitor)			Indirect Tools	Relationship between Health Indicator & Sedentary Time
	Threshold (i.e., counts or energy expenditure)		Triaxial Accelerometer				Uniaxial Accelerometer		Heart Rate Monitor & Triaxial Accelerometer	Postures		activPAL	activPAL 3	VitaMove	International Physical Activity Questionnaire	
	Validated in TDP	Validated in CP	ActiGraph GT3X	ActiGraph GT3X+	RT3	Acti-Trainer	Actigraph GT3M	ActiGraph 7164	ActiheartVR	Sitting	Lying					
Aviram [57], Bar-Haim [58]										X	X		X			X
Bania [30]										X	X	X				X
Bania [63]										X	X	X				
Capio [31]		X						X								
Capio [65]		X						X								
Claridge [33]	X		X													
Halma [67]	X			X												
Keawutan [37], Keawutan [36]	X		X	X												
Keawutan [62], Oftedal [60], Oftedal [61]		X	X	X												X
Lauruschkus [66]	X					X				X	X				X	
McPhee [40]	X		X													X
Mitchell [41]	X			X												
Nooijen [5]										X	X			X		
Obeid [42]	X							X								
Oftedal [44]		X	X													
Reedman [68]		X		X												
Riquelme [46]									X							X
Ryan [51], Ryan [52], Ryan [48]	X				X											X
Ryan [47]	X				X											X
Shkedy [6]										X	X		**X**			
Slaman [64]										X	X			**X**		
Williams[56]	X								X							X

Supplementary Tables for Scoping Review on the Operationalization, Measurement and Health Indicators of Sedentary Behaviour in Individuals with Cerebral Palsy

Table S1. Measurements of energy expenditure, oxygen consumption or muscle activation in typically sedentary postures in individuals with cerebral palsy

Author (year)/ Country	Demographics	GMFCS Level	CP Subtype	Measured Variable and Calculation	Instrument	SB definition: SB postures measured with citations used	SB definition: SB thresholds for classifying postures as sedentary	Key findings
Balemans [28], 2019 Netherlands	n=22, 45% females; age=18-86 Y; n=18 TDP	I-IV	Predominantly spastic	Oxygen Consumption	MetaMax 3B; Cortex	Lying in supine position (used to determine resting oxygen uptake to calculate MET) and sitting (with and without back or arm support), Tremblay [81]	N/A	During lying, VO2 did not differ significantly between persons with CP in GMFCS I (4.4 ± 1.4 mL/kg/min), GMFCS II (4.4 ± 2.0 mL/kg/min), GMFCS III-IV (3.1 ± 1.0 mL/kg/min), and healthy controls (p = 0.22).
				Energy expenditure: MET was calculated by dividing the V˙ O2 per condition by the average V˙ O2 during lying of each individual participant	MetaMax 3B; Cortex			During sitting supported and unsupported, persons with CP do not differ significantly in MET values from controls. The MET range for sitting support for GMFCS I was 1 1.02 ± 0.15 MET, for GMFCS II was 0.92 ± 0.18MET, and for GMFCS III-IV 1.04 (0.93–1.30) MET. The MET range for sitting support for GMFCS I was 1.00 ± 0.12 MET, for GMFCS II was 1.02 ± 0.17MET, and for GMFCS III-IV was 1.15 (1.03–1.19) MET.
				Muscle activation measured in percentage of EMG walking	Surface electromyography (EMG)			The percentage of EMG walking for lying in a supine position for GMFCS I was 10.7 ± 4.2%, GMFCS II was 12.2 ± 9.6%, and GMFCS III-IV was 9.7% (8.3–11.4%). The percentage of EMG walking for the supported sitting position in GMFCS I was 11.0 ± 3.1%, in GMFCS II was 9.0 ± 4.4%, and in GMFCS III-IV was 9.7% (8.5–11.6%). The percentage of EMG walking for the unsupported sitting position in GMFCS I was 12.9 ± 4.6%, in GMFCS II was 15.9 ± 18.5%, and in GMFCS III-IV was 17.8% 11.9–18.9%). During sitting supported and unsupported, persons with CP do not differ significantly in EMG values from healthy adult controls.
Clanchy [32], 2011 Australia	n=29, 43% females; age=8-16 Y	I-III	Spastic	Energy expenditure: MET was calculated by dividing mean VO2 by resting	Cosmed K4b2, Cosmed S.r.l, Rome, Italy	10-minute rest period described as being "sedentary" (no posture specified)	≤1.5 METs	Energy expenditure ranged from 1.2 (1.1–1.3) METs for GMFCS I-III during rest.

				metabolic rate (RMR). RMR was predicted from the participant's sex, age, body mass and height using Schofield's equation [70] for children aged 10–18 years				
Koehler [38], 2015 Germany	n=10, 30% females; age=13.4 ± 1.6 years Y	II	Hemiplegia	Energy expenditure	ZAN 600 Spirometric System (nSpire, Oberthulba, Germany)	Sitting	N/A	The energy expenditure measured during sitting for the non-hemiparetic and hemiparetic side were 0.3 ± 0.5 kcal/min and 0.4 ± 0.6 kcal/min, respectively.
O'Neil [45], 2016 USA	n=57, 51% females; age=6-20 Y	I-III	Spastic hemiplegia	Energy expenditure: MET was calculated by dividing mean V˙O2 by resting V˙O2, which was estimated using the Schofield's equation [70] for children aged 3-10 years or 10-18 years.	COSMED K4b2	1. Supine rest: lying down resting but not sleeping. 2. Writing: using a pencil or pen and copying text from a printed sheet while sitting at a desk[a]	≤1.5 METs	Energy expenditure for supine rest across GMFCS I-III was 1.1 (0.94–1.3) MET. Energy expenditure for writing across GMFCS I-III was 1.2-1.3 (1.1-1.4) METs.
				Oxygen Consumption	COSMED K4b2			Oxygen consumption at supine rest across GMFCS I-III was 5.1 (4.0–6.1) mL/kg/min. Oxygen consumption for writing across GMFCS I-III was 5.2 (4.5–6.3) mL/kg/min
Trost [54], 2016 Australia	n=47, 37% females; age=9-16 Y	I-III	Hemiplegia; Diplegia; Quadriplegia	Energy expenditure: MET calculated as mean VO2 value during each condition divided by predicted resting VO2 calculated from Schofield's equation [70] for youth with CP	Cosmed K4b2 portable indirect calorimetry system (Rome, Italy)	Lying down and handwriting[a]	N/A	The MET range for lying down for GMFCS I-II was 1.1-1.2 (1.0-1.4) MET and for GMFCS III was 1.0 (0.9-1.2) MET. The MET range for handwriting for GMFCS I-II was 1.2-1.3 (1.1-1.4) MET and for GMFCS III was 1.1 (0.9-1.2) MET.
		I-V		Energy expenditure:	Cortex Metamax	Sitting with support, sitting without support,	≤1.5 METs	The MET range for sitting with support ranged from 0.77 ± 0.57 MET for GMFCS V to 1.58 ± .46

Verschuren [55], 2014 Netherlands	n=19, 32% females; age=4-20 Y		Unilateral and bilateral spastic	METs were calculated as mean VO2 value during each condition divided by the mean VO2 value during lying for each child		Sedentary Behaviour Research Network [9]		MET for GMFCS IV. The MET range for sitting without support ranged from 1.11 ± .17 MET for GMFCS I to 1.33 ± .25 MET for GMFCS III.
				Muscle activation	“Porti” (TMS International)			Only muscle activity during standing in GMFCS III is significantly different from rest (ie., lying down) ($p<0.05$). Root mean square (RMS) for sitting with support ranges from 0.77 RMS for GMFCS V to 1.58 RMS for GMFCS IV. The RMS for sitting without support ranges 1.17 RMS for GMFCS I to 1.69 RMS for GMFCS III.
Williams [56], 2019 Australia	n=12, 53% females; age=16-25 Y	II-V	NR	Peak energy expenditure	ActiheartVR accelerometry	Sitting and lying	≤1.5 METs using (branched equation combined accelerometry counts and heart rate into metabolic equivalent of the task [75]	The peak energy expenditure and the parent-reported activity corresponding to the time of peak energy expenditure were reported. The peak energy expenditure for two individuals was in sitting and in lying. For one individual classified as GMFCS III, the peak energy expenditure of 5.9 METs was during sitting while for another individual classified in GMFCS V, the peak energy expenditure of was 5.98 METs was during lying.

[a]There was no designation of the stated postures as being “sedentary”, but rather the study validates postures typically characterized as sedentary

Table S2. Objective measurement tools for time spent in sedentary behaviour by individuals with cerebral palsy: Accelerometers and heart rate monitors

Author (year)	**Wear Time**			**Non-wear/sleep criteria**	**SB Threshold: Cut points/Epoch (axis where indicated)**	**SB Threshold Citation**	**Age group**	**Placement**
	Protocol	**Valid**	**Total Average[b]**					
			ActiGraph GT3X Triaxial Accelerometer					
Claridge [33], 2015	7 consecutive days	≥5 h on 4 consecutive days	Hip and wrist-worn accelerometers: 6.83 d (range: 422.9 to 934.7 min/d); Hip- and wrist-worn accelerometers, respectively: 682.4 (127.1) and 681.5 (128) min/d	Activity diary to track times and reasons for accelerometer removal and replacement	100 CPM VA and VM	Healy [1], 2011; Matthews [7], 2008	A/O	Hip and wrist
Oftedal [44], 2015	3 d during waking h (2 d wk and 1 d w/e)	3 d (>50% of waking h)	GMFCS: I–II 580 mins (89); GMFCS III: 591 mins (78)	Wear-time log	GMFCS I–III (age 18 m to 3 Y): 40 counts/5 sec;	Oftedal [59], 2014	T	Lower back (center)

					GMFCS IV–V (age 18 m to 3 Y): 10 counts/5 sec VM			
McPhee [40], 2017	7 consecutive days	≥5 h on 4 consecutive days	NR	NR	100 CPM VA	Freedson [71] 1998	A/O	Hip
ActiGraph GT3X+ Triaxial Accelerometer								
Halma [67], 2019	7 d	≥ 8 h during daytime	6.1 d (1.2)	Zero counts for ≥15 minutes	25 counts per 15 seconds VA	Evenson [73] 2008	T/C	NR
Mitchell [41], 2015	4 d during waking h (2 d wk and 2 d w/e)	1 d (>8 h/d)	11h 44 mins (11 h 22 mins, 12 h 05 mins)	Zero count for >20 minutes	100 CPM VA	Evenson [73], 2008	C	Least affected hip
Reedman [68], 2019	Waking hours for 7 consecutive days	≥10 h/d; episodes with at least 1 valid wear day were included in analyses	Intervention baseline: 5.87 d (2.29); Waitlist control baseline: 5.29 d (1.77)	≥60min of consecutive zero counts with allowance for 1-2min of nonzero counts<25 counts/15s)	40 counts/15 sec VA	Trost [54], 2016	C	Hip
ActiGraph GT3X and GT3X+ Triaxial Accelerometer								
Keawutan [36] 2017, Keawutan [37] 2018 [c]	3 d during waking h (2 d wk and 1 d w/e)	3 d (2 d wk and 1d w/e, (>6 h/d)	Wk: 10.9 h (1.6); W/e: 10.1h (2.3); GMFCS I: 10.6 h (1.4); II: 10.9 h (1.3); III: 10.9 h (1.4); IV: 10.1 h (0.9): V: 10.4 h (2.3)	Activity diaries	820 CPM VM	Butte [74], 2014	T/C	Lower back
Oftedal [60] 2016, Oftedal [61] 2017, Keawutan [62] 2017 [c]	3 d during waking h (2 d wk and 1 d w/e)	3 d (2d wk and 1 d w/e)	Mean wear time of 10 h a day for 3d	Activity diaries	GMFCS I–III (age 18 m to 3 Y): 40 counts/5 sec; GMFCS IV–V (age 18 m to 3 Y): 10 counts/5 sec; GMFCS I–V (age 4–5Y): 68 counts/5 sec VM	Keawutan [35] 2016, Oftedal [59] 2014	T/C	Lower back
ActiGraph GT1M Uniaxial Accelerometer								
Obeid [42] 2014	7 d during waking h	≥4d (≥5 h/d on 3 wk and 1 w/ e d)	720.0 mins/d (81.1), 6.2 d (0.9)	Activity diary to track of the waking hours and reasons for not wearing the device	100 CPM	Evenson [73], 2008	C	Right hip
ActiGraph 7164 Uniaxial Accelerometer								
Capio [31] 2012	7 d during waking h	5 d (5–18 h/d on 3 wk and 2 w/ e d)	11.72 h (2.11) on wk; 10.73 h (2.19) on w/e	Continuous counts of zero for ≥20 min	100 CPM	Evenson [73], 2008	C[a]	Hip (side NR)
Capio [65] 2015	7 d during waking h	5 d (5–18 h/d on 3 wk and 2 w/ e d)	NR	Continuous counts of zero for ≥20 min	100 CPM	Evenson [73], 2008	C[a]	Hip (side NR)

RT3 Triaxial Accelerometer								
Ryan [47] 2014	7 d during waking h	4 d (>10 h/d)	Median (IQR): 840.5 min/d (88.5) for 7.0 d (1.0)	Activity diary to record times that device was removed, and activities completed while not wearing monitor	100 CPM VM	Rowlands [76], 2004	A	Right hip (or least affected side in the case of asymmetry)
Ryan [48] 2014, Ryan [51] 2015, Ryan [52] 2015[c]	7 d during waking h	4 d (>10 h/d)	Median (IQR) 7.0 d (1.0); Mean (SD): 761.0 min/d (58.4)	Ryan [48] 2015, Ryan [52], 2015: Activity diary to record times that device was removed, and activities completed while not wearing monitor Ryan [51], 2015: ≥10 min of consecutive 0 counts, allowing for 1 min of ≤10 counts within a given 10-minute period.	41 CPM VM	Vanhelst [75], 2010	C	Right hip (or least affected side in the case of asymmetry)
ActiTrainer Triaxial Accelerometer								
Lauruschkus [66] 2017	7 d during waking h	≥5 h of wear time on ≥2 d	≥ 5h of monitoring time on ≥ 4d	NR	100 CPM	Freedson [51], 2005	C	Right hip
Bodyguard Heart Rate Monitor								
Riquelme [46] 2018	24 h	24 h	24 h	Device removed 24-hour later by trained member of the research team, ensuring wear	Intensity < 20% of MET maximal	NR	T/C	2 electrodes on skin: one on the right subclavicular area and another on left submamilar area
ActiheartVR: Heart Rate Monitor & Triaxial Accelerometer								
Williams [56] 2019	7 d	Over 3 d, with 1 d w/e, and ≥6 h on each day	3 full days (≥6 h/d) with 1 w/e day available from 11 participants, and 5 full days from 9 participants	Activity diary	≤1.5 METs per 15 sec	Ward [78], 2007	C/A	Chest

Early Years (0-4 years of age) (T), Children & Youth (5–17 years of age) (C), Adults (18–64 years of age) (A), Older adults (65+ years of age) (O)
[a] Mean age, age range NR.
NR: Not reported; CPM: Counts per Minute; wk: weekday; w/e: weekend
[b]Values are mean (SD) unless otherwise specified
[c]Publications grouped together are from the same study

Table S3. Objective measurement tools for time spent in sedentary behaviour by individuals with cerebral palsy: Activity monitors

Author (year)	**Wear time**			**Non-wear/sleep criteria**	**SB activities**	**Age group**	**Placement**
	Protocol	**Valid**	**Total Average[b]**				
activPAL							

Bania [63] 2016	7 consecutive days	≥2 d (24 h/d)	NR	Activity diary	NR	C/A	Thigh
Bania [30] 2014	7 consecutive days	≥ 2 d (24 h/d)	5.8 days (SD 1.3)	Activity diary	NR	C/A	Thigh
activPAL3							
Aviram [57] 2019, Bar-Haim [58] 2019[c]	4 consecutive days; allowed missing data from <4 h per nighttime and/or <2 h during daytime	NR (excessive missing data excluded)	NR	Activity diary (Sleep is sedentary activities >2.5 hours and began between 10 PM and 6 AM)	Nonupright inactivity (specifically stating sitting and lying)	C/A	Thigh
Shkedy [6] 2014	4 d	Not specified	4 d (SD 5h); majority of the records consisted of 2 d w/e, but on occasion consisted of 4 d wk	Activity diary	Sitting or lying down (Wheelchair mobility in this study was considered to be sedentary)	C/A	Thigh (less affected leg)
VitaMove							
Nooijen [5] 2014	72 h	24 hours	Measurement duration was 72 hours for 37%, 48 hours for 51% and 24 hours for 12% of participants	Activity diary	Lying and sitting	C/A	One recorder attached to each thigh and a third recorder attached to sternum
Slaman [64] 2015	3 consecutive weekdays	3 days for 24 consecutive hours a day	Successful measurement of 24h/day at baseline (# participants): 1 day (6), 2 days 25), 3 days (18)	Activity diary	Lying and sitting	C/A	One recorder attached to each thigh and a third recorder attached to sternum

Early Years (0-4 years of age) (T), Children & Youth (5–17 years of age) (C), Adults (18–64 years of age) (A), Older adults (65+ years of age) (O)
[a] Mean age, range NR.
[b]Values are mean (SD) unless otherwise specified
NR: Not reported
[c]Publications grouped together are from the same study

Table S4. Subjective measurement tool for sedentary behaviour in individuals with cerebral palsy

Author (year)	**Measurement tool**	**Sedentary Behaviour Activities**	**Age group**
Lauruschkus [66] 2017	International Physical Activity Questionnaire (IPAQ)	NR	C

Early Years (0-4 years of age) (T), Children & Youth (5–17 years of age) (C), Adults (18–64 years of age) (A), Older adults (65+ years of age) (O)
NR: Not reported

Table S5. Key findings of studies measuring time spent in sedentary behavior by individuals with cerebral palsy

Author (year)/country	**Demographics**	**GMFCS level**	**CP Subtype**	**Objectives**	**Methodology**	**Key Findings**
	n=54, 35% females;	I–III	Bilateral spastic	Aviram [57], 2019: To compare habitual physical activity (HPA) and sedentary	Cross-sectional	Teenagers and young adults with CP spend 53 more mins a day than TDP in SB and more time in

Aviram [57] 2019, Bar-Haim [58] 2019, Middle East[c]	age=12-20 Y CP training group; n=41 children with TD			behaviour (SB) between children and young adults with cerebral palsy (CP) and typically developing peers (TDP) Bar-Haim [58], 2019: (1) To evaluate the relationship between levels and changes HPA and motor function in adolescents and young adults with CP, before and after resistance and treadmill interventions (2) To differentiate these levels and changes between participants at GMFCS levels II and III and between the exercise interventions.		sedentary events longer than 120 mins (p=0.048). The shorter sedentary events (<60 min) were similar for the TD and CP groups. For GMFCS III, the total sedentary time was the same as for those at GMFCS II, but the percentage of sedentary time while awake was higher because they had less sleep. Individuals in GMFCS II and III spent 74.0% and 80.0% in sedentary time, respectively.
Bania [30] 2014, Australia	n=45, 49% females; age=14-22 Y	II-III	Spastic/ Bilateral	(1) To describe the daily PA levels of adolescents and young adults with bilateral spastic CP who have difficulty walking; (2) to determine which factors predict daily PA in this group	Cross-sectional	On average, participants spent 20.0 hours/day sitting/ lying.
Bania [63] 2016, Australia	n=36, 46% females; age=14-22 Y	II-III	Spastic/ Bilateral	(1) To determine whether a 12-week progressive resistance training programme for the lower limbs increased the daily PA levels of adolescents and young adults with CP with difficulty walking	Randomized controlled trial	At baseline, the intervention group spent a mean of 19.4 h/day of sedentary time while the control group spent a mean of 20.1 h/day sedentary time.
Capio [31] 2012, China	n=31, % females NR; age= 7.41± 2.48Y CP training group; n=31 children with TD	I-III	NR	(1) To measure the PA of a sample of children with CP, examine the association between PA and fundamental movement skills proficiency, and compare the data with TD children	Cross-sectional	Children with CP spend a higher percentage of time sedentary compared to the TD group (p = .014) and spent a greater percentage of time in sedentary activity on weekends compared to weekdays (p <0.001)
Capio [65] 2015, China	n=24, 50% females; age= 6.92±3.04 Y CP training group; n=26 children with TD	I-III	NR	(1) To explore the causal mechanism between fundamental movement skills and PA in children with and without disability	Pre-post-test	For both children with CP and their TDP, the percentage of sedentary time was higher on weekends compared to weekdays.
Claridge [33] 2015, Canada	n=42, 50% females; age=18-75 Y	I-V	Spastic, mixed, other (Other refers to non-spastic forms of CP were ataxic and dyskinetic CP); unilateral, bilateral; unknown	(1) To measure HPA and sedentary time in adults with CP across the severity spectrum; (2) to evaluate the effect of using different accelerometer-based activity counts on MVPA levels and to assess the validity of the Physical Activity Recall Assessment for People with Spinal Cord Injury tool compared to accelerometer measures of HPA	Cross-sectional	Adults classified in GMFCS levels IV and V spent more sedentary time and had a lower frequency of sedentary breaks compared to those in GMFCS levels I, II, and III. The overall sample spent a mean of 10.5 h/d in sedentary time.

Halma [67] 2019 Netherlands	n=65, 43% females; age=4-12 Y	I-III	Spastic/ Unilateral, bilateral	(1) To examine if changes in motor capacity outcomes are accompanied by a change in objectively measured motor performance immediately after a 3-month intensive treatment period in ambulatory children with CP, and 3 months thereafter	Quasi-randomised trial	On average, children with CP spent 72.1% of their waking hours in sedentary time at baseline.
Keawutan [36] 2017, Keawutan [37] 2018 Australia[c]	n=67, 36% females; age=4-5 Y	I-V	Spasticity, dystonia; ataxia; hypotonia / Unilateral; bilateral	Keawutan [36], 2017: To objectively measure HPA and SB levels in children with CP aged 4 to 5 years across all functional abilities and compare them to the Australian Physical Activity Guidelines Keawutan [37], 2018: To investigate the relationship between HPA, sedentary time, motor capacity, and capability in children with CP across all functional abilities	Cross-sectional	Children with CP spent 66.9% of their waking hours sedentary on weekdays, compared to 64.9% on weekends (p=0.041). Boys with CP spent less time being sedentary than girls. Children in GMFCS I-II, III and IV-V spend 57.6%, 73.6%, and 92.7% of their time sedentary, respectively. Children in GMFCS III-V spend significantly more time being sedentary than those in GMFCS I ($p \leq 0.001$). For all 3 days of monitoring, 99% and 86% of children in GMFCS I-II and GMFCS III, respectively, spent ≤ 60 minutes in total sedentary time, while almost all children in GMFCS IV-V spent over 60 minutes in total sedentary time.
Oftedal et [61] 2017, Oftedal [60] 2016, Keawutan [62] 2017 Australia[c]	n=95, 35% females; age=2-5 Y	I-V	Bilateral and unilateral spastic; Dyskinetic; Ataxic, hypotonia	Keawutan [62], 2017: (1) To describe HPA and SB in young children with CP from 1 year 6 months to 5 years of age; (2) To compare HPA and SB between time points; (3) To examine the rate of change in HPA and SB across all gross motor functional abilities Oftedal [60], 2016: (1) To investigate the longitudinal relationship between stature, growth velocity, energy intake, HPA, and sedentary time for each GMFCS level in children with CP Oftedal [61], 2017: 1) To investigate the longitudinal relation between anthropometric and body-composition measures and modifiable lifestyle factors across the spectrum of functional capacity; 2) To compare anthropometric and body-composition measures between children with GMFCS I–V with those of age- and sex-specific reference children with TD	Longitudinal	Children in GMFCS III-V spent more time sedentary than those in GMFCS I-II at all timepoints between 18months -5 y. On average, percentage of time spent sedentary during waking hours between 18months -5 y increased by 2.4%/year for GMFCS I-II and by 6.9%/year for GMFCS III-V. Children with CP aged 18–24 months, 30–36 months, 48 months and 60 months spend 56%, 59%, 66% and 66% of their time sedentary, respectively.
Lauruschkus [66] 2017, Sweden	n=11, 55% females; age=7-11 Y	I-V	NR	(1) To evaluate the feasibility of the Physical Activity on Prescription intervention, which focuses on increasing PA participation and reducing SB, for children with CP and its effectiveness on participation in PA and SB.	Pre-post-test	The median baseline sedentary time for GMFCS I-II was 360mins/day, as measured by the International Physical Activity Questionnaire (IPAQ), 543 mins/day by diary and 464 mins/day by accelerometer. The median baseline sedentary

						time for GMFCS III-V was measured to be 240 mins/day by IPAQ, 305 mins/day by diary and 673 mins/day measured by accelerometer.
McPhee [40] 2017, Canada	n=41, 51% females; age=18-75 Y	I-V	Spastic, dyskinetic, or ataxic/ unilateral, bilateral	(1) To describe the experience of fatigue in adults with CP across all GMFCS levels; (2) To determine if PA level, sedentary time, age, waist circumference, and/or BMI can predict fatigue in adults with CP	Cross-sectional	The mean time spent in SB was 55.7 (SD: 4.29) min/hour.
Mitchell [41] 2015 Australia	n=102, 49% females; age=8-17 Y	I-II	Spastic hemiplegia	(1) To assess PA of independently ambulant children and adolescents with unilateral CP and to determine the proportion adhering to the recommended level of MVPA (60 minutes daily); (2) To examine PA and inactivity in this population by comparing: (a) GMFCS levels I and II, (b) children and adolescents, (c) boys and girls, and (d) weekday and weekend activity	Cross-sectional	Participant displayed most SB at 9 am and between 7 to 9 pm on weekdays. On a typical day, participants spent 8:36 (SD 1:09) hours or 72% of their waking hours in SB. SB was highest in adolescents and girls. Boys performed more activity than girls and adolescents s (≥13 y) performed significantly less activity than children (<13 y) ($p<0.001$).
Nooijen [5] 2014, Netherlands	n=48, 52% females; age= 16-24 Y CP training group; n=32 children with TD	I-III	Spastic/ Unilateral, bilateral	(1) To assess physical behaviour of ambulatory adolescents and young adults with spastic CP.	Randomised controlled trial	Compared to their TDP, individuals with CP spent 80 minutes more being sedentary (i.e., sitting and lying) per 24 hours ($p < 0.01$) but did not spend more % time just sitting ($p=0.79$). The number of short sitting bouts (0-10sec and 10-60 sec) were significantly higher in individuals with bilateral CP compared to unilateral CP ($p < 0.05$).
Obeid [59] 2014, Canada	n=17, 12% females; age= 8-17 Y CP training group; n=17 children with TD	I-III	Spastic, dyskinetic/ Bilateral, unilateral	(1) To objectively measure time spent sedentary and frequency of breaks from sedentary time in ambulatory children and adolescents with CP compared with age-, sex-, and season-matched youth with typical development	Cross-sectional	Children with CP engaged in significantly more sedentary time (min/h) ($p=0.02$) compared with the TD group, with significantly fewer sedentary breaks (n/h of sedentary, $p=0.03$).
Oftedal [44] 2015, Australia	n=58, 46% females; age= 1-3 Y CP training group; n=20 children with TD	I-V	NR	(1) To compare children with CP according to functional capacity and TD in the following: (i) sedentary time, (ii) duration of sedentary bouts and breaks in sedentary time; and (iii) levels of HPA and SB compared to Australian PA recommendations	Cross-sectional	Children in GMFCS III was more sedentary than those who were TD and in GFMCS I-II (62% of waking hours being sedentary, $p<0.05$). The GMFCS IV–V group was more sedentary than the GMFCS I-III group (74% of day being sedentary, $p<0.05$). The GMFCS IV–V group was more likely to have sedentary bouts ≥60 min or longer compared to all other groups ($p<0.05$). For children in the TD group, ambulatory, or marginally ambulatory groups, all sedentary bouts were ≤60 min during the 3-day measurement period. On the other hand, a third of the children in the non-ambulatory CP group had only 1 out of 3

						days of wearing the accelerometer in which a sedentary bout did not exceed 60 min.
Reedman [68] 2019, Australia	n=37, 51% females; age=18-75 Y	I-III	Spastic, other/Unilateral, bilateral	(1) To determine efficacy of participation-focused therapy on participation in leisure-time PA goals; (2) To objectively measured PA behaviors in children with CP	Randomized waitlist-controlled	At baseline, the intervention group spent a mean of 435 mins/day in sedentary time while the waitlist control group spent a mean of 434 mins/day sedentary time.
Riquelme [46] 2018, Spain	n=26, 27% females; age= 4-16 Y CP training group; n=26 children with TD	I-V	Bilateral spastic, Unilateral spastic; Dyskinetic; Ataxic	(1) To compare activity between children with CP and TDP, with and without chronic pain	Cross-sectional	No information was reported on time spent in SB. The study reported differences in sedentary time between individuals with CP and TDP with chronic pain and between individuals with CP and TDP without chronic pain (reported in Table S7).
Ryan [47] 2014, Republic of Ireland	n=41, 54% females; age=18-62 Y	I-III	Bilateral or unilateral spastic	(1) To compare levels of PA and SB between adults with and without CP; (2) To examine the relationship between PA levels, SB and cardiometabolic risk factors in adults with CP	Cross-sectional	Adults with CP spent more sedentary time than their TDP ($p < 0.001$). Adults with CP in GMFCS I did not spent more sedentary time than their TDP while those in GMFCS II did ($p < 0.001$). Adults in GMFCS III spent significantly more sedentary time than adults in GMFCS II ($p < 0.05$). Adults with unilateral spastic CP spent less sedentary time than adults with bilateral CP. Percentage of waking hours spent in sedentary time for individuals classified in GMFCS I, II and III are 61.95%, 60.95% and 69.17%, respectively. The corresponding daily time spent in SB for GMFCS I, II and III are 505.3 min/day, 506.1 min/day and 579.0 min/day, respectively.
Ryan [48] 2014, Ryan [51] 2015, Ryan [52] 2015, Republic of Ireland[c]	n=90, 37% females; age=6-71 Y	I-III	Bilateral or unilateral spastic; non-spastic forms	Ryan [48], 2014: (1) To investigate the prevalence of overweight/obesity and elevated blood pressure among a cohort of ambulatory children with CP and (2) to investigate the association among PA, SB, overweight/ obesity, and blood pressure in children with CP Ryan [51], 2015: (1) To utilize accelerometry to describe light, moderate, and vigorous PA and SB in preadolescent children with and without CP and compare PA and SB between the 2 groups. Ryan [52], 2015: (1) To determine the association between SB, PA and cardiorespiratory fitness in children with CP; (2) To determine the association between cardiorespiratory fitness, anthropometric measures and blood pressure in children with CP	Cross-sectional	There was a significant difference in sedentary time across GMFCS levels ($p=0.022$). Percentage of waking hours spent in sedentary time for individuals classified in GMFCS I, II and III are 31.7%, 37.2% and 42.3, respectively. Individuals with CP with low, middle and high cardiorespiratory fitness, spent 29.0%, 33.6% and 32.8% of their waking hours being sedentary. Time spent sedentary significantly differed between individuals CP and their TDP, with 193 min/day (SD: 68.03 mins) or 25.5% mean sedentary time, compared to the TD group, with 123 min/day (SD: 49 mins) or 17.54% mean sedentary time ($p < 0.01$).

Slaman [64] 2014, Netherlands	n=11, 55% females; age=7-11 Y	I-IV	Spastic	(1) To evaluate the effectiveness of a lifestyle intervention programme on physical behaviour.	Randomized controlled trial	Children with CP spent 41.09% of their waking hours sedentary and had 3.02 sitting periods lasting>30 mins over 24 hours
Shkedy [6] 2014, Middle East	n=222, 41% females; age=13-22 Y	II-IV	Spastic, mixed/ bilateral (diplegia, quadriplegia)	(1) To document HPA and SB in adolescents and young adults with CP, and compare patterns in individuals with different levels of mobility dysfunction; (2) to document the long-term HPA of adolescents and young adults with CP and to compare individuals in educational institutions with those who were homebound	Cross-sectional	Adolescents and young adults classified as GMFCS II, III and IV spent 82.5%, 86.6% and 95.7%/24 hours in sedentary time, respectively; The corresponding daily sedentary times are 122 min, 158 min and 514 min, respectively. Those in GMFCS III spend significantly more sedentary time than those in GFMCS II while those in GMFCS IV spend significantly more sedentary time compared to those in GMFCS II and III.
Williams [56] 2019, Australia	n=12, 50% females; age=12-19 Y	II-V	NR	(1) To describe time spent in sedentary, light, and moderate-vigorous PA, percentage body fat, body mass index, and nutritional intake in adolescents with CP; (2) To compare these values with guidelines and CP specific recommendations	Cross-sectional	Adolescents with CP spent a median of 413.3 min per 12-h day or 57.4% of a 12-h day engaged in SB. They exceeded 30 min bouts of SB a median of 2 times per day (range 0–10). Only 9% of participants met the SB guidelines of <120 min of sedentary behaviour per day.

NR: Not reported

[c]Publications grouped together are from the same study

Table S6. Description of reliability and validity of tools for measuring and classifying sedentary behaviour in individuals with cerebral palsy

Author (year)/ Country	**Demographics**	**GFMCS Level**	**CP Subtype**	**Description of Measurement Tool**	**Method of Determining Validity/ Reliability**	**Operationalized definition of Sedentary Behaviour**		**Results of Validity/ Reliability**	**Rating of Results**
						SB threshold (cut-points/ epoch) being tested	**Criterion SB activities**		
Ahmadi [27] 2018, Australia	n=22, 50% females; age=6-20 Y	I-III	NR	Machine learning algorithms: Models that classify physical activity (PA) type and predicts energy expenditure or PA intensity using specific regression equations for results from the ActiGraph GT3X+ triaxial accelerometer	Criterion validity: Three types of machine learning algorithms, binary decision trees (BDT) vs. support vector machines (SVM) vs. random forests (RF) placed on the hip vs. wrist vs. both, were validated against video observation	N/A	Supine rest, seated handwriting	Represented with confusion matrices, there was excellent agreement between video observation and machine learning model prediction of sedentary activities ranging between 0.95-0.97 for wrist, 0.95-0.96 for hip, and 0.94-0.98 for hip and wrist. For classification of sedentary activities, there was no differences between different types of machine learning algorithms and placement; however, SVM and RF outperformed BDT, and both hip and wrist outperformed either one alone for non-sedentary.	Excellent agreement between all models and locations of placement with video observation for supine rest and handwriting.

Bania [29] 2014, Australia	n=10 validity, n=24 reliability, 50% females; age=14-22 Y	II-III	Spastic diplegia	ActivPAL: AM	Criterion validity: AM data was compared against video observation	N/A	Sitting	A Bland-Altman analysis showed a systematic difference in activity monitor and video observation, but the mean difference was small.	NR
					Test-retest reliability: AM data was conducted at the beginning and at the end of 6 days	N/A	Sitting and lying	Retest reliability was moderate for time spent in sitting and lying (ICC: 0.60–0.66). There were no systematic changes between measurements 6 days apart. Across the groups, changes in time spent in sitting and lying of 1% could be considered as being within measurement error, with 95% confidence. For individuals, changes in time spent in sitting and lying of 11% could be considered as being within measurement error, with 95% confidence.	There is substantial relative test-retest reliability for time spent sitting and lying.
Clanchy [32] 2011, Australia	n=29, 43% females; age=8-16 Y	I-III	Spastic	ActiGraph 7164: Uniaxial accelerometer	Criterion validity: Several accelerometer cut-points for SB were validated against indirect calorimetry	Evenson [73]: s ≤ 100 CPM; Treuth [106]: SED ≤ 100 CPM; Freedson [72]: ≤ 100 CPM; Puyau [107]: VA < 800 CPM; Current study: ≤ 100 CPM	Resting protocol	Classification accuracy, shown with ROC-AUC, is excellent (>90%) for all models. The Puyau [107] cut point had the lowest classification accuracy (90%) compared to the other three models (92%) (Freedson [72], Evenson [73], Treuth [106], current study).	All models displayed excellent classification accuracy for the resting protocol.
Claridge [8] 2019, Canada	n=14, 36% females; age=18+ Y	I-III	Spastic diplegia, hemiplegia, quadriplegia	Activ8: AM	Criterion validity: AM data from three different body placements was compared against video observation for typically sedentary postures	N/A	Sitting[a]; SB Break: Upright activity	The Spearman rho correlation coefficient between AM and video observation for sitting, positioned at frontolateral thigh, frontal thigh and pant pocket were 0.99 (significantly higher correlation p < 0.01), 0.72 and 0.79, respectively. Lying is unable to be distinguished from sitting.	The agreement between AM and video observation at 3 different location was substantial for the frontal thigh and pant pocket and excellent for the frontolateral thigh in the sitting posture.

Gerber [69] 2019, Switzerland	n=15 validity, n=10 reliability, 60% females; age=8-20 Y	I-III	Affected sides: right, left and both	Set-up comprised of 5 inertial measurement unit devices (Physilog4, Gait Up, Renens, Switzerland): set-up of 5 sensors attached to different body parts, with each sensor device (called the Physilog4) composed of a triaxial accelerometer, triaxial gyroscope, and barometer to detect walking episodes	Construct validity: Measurement of a sitting and lying is compared between two school days vs a school day and a weekend	N/A	Sitting and lying[a]	With the instrument, the agreement (ICC) of the sedentary behaviour levels between a school day and a weekend was 0.30 (0.00–0.73) and 0.47 (0.00–0.80) between two school days of the same week.	There was fair agreement between sedentary levels on a school day vs weekend and moderate agreement between two school days of the same week.
					Test-retest: Measurement of sitting and lying is compared between the same standard school day 2 to 4 weeks apart	N/A		The agreement (ICC) between the same standard school day 2 to 4 weeks apart was 0.90 (0.64–0.97).	There was excellent agreement between sedentary levels on the same standard school day 2 to 4 weeks apart
Hegde [34] 2018, USA	N=10, 40% females; age=4-9	I-II	Some with spastic; diplegia, hemiplegia	Pediatric SmartShoe: five pressure sensors and a 3-D accelerometer that can differentiate between major postures and activities, accurately estimate energy expenditure, measure temporal gait parameters, and estimate body weight	Criterion validity: AM data was compared against direct observation for sedentary postures	N/A	All activities including typically SB were included in the results[b]: 1. Sitting in different contexts (i.e., on child chair, adult chair, parent's lap, floor playing with toys) 2. Standing 3. Walking	A leave-one-out cross validation method yielded an overall classification accuracy of 95.3% for sitting, standing and walking. There were no separate results for sitting.	There was excellent agreement between the Pediatric SmartShoe and in-person observations for all activities measured.
Keawutan [35] 2016, Australia	n=84, 43% females; age=4-5 Y	I-V	Unilateral spasticity, bilateral spasticity, dystonia, ataxia, hypotonia, and athetosis	ActiGraph GT3X and GT3X+: Triaxial accelerometers	Criterion validity: Several accelerometer cut-points for SB were validated against video observation	Butte [74]: 68 counts/ 5 seconds; Current study GMFCS I: 100 counts/ 5 seconds, GMFCS II: 93 counts/ 5 seconds, GMFCS III:	Stationary with or without limb movement	The Butte cut point [73] demonstrated lower bias in GMFCS I, II, IV, and V and narrower 95% limits of agreement in GMFCS I, II, III, and V. The CP cut-point for GMFCS I for the current study significantly overestimated time spent sedentary (p = 0.037). The Butte cut-point overestimated time spent sedentary for GMFCS I, but this difference was not statistically significant (p = 0.091). In the GMFCS	NR

						63 counts/ 5 seconds, GMFCS IV: 60 counts/ 5 seconds, GMFCS V: 16 counts/ 5 seconds		II-IV groups, no significant differences in time spent sedentary between observed and predicted values were found for the current study and Butte cut-off points [74]. For the current study's cut-off points, there was a small bias but wide 95% limits of agreement, which indicates that although the results were accurate on a group level, the difference between observed and predicted time spent sedentary for an individual child may be significant.	
Koehler [38] 2015, Germany	n=10, 30% females; age=13.4 ± 1.6 years Y	II	Hemiplegia	SenseWear Armbands: Multisensor device, worn on the upper arm, that combines accelerometry data with information from several heat-related channels to estimate energy expenditure	Criterion validity: AM data was compared against indirect calorimetry for sedentary postures	N/A	Sitting[a]	There was no significant difference between energy expenditure measured by indirect calorimetry and the estimates of energy expenditure provided by the Sensewear armband on the non-hemiparetic (95% confidence interval: –0.7, 1.3) nor the hemiparetic side (95% confidence interval: (–0.7, 1.5).	NR
McAloon [39] 2014, United Kingdom	n=10, % females NR; age=4-18 Y	I-III	Hemiplegia; Diplegia; Quadriplegia	ActivPAL: AM	Criterion validity: AM data was compared against video observation for sedentary postures	N/A	Sitting	'Sit' was misclassified as a stand which resulted in greater intervals of standing time being recorded. There is wide 95% limits of agreement for sitting, indicating greater systematic variation.	NR
O' Donoghue [43] 2014, Republic of Ireland	n=17, 47% females; age=5-18 Y	I	Hemiplegia; Asymmetric Diplegia	activPAL: AM	Criterion validity: AM data was compared against video observation for sedentary postures	N/A	Sitting[a]	There were moderate ICC results (ICC: 0.49) and high ICC results (ICC: 0.95) in the first and second test protocols, respectively. These results showed moderate and high agreement of the activPAL AM with observational values, respectively. A Bland-Altman analysis showed that the AM overestimated sitting time to a clinically significant degree.	There was moderate agreement between observation and AM results for test 1 and excellent agreement for test 2 for sitting.
Oftedal [59] 2014, Australia	n=103, 61% females; age=2-3 Y	I-V	Spastic, dyskinetic, hypotonic/ diplegia,	ActiGraph GT1M: Uniaxial accelerometer	Criterion validity: Several accelerometer cut-points for SB were validated against video observation	Uniaxial accelerometer (ActiGraph GT1M), GMFCS I-V:	Lying or sitting with or without limb movement, or standing completely still	A Bland-Altman analysis showed that the ActiGraph GT1M significantly overestimated time spent sedentary using the uniaxial cut-point for children in GMFCS I-III (p<0.001),	NR

			hemiplegia, quadriplegia	ActiGraph GT3X and GT3X+: Triaxial accelerometers		2 counts/5 sec; Triaxial accelerometer (GT3X and GT3X+), GMFCS I-III: 40 counts/ 5 sec, GMFCS IV-V: 10 counts/ 5 sec		whereas the GMFCS IV–V cut point only showed a small bias. The difference between observed and predicted time spent sedentary for the triaxial cut points for GMFCS I–III and GMFCS IV–V were minimal and nonsignificant, with similar limits of agreement.	
O'Neil [45] 2016, USA	n=57, 51% females; age=6-20 Y	I-III	Spastic hemiplegia	ActiGraph GT3X: Triaxial accelerometer: ActiGraph data analyzed in this study were step counts, VA counts, and VM outputs Sensewear armband monitor: Uses multiple sensors to record acceleration; can provide various types of data including energy expenditure and step count StepWatch: Uniaxial monitor that records step counts	Concurrent validity: The three accelerometer results for a range of activities were compared with VO2 output	N/A	All activities including typically SB were included in the results[b]: 1. Supine rest 2. Writing 3. Wiping a table 4. Folding laundry 5. Active video game 6. Active video game 7. Comfortable/slow walk 8. Brisk walk 9. Fast walk	The concurrent validity ranged from 0.82-0.85 for the ActiGraph, 0.77-0.79 for the StepWatch and 0.73-0.75 for the Sensewear for all activities. There were no separate results for supine rest and writing.	The concurrent validity is good for the ActiGraph, fair for the StepWatch and for the Sensewear in measuring all included activities.
					Inter-instrument reliability was calculated by comparing left-to-right accelerometer placement for step count and VA+VM (ActiGraph only) for each of the three accelerometers tested	N/A		ActiGraph and StepWatch accelerometers demonstrated the highest levels of inter-instrument reliability between measurements of the right and left side, with ICCs exceeding .975, whereas inter-instrument reliability for the SenseWear monitor was slightly lower (ICC=.94). The reliability was calculated for all activities. There were no separate results for supine rest and writing.	The inter-rater reliability is excellent for VA, VM (ActiGraph only) and step counts between placement on the left and on the right side of the body for all three accelerometers in measuring all included activity.

Ryan [49] 2014, Ryan [50] 2014, Republic of Ireland[c]	n=36, 56% females; age=6+ Y	I-III	Unilateral and bilateral spastic	Sensewear ProArmband: Multisensor device, worn on the upper arm that combines accelerometry data with information from several heat-related channels to estimate energy expenditure Intelligent Device for Energy Expenditure and Activity: Collects raw data from five accelerometer sensors and converts it to energy expenditure RT3 accelerometer: Triaxial accelerometer	Criterion validity: The level of agreement for detecting energy expenditure while at rest was compared between three activity monitors and indirect calorimetry. Several cut-points for SB for the RT3 accelerometer were validated against indirect calorimetry.	RT3 accelerometer: < 41 CPM Vanhelst cut-point [75]; Current study cut-off point < 51.9 CPM	Supine position[a] and energy expenditure threshold of < 2.0 MET	There was a significant difference between the results of the Sensewear ProArmband and indirect calorimetry (p<0.01) but no difference between Intelligent Device for Energy Expenditure and Activity and RT3 accelerometer with indirect calorimetry (no specification of cut-point used). The study's cut-point had excellent classification accuracy, based on a ROC-AUC of 96.5% used to derive the cut-point. The Cohen's kappa coefficient, which indicated degree to which published cut-points applied to the sample of children and adolescents with CP, was excellent for the study by Valnhelst [75] and the study by Ryan [49, 50] [0.92 (0.82 - 1.00) and 0.96 (0.89 - 1.00), p < 0.001, for the cut points by Vanhelst [75] and the study, respectively].	There was an excellent ability of the RT3 accelerometer to distinguish SB using the study's cut point. Both the Vanhelst [75] and study cut-points had excellent applicability to children and adolescents with CP.
Tang [53] 2013, United Kingdom	n=15, 27% females; age=5-17 Y	I-III	Diplegia, hemiplegia, quadriplegia	activPAL: AM	Criterion validity: AM data was compared against video observation for sedentary postures	N/A	Sitting and lying[a]	The agreement between AM outcomes and video observations was 97.4%±2.7% for sitting/lying using a simple correlation calculation.	There was excellent agreement between the AM output and video observations for sitting and lying.
Trost [54] 2016, Australia	n=47, 37% females; age=9-16 Y	I-III	Hemiplegia; Diplegia; Quadriplegia	ActiGraph GT3X: Triaxial accelerometer	Criterion validity: Several accelerometer cut-points for SB were validated against indirect calorimetry.	Mean VA counts Evenson [73]: < 25 counts per 15; Mean VA counts Clanchy [32]: < 25 counts per 15 s; Current study: VA < 8 counts per 15 s, VM < 72 counts per 15 s	Supine rest, seated handwriting	For all 4 cut points, the misclassification rate for sedentary activities was relatively low at < 12% and classification accuracy, shown with ROC-AUC, was excellent (>90%). Classification accuracy was highest for the VA decision tree model (97%), second for the VM decision tree model (96%) and tied for the Clanchy [32] and Evenson models [73] (93%).	All models displayed excellent classification accuracy for sedentary activities.

NR: Not Reported; VA: Vertical axis; VM: Vector magnitude; CPM (counts per minute); AM: Activity Monitor
[a]There was no designation of the stated postures as being "sedentary" by the study authors, but rather the study validates postures typically characterized as sedentary
[b]No results were provided on the validity and/or reliability solely for the sedentary activities, but rather on the range of activities of varying intensities

Table S7: Description of relationship between sedentary behavior and health indicators in individuals with cerebral palsy

Author (year)/ Country	**Demographics**	**GMFCS Level**	**CP Subtype**	**Health Indicator**	**Relationship between Health Indicator & Sedentary Behaviour**	**Association between SB and Health Indicator (+/-/none/NR)**
			Cardiometabolic Disease Risk Factors			
Aviram [57] 2019, Bar-Haim [58] 2019, Middle East[c]	n=54, 35% females; age=12-20 Y CP training group; n=41 children with TD	II–III	Bilateral spastic (38 can walk w. limitations, 16 require mobility devices)	BMI	Increased sedentary time is associated with higher BMI and is fair in strength (r=0.38, p= 0.004).	**+**
Bania [30] 2014, Australia	n=45, 49% females; age=14-22 Y	II-III	Bilateral spastic	Height	Height was not significantly correlated to time spent sitting or lying (Pearson r = -0.07, p>0.05)	None
McPhee [40] 2017, Canada	n=41, 51% females; age=18-75 Y	I-V	Spastic, dyskinetic, or ataxic/ unilateral, bilateral	BMI	Sedentary time per hour was not significantly related to BMI (B= 0.066, p>0.05)	None
				Waist circumference	Sedentary time per hour was not significantly related to waist circumference (B= 0.107 p>0.05)	None
Oftedal [60] 2016, Oftedal [61] 2017, Keawutan [62] 2017, Australia[c]	n=95, 35% females; age=2-5 Y	I-V	Bilateral spastic, Unilateral spastic; Dyskinetic; Ataxic, hypotonia	Height-for-age z score (HZ)	Percent time spent sedentary was not significantly correlated to HZ before (r=–0.01, p=0.14) and after adjusting for GMFCS level (r=–0.008, p=0.34)	None
				Fat-free mass controlled for height	The percent time spent sedentary was weakly correlated to fat-free mass controlled for height (r=-0.03, p=0.003), but not when adjusted for GMFCS level (r=-0.003, p=0.841)	**-** but none when adjusted for GMFCS level
				Body fat percentage	Percent time spent sedentary was moderately correlated to body fat, % (r=0.2, p<0.001), but not when adjusted for GMFCS level (r= 0.002, p=0.965)	**+** but none when adjusted for GMFCS level
Ryan [47] 2014, Republic of Ireland	n=41, 54% females; age=18-62 Y	I-III	Bilateral spastic, Unilateral spastic	C-reactive protein levels	SB was not associated with high risk c-reactive protein*	None
				Metabolic syndrome (the presence of three or more of the following: (1) central obesity, (2) elevated triglycerides (≥150 mg/dL [1.7 mmol L–1]) or drug treatment for elevated triglycerides, (3) reduced height	SB was not associated with metabolic syndrome*	None

				density lipoprotein cholesterol (HDL-C) (<40 mg/dL [1.0 mmol L–1] in men; <50 mg/dL [1.3 mmol L–1] in women) or drug treatment for reduced HDL-C, (4) elevated blood pressure (systolic ≥ 130 and/or diastolic ≥ 85 mm Hg) or antihypertensive drug treatment, and (5) elevated fasting glucose (≥100 mg/dL) or drug treatment for elevated glucose) [81]		
Ryan [48] 2014, Ryan [51] 2015, Ryan [52] 2015, Republic of Ireland[c]	n=90, 37% females; age=6-71 Y	I-III	Unilateral spastic, bilateral spastic CP; non-spastic forms	Blood pressure	Increased SB is associated with elevated blood pressure (B=0.064, p=.047)	**+**
				Cardiorespiratory fitness (10 m shuttle run test)	Sedentary activity was not associated with cardiorespiratory fitness*	None
Williams [56] 2019, Australia	n=12, 50% females; age=12-19 Y	II-V	NR	Body fat percentage	No statistically significant associations were found between SB and percentage body fat (Spearman's ϱ=-0.36, p=0.31)	None
			Pain			
Riquelme [46] 2018, Spain	n=26, 27% females; age= 4-16 Y CP training group; n=26 children with TD	I-V	Bilateral spastic, Unilateral spastic; Dyskinetic; Ataxic	Chronic Pain (Self-reports of pain, lasting more than 3 months, and fatigue experienced during each activity in monitoring period in an 11-point faces scale)	Children with CP and chronic pain spent less time sedentary (p=0.05) than their TDP with chronic pain; no significant differences in sedentary times were found for children with CP and their TDP without pain. No significant differences were observed for energy expenditure at sedentary level between children with CP with or without chronic pain or between children with CP and TD children.	NR
			Fatigue			
McPhee [40] 2017, Canada	n=41, 51% females; age=18-75 Y	I-V	Spastic, dyskinetic, or ataxic/ unilateral, bilateral	Fatigue measured through the Fatigue Impact and Severity Self-Assessment (FISSA) questionnaire	Sedentary time per hour was not significantly related to FISSA scores (B=2.1, p=0.060)	None

[c]Publications grouped together are from the same study
NR: Not reported
*No value for correlation nor statistical significance provided

Chapter 2: Identifying Critical and Important Health Outcomes for Physical Activity Promotion in Cerebral Palsy Using Perspectives from Clinicians and Researchers: A Conference-Based Survey

Abstract

Background

Physical activity (PA) helps reduce negative health outcomes in individuals with cerebral palsy (CP), a population already at higher risk because they perform lower PA. Knowing the health outcomes relevant to PA is necessary towards endeavours such as clinical practice guidelines that help clinicians create management plans to increase PA in the population.

Objective

To rate the importance of pre-determined health outcomes, related to PA, based on expert opinions for individuals with CP, across all ages

Methods

We developed a self-administered electronic survey consisting of 9 health outcomes that cover both the physical and mental aspects of health informed by prior literature. The 9 health outcomes included were: (1) cardiorespiratory endurance; (2) body size (i.e., height and weight) ; (3) body composition (i.e., waist circumference); (4) sleep; (5) nutrition; (6) blood lipids and glucose; (7) blood pressure; (8) depression; (9) anxiety. Ratings for the nine outcomes lie on a 9-point Likert-type scale (1=least important, 9=most important) as set out by the Grading of Recommendations Assessment, Development and Evaluation (GRADE) approach. Additional health outcomes not listed in the survey were collected through a free-text box. The open survey was conducted at the combined annual American Academy for Cerebral Palsy and Developmental Medicine and triennial International Alliance of Academies of Childhood Disability Meeting. Registered experts to a conference workshop, primarily physicians,

occupational therapists, physiotherapists, nurses and researchers, were invited to participate in this survey; those attending the workshop completed the survey on-site. The results were descriptively analyzed by health outcome. Statistical analysis was conducted to determine pairwise differences between critical and important health outcomes using the Kruskal-Wallis test with Bonferroni correction. Based on the scale from the GRADE approach, ratings of 4-6 were "important" and ratings of 7-9 were "critical" for decision-making and should be included in the evidence profile for future clinical practice guideline development.

Results

Demographic information was available from all 75 experts who registered for the workshop. Respondents were from North America (60.0%), Asia (17.3%), Europe (13.4%), Oceania, (8.0%), and South America (1.3%). The clinical and research backgrounds represented were primarily physiotherapists (42.7%), followed by physicians (22.7%), individuals with solely Master's or PhD degrees (20%), occupational therapists (9.3%), and nurse-practitioners (2.7%). A convenience sample of 55 participants out of 75 registered experts actually attended the workshop and completed the survey. They rated five health outcomes as critical (i.e., sleep; nutrition, cardiorespiratory endurance, depression; anxiety) and four as important (i.e., body size, body composition, blood lipids and glucose, blood pressure). Pairwise comparisons showed that five critical health outcomes (i.e., cardiorespiratory endurance, sleep, nutrition, depression and anxiety) (median rating=7) were each rated higher than four important health outcomes (i.e., body size, body composition, blood lipids and glucose, and blood pressure) (median rating=5, except for blood pressure where median rating=6, $p<0.05$). The exception was between the rating for sleep and for nutrition, both rated critical outcomes, and blood pressure, rated an important outcome ($p>0.05$). Free-form responses from 6 respondents identified the following additional health outcomes for consideration: (1) bone density (n=1); (2) quality of life (n=1); (3) emotional well-being (n=1); (4) global feeling of well-being and accomplishment (n=1); (5) a measure of insulin resistance (n=1); and (6) level of social participation (n=1).

Conclusions

Health outcomes which cover both the physical and psychological aspects of health were determined to be critical or important by experts. The important and critical health outcomes identified could help guideline-makers in the decision of the evidence profile to include in the development of future clinical practice guidelines in individuals with CP. These guidelines can help clinicians create management plans to increase PA in this population.

Introduction

Background

Cerebral palsy (CP) is a group of permanent disorders of the development of movement and posture that appear in early childhood [1]. A population-based estimate of the prevalence of the motor disorder is 2-3 per 1000 live births, making it the most common childhood physical disability [2–4]. Trends indicate that individuals with CP have a long lifespan; more than 80% have a life expectancy beyond 58 years and longer [5]. However, aging with CP is associated with heightened noncommunicable disease risk [6], including cardiovascular disease [7], and mental health issues [8,9]. It is necessary to identify and address unique comorbidity or multimorbidity, including both physical and mental health problems, within this population.

Both children and adults with CP engage in less physical activity (PA) than the typically developing population, which could be attributed to difficulties of movement associated with the motor disorder [10–13]. In these individuals, lower PA levels are strongly associated with physical functioning, such as loss of mobility or deterioration in walking abilities [14]. In the typically developing population, low PA leads to increased risk of negative health conditions, such as cardiovascular disease, and other secondary health concerns, including early function loss [15,16]. On the other hand, the positive health effects of PA have been well established across all age groups in the typically developing population. These benefits include improved body composition, through weight control and reduced abdominal adiposity, and reduced blood pressure [17]. While PA can have a myriad of benefits, such as physical and emotional well-being, for the typically developing population, these are particular concerns in individuals with physical disabilities due to their restricted social participation [18]. Therefore, increasing PA is crucial towards improving health outcomes in individuals with CP.

Clinicians and researchers can help identify the health outcomes that are relevant to individuals with CP by integrating their clinical expertise and experience working directly with these patients. Furthermore, researchers and clinicians play a key role in PA intervention design and management in individuals with CP. While an earlier study has explored the importance of core outcomes [19], consisting of multimorbidity risk factors, in individuals with CP based on expert discussions, the sample size of experts was only eight, potentially enhancing the risk of selection bias. Furthermore, the outcomes were identified according to their importance in assessing multimorbidity risk rather than their relationship to PA and did not include psychological aspects of health.

To create PA recommendations for individuals with CP, one must establish important and critical health outcomes related to PA. Understanding these outcomes will contribute towards establishing evidence profiles for the development of future clinical practice guidelines in PA management for individuals with CP based on the standard of guideline development-the Grading of Recommendations Assessment, Development and Evaluation (GRADE) approach [20]. To inform the first step in the GRADE approach for clinical practice guideline development, we initiated a survey with experts to identify important and critical health outcomes for PA in individuals with CP using the rating system provided by the GRADE approach. The GRADE approach is the current established standard for developing CP population-specific clinical practice guidelines [20]. Clinical practice guidelines ultimately inform clinicians in designing effective PA programs and resources for people with CP.

Objectives of study

The primary purpose of the survey was to rate the importance of pre-determined health outcomes, related to PA, based on expert opinions for individuals with CP, across all ages. The study will contribute towards the development of a clinical practice guideline for managing PA in individuals with CP following the GRADE methodology [20].

Research Question

What are important and critical health outcomes related to PA for all individuals with CP based on the opinions of clinicians and researchers?

Methods

Survey Development: GRADE Approach

We followed the "Checklist for Reporting Results of Internet E-Surveys" [21]. The survey options followed the GRADE rating scale to determine the importance of the health outcomes in development of clinical practice guidelines for individuals with CP [22].

Rationale for Item Generation of Included Health Outcomes

The criteria used to select studies reporting on the nine pre-determined health outcomes was that it included expert input on the outcomes. In addition, we also selected outcomes which are among the most prevalent in the typically developing population and in addition, have a heightened prevalence, compared to that of the typically developing population, in individuals with CP; it was preferable that the prevalence of these outcomes in individuals with CP was determined through large cohort studies. We selected both physical and psychological outcomes to cover the spectrum of health, as done in the development of other PA guidelines [23-25].

Physical Health Outcomes (7 Outcomes)

The survey included seven core outcomes reported in a study aimed to establish a core outcome set for assessing multimorbidity risk in adolescents (14-18 years of age) and adults (> 18 years of age) with CP [19]. Outcomes from the study were selected because in the study, consensus on these seven health outcomes was achieved through an in-person meeting between a total of eight clinicians and/or research experts. A clinical expert must have worked with adolescents and/or adults with CP for minimum 5 years while a research expert must have published at least one article on an identified outcome of multimorbidity risk in the study's population. The core outcomes selected for their potential relevance, as related to PA, for both adolescents and adults with CP were: (1) cardiorespiratory endurance; (2) body size (i.e., height and weight) ; (3) body composition (i.e., waist circumference); (4) sleep; (5) nutrition; (6) blood lipids and glucose; and (7) blood pressure. The selection process for these outcomes were outlined in the protocol [19]. The protocol described the selection of these core outcomes for a study [26] which aimed to identify outcome measurement instruments for each of these seven core outcomes.

Mental Health Outcomes (2 Outcomes)

To cover the psychological aspects of health, consultation was done with a clinical expert and researcher (JWG) and a researcher (PM) to identify health outcomes of high prevalence through cohort studies that cover the psychological aspect of health, in individuals with CP. Two large cohort studies were identified. Anxiety and depression were chosen as health outcomes because they are the two most common mental illnesses in the general population [27]. Depression [28,29] and anxiety [30] have a two-to-three-fold greater likelihood of occurring in individuals with a long-term condition or disability.

A large cohort study, conducted in the United Kingdom, consisting of 1705 adults with CP and a matched group of 5115 adults without CP, found that adults with CP are at an increased risk of developing anxiety and depression [31]. The risk for depression was 28 percent higher and the risk of anxiety was 40 percent higher among adults with CP who have intellectual difficulties compared to the typically developing population. For those who had CP but did not have an intellectual disability, the probability of developing depression and anxiety increased even more; with individuals having a 44 percent higher risk of depression and 55 percent higher risk of anxiety than their typically developing peers.

In another cohort study, conducted in the United States, consisting of 111 children with CP and 29 909 children without CP, anxiety was found to have the highest prevalence out of the mental health disorders studied. Children with CP had higher prevalence and unadjusted odds of both depression and anxiety compared to controls [8].

In total, nine health outcomes were chosen to be included in the pre-determined set of health outcomes for rating in the survey. These consisted of seven physical health and two mental health outcomes. The final nine health outcomes were selected by JX based on a review of the literature, and then reviewed by two experts for thoroughness (JWG, PM).

Question Stems

The question posed to experts was: "Which health outcomes are most important for individuals with CP, based on your expertise and experience?", intended to capture the expert's opinions on the importance of the nine health outcomes. The survey question was posed within the context of a workshop on PA in individuals with CP to give context to the question posed. The abstract for the presentation is shown in Additional Figure 1. The content of the workshop gave an overview of interventions to increase PA participation in individuals with CP and did not pertain to any of the health outcomes in the survey. The content of the workshop also included the proposal for the development of a Care Pathway on promoting PA and reducing SB in individuals with CP. The survey (Additional Figure 2) asked the experts to rate the 9 health outcomes with the GRADE rating scale. A free-text box titled "Other (please specify)" offered respondents an opportunity to suggest additional health outcomes.

Responses

The GRADE approach calls for selecting critical and important health outcomes through a transparent method for documenting and considering them. The approach specifies deciding the importance of health outcomes to decision-making in guideline development to be done by numerically rating outcomes on a 9-point Likert type integer scale with ratings from 1 to 9 (7 to 9 – critical; 4 to 6 – important; 1 to 3 – of limited importance) [20]. A visual of the scale is presented in Figure 1. This is not a rank order of importance and more than one outcome can have the same importance rating (e.g., two health outcomes can each have a score of 7). Both critical and important outcomes are included in the evidence profile for guideline development. Decisions on the overall quality of evidence for a guideline recommendation may depend on which outcomes are designated as "critical" and which are "important" [32]. The GRADE rating scale is not specific to any health outcome and can include any outcomes that may be important for guideline development. Each respondent scored the nine health outcomes on the scale. The GRADE rating scale and associated meaning for each group of rating was projected on a large screen (an image similar to Figure 1) for experts during the voting period.

Formatting

The survey was conducted through SurveyMonkey, an online survey tool chosen to deploy the survey and obtain analytics immediately after voting for discussions during the workshop. To avoid duplicate responses, SurveyMonkey settings were configured so that only one response was allowed per browser or email address. All participants completed the survey during one designated time at the workshop. As per conference guidelines, the workshop allowed 40 minutes for giving a presentation, followed by 20 minutes for interactive audience engagement, so the survey and expert discussion of real-time results were limited by this time frame. Survey was restricted to those who attended by only including surveys completed during this 20-minute voting period between 10:40am-11am Pacific Standard Time Thursday, September 19, 2019.

Setting

An open and cross-sectional survey was conducted during a workshop at the combined 73rd American Academy of CP and Developmental Medicine (AACPDM) Annual and 2nd *International Alliance of Academies of Childhood Disability* (IAACD) Triennial Meeting in September 2019. The workshop was titled “Promoting Physical Activity in Individuals with Cerebral Palsy”.

Participants

A convenience sample was composed of 55 respondents out of the 75 eligible respondents (73% participants who registered for the workshop). The demographic information was obtained from the list of registered attendees provided to us as the presenters of the workshop. The list included the participant's country and specialty description.

The survey results of 55 respondents were anonymous, and no demographic information was asked during the survey portion. Therefore, the demographic information presented in the results reflect the profile of 75 registrants rather than those who completed the survey.

Administration

An abstract summarizing the content of the workshop was submitted to the AACPDM. The workshop was advertised in the printed "Final Program" for the conference, which was also available on the AACPDM website, and in the conference mobile app. We invited all registered attendees of the workshop to participate and excluded those who did not attend. Attendees were provided with a QR code during the workshop, projected onto a screen, to access the online survey on SurveyMonkey. The survey was also sent electronically to them in an email the day prior with instructions to wait until the workshop to complete voting in case they could not access the QR code during the workshop. Participants were not able to review nor change their answers once submitted. However, they could scroll through all their responses, at once, before submitting, since rating scales for each of the nine health outcomes were presented on one webpage. Survey completion, both for the survey and for the voting of each item, was voluntary, and responses were anonymous.

Analysis

The descriptive statistics were summarized using numbers and percentages to describe the occupation, education and country of participants. The median and interquartile range (IQR) for each of the nominal data on the rating for each health outcomes were reported. Statistical analysis was performed using the STATA/IC software (STATA 13.0 for Mac, College Station, TX, USA).

Since the GRADE ratings are ordinal, non-parametric statistical tests were conducted. Since the group of experts are from an independent random sample, and the ratings for 9 categorical groups of health outcomes were compared, the Kruskal-Wallis test was used to determine if any pairwise comparisons between the ratings of the nine independent health outcomes were statistically significant. The purpose of the Kruskal Wallis test was to determine if there were significant differences between the health outcomes with median ratings of "critical" and health outcomes with median ratings of "important". A resulting p-value of $p < 0.05$ for the Kruskal-Wallis test indicates there is a significant difference between at least 2 health outcomes. In this case, pair-wise comparisons were performed to determine which outcome(s) were significantly different using the post-hoc Dunn's test, which consists of performing Wilcoxon Rank-Sum (Mann-Whitney U) tests. The Dunn's test with programmed Bonferroni corrections was used to determine where significant differences existed between pairwise comparisons of health outcomes (i.e., $p<0.05$). The Dunn's test can simultaneously conduct multiple Wilcoxon Rank-Sum (Mann-Whitney U) tests with Bonferroni adjustments (i.e., $\alpha=0.05$, p-value calculated is multiplied by 9). As the future step from this study is to develop an exhaustive list of health outcomes with patient input, assessing if there is a significant difference between two outcomes may help narrow down the selection of outcomes from within the nine pre-determined health outcomes, if there was contention for the inclusion of any two outcomes from this study. The analysis would help enhance the usability of data from this study for such a future initiative.

Ethical Considerations

According to Article 2.4 of the "Ethical Conduct for Research Involving Humans" , research ethics board review is not required for research that relies exclusively on secondary use of anonymous information as long as the process of data linkage or recording or dissemination of results does not generate identifiable information [33]. Secondary use, in research, refers to the use of information originally collected for a purpose other than the current research purpose. The original purpose of the data from this study was to inform the development of an AACPDM Care Pathway on PA promotion rather than be analyzed separately in a survey. Anonymous information refers to information that never had identifiers associated with it and the risk of identification of individuals is very low. The information proposed for our research is not identifiable because we cannot link the data to respondents since we do not know the demographics for attendants of the workshop who voted in the survey, but rather only those who registered for the workshop. Based on the application of Article 3.12 [33], consent may be demonstrated solely by the actions of the participant (e.g., through the return of a completed questionnaire).

Results

Description of the Sample of Registered Experts

All questionnaires were filled out during the date and time period of the workshop. A summary of the specialty and country of study or practice is numerically shown in Table 1. The experts were classified, foremost, by their clinical background. Of the 75 individuals who registered for the workshop, physiotherapists made up the largest proportion of experts (42.7%). Physicians made up the second largest specialty (22.7%), and individuals holding solely Master's or PhD degrees made up the third largest specialty (20%). Occupational therapists made up the fourth largest specialty (9.3%), followed by nurse-practitioners (2.7%). Finally, there was one person holding solely a Bachelor's degree and one person in administration (1.3% each).

As shown by Table 1, the majority of the 75 individuals who registered for the workshop came from North America (60.0%), which aligns with the conference's geographic location being in the USA. The second largest continent represented was Asia (17.3%), followed by Europe (13.4%), then Oceania, which consisted only of Australian participants (8.0%), and finally, South America (1.3%), which consisted only of a participant from Brazil. Africa was not represented among the list of registered participants.

Convenience Sample of Participants

As not all the experts who registered for the workshop attended the event, the descriptive statistics presented for the voting population only gives an estimate of the specialty and demographics of the voting sample. Reasons for non-attendance were unknown. The flow diagram of survey respondents is shown in Figure 2. All participants (n=55) attending the workshop completed the survey during the allotted time window. All respondents answered the survey questions in its entirety, so there was no missing data.

Descriptive Statistics: Ratings for each health outcomes

Table 2 shows the median and IQR for each health outcome. Additional Table 3 provides the frequency and proportion of responses for the GRADE ratings for each of the nine pre-determined health outcomes from the survey.

The medians for all outcomes lie between 4-7, which indicates that all outcomes are at least important, and at most, critical. Across the nine health outcomes, the highest rated outcomes, which were deemed "critical" based on the GRADE rating scale, were cardiorespiratory endurance, sleep, nutrition, depression and anxiety, all with the highest median rating of 7 (IQR for sleep, depression and nutrition: 5-8; IQR for cardiorespiratory endurance: 6-8). The median was second highest for blood pressure 6 (IQR: 3-7) and lowest for body size, body composition and "blood lipids and glucose" 5 (IQR for BS: 4-7; IQR for BC and "blood lipids and glucose": 3-7); these health outcomes were deemed "important" based on the GRADE rating scale.

The interquartile ranges show the least amount of variability for cardiorespiratory endurance, and anxiety (IQR= 2); second for body size, sleep, nutrition, blood pressure, and depression (IQR=3); the most variability for body composition, and blood lipids and glucose (IQR=4).

Relative Importance of Health Outcomes

Table 3 summarizes pairwise comparisons for the health outcomes based on the median of their GRADE ratings. The Kruskal-Wallis test showed significant differences between the medians of the nine health outcomes. The Dunn's test, with Bonferroni corrections, showed statistically significant differences between pairwise comparisons of the medians for the health outcomes. The z-test of pairwise comparisons performed on the nine health outcomes and the significance values are shown in Additional Table 4.

Additional health outcomes

Free-form responses were provided by six respondents. These comments indicated additional health outcomes. Some respondents suggested more than one health outcome. These outcomes are: (1) bone density (n=1); (2) quality of life (n=1); (3) emotional well-being (n=1); (4) global feeling of well-being and accomplishment (n=1); (5) a measure of insulin resistance (n=1); and

(6) level of social participation (n=1). One respondent highlighted the importance of nutrition, which was already included in the survey.

Discussion

We conducted an international, electronic survey with primarily clinicians and researchers with an interest in PA promotion in individuals with CP, with the aim to determine the critical and important health outcomes in individuals with CP across all ages, based on the experts' opinions. We obtained data from 55 experts by conducting a survey, in real time, at a conference devoted to topics in neurodevelopmental disabilities. Using the rating scale from the GRADE approach [20], we identified general agreement that nine health outcomes were important or critical for decision-making for future clinical practice guideline development for individuals with CP. The traditional outcomes of health are physical factors such as blood pressure and cardiorespiratory fitness [34]. While more than half of traditional outcomes, consisting of physical health, were rated as being important (i.e., body size [i.e., height and weight] ; body composition [i.e., waist circumference]; blood lipids and glucose; blood pressure) and the other half rated as being critical (i.e., sleep; nutrition, cardiorespiratory endurance), both the psychological health outcomes were rated as being critical (i.e., depression; anxiety). These findings indicate that experts considered the inclusion of psychological health outcomes to be critical in the evidence profile for decision-making in PA for individuals with CP.

The pairwise comparison of the nine health outcomes suggests that the ratings for the health outcomes of cardiorespiratory endurance, sleep, nutrition, depression and anxiety, which were all rated as critical, were significantly higher than for body size, body composition, blood pressure and blood lipids and glucose, which were all rated as important. The exception was between the pairwise comparisons in ratings for sleep and for nutrition, rated as critical, with blood pressure, rated as important. The ratings were classified as "important" or "critical" based on the importance attached to each group of numerical ratings according to the GRADE rating scale. These results suggest that, in general, critical outcomes were rated as being significantly higher than important outcomes and could help determine the overall quality of evidence in future guideline recommendations for PA management in individuals with CP.

Clinical endpoints are outcomes of direct patient relevance, measuring the primary interest for patients in and of itself. On the other hand, surrogate outcomes are used as substitutes for a direct measure of how a patient feels, functions, or survives [35]. All clinical endpoints are rated as critical while surrogate outcomes, which are blood pressure, blood lipids and glucose, body composition and body size, are rated as important. Therefore, while these surrogate outcomes are suggested to be on the causal pathway to endpoints of direct relevance to patients, they themselves are not of direct relevance to patients. They are not as important to measure, based on the opinions of clinical and research experts. However, these surrogate endpoints do have the advantage of being easier to measure. Ideally, surrogate outcomes could act as proxies for the clinical endpoints when all mechanisms of action to the clinical endpoint are mediated by the surrogate outcome [36].

The six additional health outcomes identified in the free-form responses all overlapped with the critical and important health outcomes used in the development of the 24-Hour Movement Guidelines, namely bone density, quality of life, emotional well-being, global feeling of well-being and accomplishment, measure of insulin resistance (a cardiometabolic biomarker); and level of social participation [23–25]. These results indicate that individuals with CP may have overlapping needs with their typically developing peers. This suggests that future studies on important health outcomes, relevant to PA, for individuals with CP should also consider health outcomes important in the typically developing population. A study which reported on the research priorities in the field of CP, discussed with caregivers and individuals with CP, clinicians, researchers and CP community advocates, found that participation and quality of life were important outcomes of future intervention studies in individuals with CP [38]. However, these outcomes were not decided for their relevance to PA and agreement was reached not specifically for the health outcomes but rather for the research priority with these outcomes as emerging themes. Nevertheless, the study results support the freeform responses in our study, suggesting that quality of life and participation could potentially be important or critical health outcomes in PA for people with CP.

The results from this study should be interpreted with caution because the nine pre-determined outcomes were not an exhaustive list of all health outcomes that relevant to PA in this population. Rather, this study provides a list of health outcomes found to have importance through literature, which potentially among others, are important for PA in individuals with CP.

Implications/ Future Steps

To mitigate the negative health consequences of inactivity, a systematic effort to promote PA in the CP population is needed, but it is difficult to design effective PA programs and resources for people with CP in the absence of population-specific clinical practice guidelines. This study aimed to inform one of the first steps in the GRADE approach to guideline development, which is the preliminary classification of outcomes, by an expert panel, as important, critical or of limited importance before reviewing evidence. The next step is to obtain patient input for their ratings and thoughts on important and critical health outcomes relevant to PA. In this way, the final outcomes informing guideline development can be "patient-important" [20].

As with quantitative surveys, there is little contextual information surrounding the health outcomes [39]. For example, different clinician specialties and researchers hold unique perspectives on the rationale for their ratings of the health outcomes, information which could affect the development or implementation of clinical practice guidelines for the population. A future step can be to conduct a focus group for clinicians, researchers and individuals with CP and their families to discuss their rationale for voting on these health outcomes. This may also generate ideas for other health outcomes. Focus groups may be the appropriate method to discover these perspectives, as it is exploratory in nature and provides qualitative descriptions of the experts' rationales for health outcome ratings and choices [40]. The experience from prior focus groups demonstrates the importance of 'warm-up' time, in which the moderator provides participants with some background information about the topic [41]. As no prior studies were conducted on important and critical health outcomes, relevant to PA, for individuals with CP, the outcomes, their rationale for inclusion in this study, and their relative importance, could serve as a starting point in focus group discussions.

Limitations

We did not conduct rigorous testing of the survey to evaluate its feasibility, and ease of administration. To ensure a comprehensive representation of potentially critical and important health outcomes in the survey, that is decided through a more systematic approach, we could have employed "sampling to redundancy", a technique in which item generation occurs until no new items emerge [42]. This could have been accomplished by in-depth interviews or focus-group sessions with a larger expert panel to decide the initial health outcomes for voting. This method may help to mitigate selection bias from only two experts reviewing the thoroughness of included studies from a non-systematic review of the literature in choosing the nine pre-determined health outcomes. However, the purpose of the study is not to rate the importance of an exhaustive list of health outcomes but rather a list of potentially critical or important health outcomes to PA for individuals with CP. Furthermore, there was no pre-testing of the survey question to ensure expert respondents would interpret the question consistently and in the intended manner [42].

As the survey was anonymous, the exact demographic information of experts who voted was unknown, such as whether the experts worked in pediatric or adult care. Furthermore, we did not connect demographic information to survey results, so we could not discern subgroup variation in ratings (e.g., regional or specialty differences in ratings).

Conference samples are prone to selection bias due to the convenience sample design [43]. However, this bias may have been offset in part by the strength of our study, which was a large sample size from different specialties, workplace settings and countries [44]. The large sample size also maximized the generalizability of the experts' responses from the sample to the population of experts working with individuals with CP.

It is possible that all health outcomes have been deemed important due to acquiescence bias, which is the general tendency of a person to provide affirmative answers to items of a questionnaire, regardless of the content of the items [45]. In this study, this would mean a tendency for respondents to rate health outcomes as "critical" or "important" rather than "of limited importance".

Conclusion

Our survey aimed to provide expert ratings to identify the importance of nine pre-determined health outcomes relevant to PA for individuals with CP. We found that for clinicians and researchers working with individuals with CP, health outcomes need to cover both the psychological and physical aspects of health and have direct relevance to patients. Given the need for increased PA in individuals with CP, it is imperative to decide on these health outcomes for clinical practice guideline development. There should be a collective effort in PA research to design tools with input from various stakeholders, as done in this study, who work directly with the population of interest.

References

1. Rosenbaum P, Paneth N, Leviton A, Goldstein M, Bax M, Damiano D, et al. A report: the definition and classification of cerebral palsy April 2006. Dev Med Child Neurol Suppl. 2007;109:8–14.

2. Accardo PJ, Capute AJ, editors. Capute & Accardo's neurodevelopmental disabilities in infancy and childhood. 3rd ed. Baltimore: Paul H. Brookes Pub; 2008. 2 p.

3. Surman G, Bonellie S, Chalmers J, Colver A, Dolk H, Hemming K, et al. UKCP: a collaborative network of cerebral palsy registers in the United Kingdom. J Public Health. 2006;28(2):148–56.

4. Cans C. Surveillance of cerebral palsy in Europe: a collaboration of cerebral palsy surveys and registers. Dev Med Child Neurol. 2007;42(12):816–24.

5. Blair E, Langdon K, McIntyre S, Lawrence D, Watson L. Survival and mortality in cerebral palsy: observations to the sixth decade from a data linkage study of a total population register and National Death Index. BMC Neurol. 2019;19(1):111.

6. Ryan JM, Allen E, Gormley J, Hurvitz EA, Peterson MD. The risk, burden, and management of non-communicable diseases in cerebral palsy: a scoping review. Dev Med Child Neurol. 2018;60(8):753–64.

7. Mcphee PG, Claridge EA, Noorduyn SG, Gorter JW. Cardiovascular disease and related risk factors in adults with cerebral palsy: a systematic review. Dev Med Child Neurol. 2018; 61(8):915-923.

8. Whitney DG, Warschausky SA, Peterson MD. Mental health disorders and physical risk factors in children with cerebral palsy: a cross-sectional study. Dev Med Child Neurol. 2019;61(5):579–85.

9. Whitney DG, Warschausky SA, Ng S, Hurvitz EA, Kamdar NS, Peterson MD. Prevalence of Mental Health Disorders Among Adults With Cerebral Palsy: A Cross-sectional Analysis. Ann Intern Med. 2019;171(5):328.

10. Carlon SL, Taylor NF, Dodd KJ, Shields N. Differences in habitual physical activity levels of young people with cerebral palsy and their typically developing peers: a systematic review. Disabil Rehabil. 2013;35(8):647–55.

11. Nooijen CF, Slaman J, Stam HJ, Roebroeck ME, Berg-Emons R, Learn2Move Research Group. Inactive and sedentary lifestyles amongst ambulatory adolescents and young adults with cerebral palsy. J NeuroEngineering Rehabil. 2014;11(1):49.

12. Balemans A.C., Fragala-Pinkham M.A., Lennon N., Thorpe D., Boyd R.N., O'Neil M.E., et al. Systematic review of the clinimetric properties of laboratory- and field-based aerobic and anaerobic fitness measures in children with cerebral palsy. Arch Phys Med Rehabil. 2013;94(2):287–301.

13. Nieuwenhuijsen C, van der Slot WMA, Dallmeijer AJ, Janssens PJ, Stam HJ, Roebroeck ME, et al. Physical fitness, everyday physical activity, and fatigue in ambulatory adults with bilateral spastic cerebral palsy: Fitness, activity, fatigue in cerebral palsy. Scand J Med Sci Sports. 2011;21(4):535–42.

14. Opheim A, Jahnsen R, Olsson E, Stanghelle JK. Walking function, pain, and fatigue in adults with cerebral palsy: a 7-year follow-up study. Dev Med Child Neurol. 2009;51(5):381–8.

15. Shuval K, Finley CE, Barlow CE, Gabriel KP, Leonard D, Kohl HW. Sedentary Behavior, Cardiorespiratory Fitness, Physical Activity, and Cardiometabolic Risk in Men: The Cooper Center Longitudinal Study. Mayo Clin Proc. 2014;89(8):1052–62.

16. Myers J. Exercise and Cardiovascular Health. Circulation. 2003;107(1): e2–e5.

17. Warburton DER. Health benefits of physical activity: the evidence. Can Med Assoc J. 2006;174(6):801–9.

18. Tough H, Siegrist J, Fekete C. Social relationships, mental health and wellbeing in physical disability: a systematic review. BMC Public Health. 2017;17(1):414.

19. McPhee PG, Benner JL, Balemans ACJ, Verschuren O, van den Berg-Emons RJG, Hurvitz EA, et al. Multimorbidity risk assessment in adolescents and adults with cerebral palsy: a protocol for establishing a core outcome set for clinical research and practice. Trials. 2019;20(1):176.

20. Schünemann H. The GRADE handbook. Cochrane Collaboration; 2013.

21. Eysenbach G. Improving the Quality of Web Surveys: The Checklist for Reporting Results of Internet E-Surveys (CHERRIES). J Med Internet Res. 2004;6(3):e34.

22. Guyatt GH, Oxman AD, Kunz R, Atkins D, Brozek J, Vist G, et al. GRADE guidelines: 2. Framing the question and deciding on important outcomes. J Clin Epidemiol. 2011;64(4):395–400.

23. Poitras VJ, Gray CE, Borghese MM, Carson V, Chaput J-P, Janssen I, et al. Systematic review of the relationships between objectively measured physical activity and health indicators in school-aged children and youth. Appl Physiol Nutr Metab. 2016;41(6 (Suppl. 3)):S197–239.

24. Saunders TJ, Gray CE, Poitras VJ, Chaput J-P, Janssen I, Katzmarzyk PT, et al. Combinations of physical activity, sedentary behaviour and sleep: relationships with health indicators in school-aged children and youth. Appl Physiol Nutr Metab. 2016;41(6 (Suppl. 3)):S283–93.

25. Carson V, Lee E-Y, Hewitt L, Jennings C, Hunter S, Kuzik N, et al. Systematic review of the relationships between physical activity and health indicators in the early years (0-4 years). BMC Public Health. 2017;17(S5):854.

26. Benner JL, McPhee PG, Gorter JW, Hurvitz EA, Peterson MD, Obeid J, et al. Focus on Risk Factors for Cardiometabolic Disease in Cerebral Palsy: Toward a Core Set of Outcome Measurement Instruments. Arch Phys Med Rehabil. 2019;S0003999319303387.

27. Demyttenaere. Prevalence, Severity, and Unmet Need for Treatment of Mental Disorders in the World Health Organization World Mental Health Surveys. JAMA. 2004;291(21):2581.

28. Katon WJ. Clinical and health services relationships between major depression, depressive symptoms, and general medical illness. Biol Psychiatry. 2003;54(3):216–26.

29. Turner RJ, Noh S. Physical Disability and Depression: A Longitudinal Analysis. J Health Soc Behav. 1988;29(1):23.

30. Roy-Byrne PP, Davidson KW, Kessler RC, Asmundson GJG, Goodwin RD, Kubzansky L, et al. Anxiety disorders and comorbid medical illness. Gen Hosp Psychiatry. 2008;30(3):208–25.

31. Smith KJ, Peterson MD, O'Connell NE, Victor C, Liverani S, Anokye N, et al. Risk of Depression and Anxiety in Adults With Cerebral Palsy. JAMA Neurol. 2019;76(3):294.

32. Balshem H, Helfand M, Schünemann HJ, Oxman AD, Kunz R, Brozek J, et al. GRADE guidelines: 3. Rating the quality of evidence. J Clin Epidemiol. 2011;64(4):401–6.

33. Canadian Institutes of Health Research. Tri-Council policy statement: ethical conduct for research involving humans. [Internet]. 2018 [cited 2020 Jul 5]. Available from: http://publications.gc.ca/collections/collection_2019/irsc-cihr/RR4-2-2019-eng.pdf

34. Svalastog AL, Donev D, Jahren Kristoffersen N, Gajović S. Concepts and definitions of health and health-related values in the knowledge landscapes of the digital society. Croat Med J. 2017;58(6):431–5.

35. Biomarkers and surrogate endpoints: Preferred definitions and conceptual framework. Clin Pharmacol Ther. 2001;69(3):89–95.

36. Gupta K, Gupta J, Singh S. Surrogate Endpoints: How Reliable Are They? 2010;6.

37. Chaput J-P, Gray CE, Poitras VJ, Carson V, Gruber R, Birken CS, et al. Systematic review of the relationships between sleep duration and health indicators in the early years (0–4 years). BMC Public Health. 2017;17(S5):855.

38. Gross PH, Bailes AF, Horn SD, Hurvitz EA, Kean J, Shusterman M, et al. Setting a patient-centered research agenda for cerebral palsy: a participatory action research initiative. Dev Med Child Neurol. 2018;60(12):1278–84.

39. Choy LT. The Strengths and Weaknesses of Research Methodology: Comparison and Complimentary between Qualitative and Quantitative Approaches. IOSR J Humanit Soc Sci. 2014;19(4):99–104.

40. Basch CE. Focus Group Interview: An Underutilized Research Technique for Improving Theory and Practice in Health Education. Health Educ Q. 1987;14(4):411–48.

41. Breen RL. A Practical Guide to Focus-Group Research. Journal of Geography in Higher Education. 2006;30(3):463–75.

42. Burns KEA, Duffett M, Kho ME, Meade MO, Adhikari NKJ, Sinuff T, et al. A guide for the design and conduct of self-administered surveys of clinicians. Can Med Assoc J. 2008;179(3):245–52.

43. Lavrakas P. Encyclopedia of Survey Research Methods [Internet]. 2455 Teller Road, Thousand Oaks California 91320 United States of America: Sage Publications, Inc.; 2008 [cited 2020 Jun 28]. Available from: http://methods.sagepub.com/reference/encyclopedia-of-survey-research-methods

44. McColl E, Jacoby A, Thomas L, Soutter J, Bamford C, Steen N, et al. Design and use of questionnaires: a review of best practice applicable to surveys of health service staff and patients. Health Technol Assess [Internet]. 2001 [cited 2020 Jun 14];5(31). Available from: https://www.journalslibrary.nihr.ac.uk/hta/hta5310/

45. Hinz A, Michalski D, Schwarz R, Herzberg PY. The acquiescence effect in responding to a questionnaire. Psycho-Soc Med. 2007;4:Doc07.

Figure 1. GRADE Rating Scale

1	2	3	4	5	6	7	8	9
Least important								Most important
Least important for decision-making (not included in evidence profile)			Important but not critical for decision-making (included in evidence profile)			Critical for decision-making (included in evidence profile)		

Figure 2. Flow diagram of survey respondents

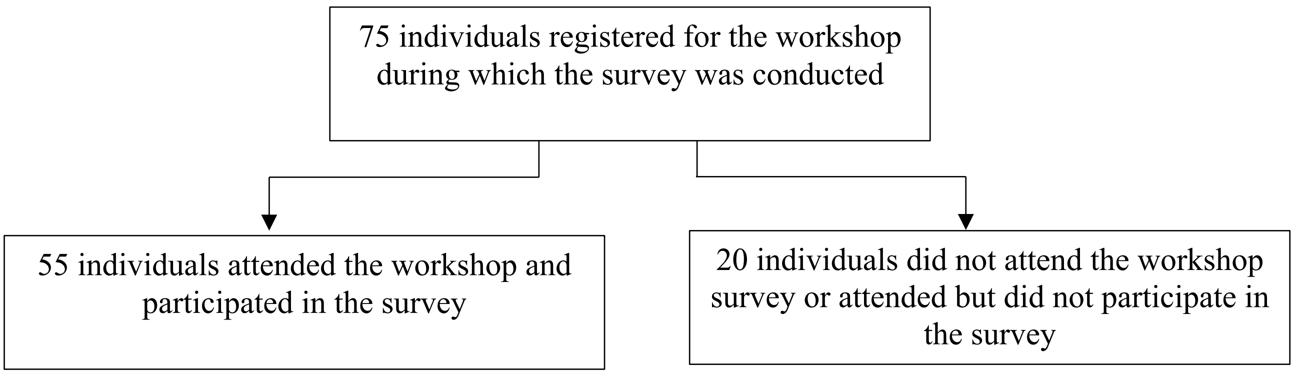


Table 1. Demographics of registered experts (n=75)

Characteristic			**Value**
Country and Continent, n respondents (% of total)	North America	USA	35 (45.5)
		Canada	10 (13.0)
		Total	45 (60.0)
	Asia	Korea	8 (10.4)
		China	2 (2.6)
		Japan	2 (2.6)
		Israel	1 (1.3)
		Total	13 (17.3)
	Europe	Netherlands	5 (6.5)
		Sweden	2 (2.6)
		United Kingdom	2 (2.6)
		Denmark	1 (1.3)
		Total	10 (13.3)
	Oceania	Australia	6 (8.0)
	South America	Brazil	1 (1.3)
Occupation/Education, n respondents (% of total)	Clinical Background[a]	PT or PT/PhD or PT/Master's	32 (42.7)
		MD or DO or MD/PhD	17 (22.7)
		OT or OT/PhD	7 (9.3)
		Nurse-Practitioner or Nurse-Practitioner/PhD	2 (2.7)
	Research/Educational Background for Respondents with no clinical background[b]	PhD or Master's	15 (20)
		Bachelor's	1 (1.3)
		Administration	1 (1.3)

Legend: [a]The registered participants were classified foremost by their clinical background, if they had one, regardless of their graduate degree level. [b] Those who did not have a clinical background were classified by their graduate degree level.

Table 2. Median, IQR and rating of importance for nine health outcomes (n=55 for each outcome) based on the GRADE 9-point Likert-type scale (1-9)

Health Outcomes	Rating	Median	IQR (1st, 3rd)
Physical Outcomes			
Cardiorespiratory Endurance	Critical	7	2 (6, 8)
Sleep	Critical	7	3 (5, 8)
Nutrition	Critical	7	3 (5, 8)
Blood pressure	Important	6	3 (3, 7)
Body size	Important	5	3 (4, 7)
Body composition	Important	5	4 (3, 7)
Blood lipids and glucose	Important	5	4 (3, 7)
Psychological Outcomes			
Anxiety	Critical	7	2 (6, 8)
Depression	Critical	7	3 (5, 8)

Table 3. Results for Dunn's test for statistically significant pairwise comparisons ($\alpha<0.05$)[a] (n=55)

Pairwise comparison of health outcomes (Health outcome 1- Health outcome 2)	Z-Score	P-value
Cardiorespiratory endurance **-** Body size	4.65	0.0001
Cardiorespiratory endurance **-** Body composition	4.18	0.0005
Cardiorespiratory endurance **-** Blood Pressure	3.51	0.0080
Cardiorespiratory endurance **-** Blood lipids and glucose	4.21	0.0005
Body size **-** Sleep	-3.89	0.0018
Body size **-** Nutrition	-3.72	0.0036
Body size **-** Depression	-4.32	0.0003
Body size **-** Anxiety	-4.27	0.0004
Body composition **-** Sleep	-3.42	0.0114
Body composition **-** Nutrition	-3.25	0.0210
Body composition **-** Depression	-3.85	0.0021
Body composition **-** Anxiety	-3.79	0.0027
Sleep **-** Blood lipids and glucose	3.45	0.0100
Nutrition **-** Blood lipids and glucose	3.28	0.0185
Blood pressure **-** Depression	-3.18	0.0261
Blood pressure **-** Anxiety	-3.13	0.0315
Blood lipids and glucose **-** Depression	-3.88	0.0019
Blood lipids and glucose **-** Anxiety	-3.83	0.0023

[a] Only the results for statistically significant pairwise comparisons were included in the table

Additional Figure 1. Submitted abstract for the workshop presented at the AACPDM on promoting physical activity in individuals with cerebral palsy

Submission Title:
Promoting physical activity and reducing sedentary behaviour in individuals with cerebral palsy over the lifecourse through the development of an AACPDM Care Pathway

Presenter(s)

---

1. Jan Willem Gorter, MD PhD (Role: Presenter)
2. Julia (Shi-Peng) Xiong, BHSc (Role: Presenter)!
3. Patrick McPhee, PhD (Role: Presenter)
4. Sarah E. Reedman, PhD, BPhty (Hons) (Role: Presenter)

Abstract Body

Learning Objectives

1. **Learn current PA promotion strategies and understand the importance of targeting PA participation and reducing sedentary behaviour**
2. **Understand the AACPDM Care Pathway development process**
3. **Learn and discuss assessment criteria of baseline PA levels and decision-points for making PA management plans, based on empirical evidence, end-user experiences, and opinions from an international, multidisciplinary expert team**
4. **Relate the Care Pathway development process to the attendees' own expertise**

Purpose

To learn about the process and preliminary results of the Care Pathway on guiding clinical decision-making on "promoting physical activity (PA) and reducing sedentary behaviour in individuals with cerebral palsy (CP) over the lifecourse", shape the development of the Pathway through alignment with current practice and research, and discuss its implementa!

tion.

Target Audience

clinicians, researchers, individuals with cerebral palsy and their families interested in physical activity promotion across the lifespan (children and adults)

Course Summary

Many interventions aim to promote PA and reduce sedentary behaviour in persons with CP by targetting physical capacity or skill development. However, recent systematic reviews show that this practice yields only a small increase in PA, with, at best, modest sustained effects at follow-up. Studies have shown promise by focusing on interventions at the activity, participation, personal and environmental levels.

A new AACPDM Care Pathway will synthesize both empirical evidence and stakeholder expertise with the goal to help clinicians promote PA and reduce sedentary behaviour across the lifespan in individuals with CP. To help accomplish this, we have built an international consortium of clinicians and researchers, and will also engage community stakeholders and people living with CP, their families and caregivers.

Dr. Gorter will introduce the objectives of the Care Pathway and provide an overview of current PA promotion methods. Dr. Reedman and MSc student Xiong will present the published research evidence. MSc student Xiong and Dr. McPhee will present initial results of the development of the Pathway. Findings will be discussed with participants to identify knowledge gaps and to help shape the algorithm of the Pathway.

Course Format

10 Minutes: Introduction

Jan Willem Gorter, MD, PhD

Introduction of the project funded by the AACPDM Care Pathway Grant (2018)

10 Minutes: PA interventions over the life course for individuals with CP

Sarah Reedman, PhD; Julia Xiong, MSc student

Results of three systematic reviews published in 2017 and updates on interventions to increase PA

20 Minutes: Planning and development of the Care Pathway

Julia Xiong, MSc student; Patrick McPhee, PhD

Work to date in the development of the Care Pathway, including results of expert and stakeholder engagement

20 Minutes: Interactive discussion (moderator: Jan Willem Gorter)

Attendees will be invited to discuss the key components of the Care Pathway, identify gaps and help shape the algorithm by considering its clinical practicality and international implementation

Additional Figure 2. The health survey in the same format that participants saw on SurveyMonkey

2. Which health outcomes are most important for individuals with cerebral palsy, based on your expertise and experience?

	1	2	3	4	5	6	7	8	9
Cardiorespiratory endurance (aerobic fitness, aerobic capacity, oxygen consumption);	○	○	○	○	○	○	○	○	○
Body size [i.e., height and weight];	○	○	○	○	○	○	○	○	○
Body composition [i.e., waist circumference];	○	○	○	○	○	○	○	○	○
Sleep (sleep disorders);	○	○	○	○	○	○	○	○	○
Nutrition (diet, food intake behaviour/pattern);	○	○	○	○	○	○	○	○	○
blood pressure;	○	○	○	○	○	○	○	○	○
blood lipids and glucose;	○	○	○	○	○	○	○	○	○
depression;	○	○	○	○	○	○	○	○	○
anxiety	○	○	○	○	○	○	○	○	○

Other (please specify)

Additional Table 1. Rating of Nine Health Outcomes

Health Outcome	Ratings									
	1	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**	**Total**
Cardiorespiratory Endurance (aerobic fitness, aerobic capacity, oxygen consumption) (%)	0 (0)	1 (2)	2 (4)	3 (5)	5 (9)	10 (18)	14 (25)	8 (15)	12 (22)	55 (100)
Body size (height and weight) (%)	0 (0)	5 (9)	5 (9)	9 (16)	11 (20)	11 (20)	12 (22)	1 (2)	1 (2)	55 (100)
Body composition (i.e., waist circumference) (%)	1 (2)	3 (5)	10 (18)	4 (7)	12 (22)	8 (15)	10 (18)	5 (9)	2 (4)	55 (100)
Sleep (sleep disorders) (%)	0 (0)	0 (0)	3 (5)	4 (7)	7 (13)	9 (16)	17 (31)	7 (13)	8 (15)	55 (100)
Nutrition (i.e., diet, food intake behaviour/pattern) (%)	0 (0)	1 (2)	4 (7)	4 (7)	9 (16)	4 (7)	15 (27)	11 (20)	7 (13)	55 (100)
Blood pressure (%)	0 (0)	4 (7)	10 (18)	1 (2)	11 (20)	9 (16)	11 (20)	7 (13)	2 (4)	55 (100)
Blood lipids and glucose (%)	2 (4)	3 (5)	9 (16)	3 (5)	11 (20)	12 (22)	9 (16)	3 (5)	3 (5)	55 (100)
Depression (%)	0 (0)	0 (0)	2 (4)	3 (5)	9 (16)	9 (16)	14 (25)	6 (11)	12 (22)	55 (100)
Anxiety (%)	0 (0)	0 (0)	2 (4)	5 (9)	5 (9)	10 (18)	17 (31)	5 (9)	11 (20)	55 (100)

Additional Table 2. Matrix of z-score of pairwise comparisons of the ratings for the nine health

Health Outcomes (column-row)	Cardio-respiratory Endurance	pv	Body size	pv	Body composition	pv	Sleep	pv	Nutrition	pv	Blood pressure	pv	Blood lipids and glucose	pv	Depression	pv
Body size	4.65	0.0001														
Body composition	4.18	0.0005	-0.47	1.00												
Sleep	0.76	1.00	-3.89	0.0018	-3.42	0.011										
Nutrition	0.93	1.00	-3.72	0.0036	-3.25	0.021	0.17	1.00								
Blood pressure	3.51	0.0080	-1.14	1.00	-0.66	1.00	2.75	0.11	2.58	0.18						
Blood lipids and glucose	4.21	0.0005	-0.44	1.00	0.036	1.00	3.45	0.01	3.28	0.019	0.70	1.00				
Depression	0.33	1.00	-4.32	0.0003	-3.85	0.0021	-0.43	1.00	-0.60	1.00	-3.18	0.026	-3.88	0.0019		
Anxiety	0.382	1.00	-4.27	0.0004	-3.79	0.0027	-0.38	1.00	-0.55	1.00	-3.13	0.032	-3.83	0.0023	0.055	1.00

Pv: p-value; Statistically significant pairwise comparisons are marked in light grey

DISCUSSION

4.1 DISCUSSION OVERVIEW

The purpose of this thesis was to investigate and better understand SB in individuals with CP and to rate the importance of pre-determined health outcomes for PA in this population, based on expert opinion. A common tendency in research and clinical practice is to apply definitions of SB and tools to promote PA, which are known to be applicable and useful in the typically developing population, to individuals with CP. The reason is perhaps attributed to the paucity in research development in SB and PA specific to the CP population [1].

To address this knowledge gap, my work began by conducting a scoping review to better understand the operationalization of SB in individuals with CP, as published in the recent literature (Chapter 2). Mapping the current operationalizations of SB used in CP literature could help inform consensus on standard operationalized definitions, measurement and reporting of SB. This could be accomplished following the steps of the Terminology Consensus Project [2], a project by the SBRN for reaching consensus on the definition for SB and related terms in the typically developing population. The project was composed, firstly, of a literature review to identify the current uses of the SBRN definition for SB. Key terms from the search were collated, and a draft list of terms and their definitions was created for consensus, through a survey, by a panel of international experts. The SBRN definition of SB is composed of two parts, a postural and energy expenditure component [3]. For the postural component, I found that sitting and lying are considered SB by instruments used to measure SB time in individuals with CP. There is large variability in the cut-points of SB used by instruments to measure SB time in this population. For the energy expenditure component, I found that the definition for intensity of ≤1.5 MET in the typically postures of sitting and lying generally applies in individuals with CP;

as shown by studies that measured energy expenditure during these postures through indirect calorimetry. No studies reported reclining as part of the definition for SB or measured physiological components (e.g., energy expenditure, muscle activation) while reclining.

There is variability in i) the tools and cut-points for accelerometers that are used and validated and ii) the reporting of sedentary time. Oftentimes, the thresholds for SB were used from studies of validation in the typically developing population (e.g., <100 counts per minute) rather than from validation in individuals with CP. Furthermore, the reporting of SB times was highly variable. While most studies reported hours per day spent sedentary on consecutive days, others differentiated between time spent sedentary on a weekday versus weekend. This could impact results of sedentary time, since children with CP have been shown to spend significantly less sedentary time on weekdays compared to weekends [1]. Three studies reported percentages of sedentary time over 24 hours rather than only waking hours [4–6]. One study reported percent sedentary time, while assuming 12 hours to be the waking time [7]. The percentage of time in SB would be impacted by the denominator used to calculate that percentage. Further consensus on the reporting and measurement of SB is needed.

Both PA and SB are part of movement-related behaviours that occur during the whole day; they also interact to influence holistic health [8]. To cover the movement continuum, I conducted a conference-based survey to identify important outcomes for PA, from a list of pre-determined health outcomes, in individuals with CP. There was no research evaluating the opinions of experts on important health outcomes of PA in individuals with CP. In following the rating system of GRADE, the international standard for guideline development, the eventual goal in identifying these outcomes is to inform the development of clinical practices guidelines to manage PA in this population. I discovered the importance of both the physical and mental aspects of health to PA in individuals with CP, based on the opinions of clinicians and researchers.

The overarching aim of the thesis was to explore the movement continuum in individuals with CP. This general discussion aggregates the main findings of my thesis and provides an overarching interpretation. Strengths and limitations from these studies are addressed and outstanding knowledge gaps and future directions for advancing the field of work are provided for each chapter. Findings from the included manuscripts were discussed separately: (Chapter 1) The operationalization of SB in individuals with CP; (Chapter 2) Identifying important health outcomes for PA in individuals with CP.

4.2 ADVANCING OUR UNDERSTANDING OF SB IN CP

The scoping review in Chapter 1, consisting of 39 studies that operationalize SB in individuals with CP, was the most substantial contribution from my thesis. The aim of the review was to report on the operationalization of SB and postures/activities that constitute SB in CP across all ages and severity levels. To accomplish this, I found studies that measured the physiological aspects of the typically sedentary postures of sitting and lying, namely muscle activation, energy expenditure and oxygen consumption. I also collected information on the definition of the term "sedentary" reported in the CP population. I identified that perhaps, there is more consistency and alignment with the SBRN definition [3] of SB in the typically developing population, than expected. While there is considerable variability in the measurement tools and thresholds used for SB, the majority of studies included in this review that employed activity monitors to measure time spent in SB used the postural definition of sitting and lying to characterize SB. Five out of the six studies that measured energy expenditure in typically sedentary postures, sitting and lying, supported the energy expenditure definition of ≤1.5 METs in these postures. The postural definition was applied mainly by activity monitors in the postures constituted as sedentary. The energy expenditure operationalization of SB was determined through validation studies, which compared the results of instruments, generally accelerometers, to measure SB against indirect calorimetry. The intensity operationalization of sitting and lying in CP was also determined through studies that were not validating a tool but aimed to directly measure energy expenditure in the sitting and lying postures. Only one study measuring energy expenditure in sitting and lying produced results of >1.5 METs [7]. However, these results for sitting and lying were only measured and observed in two participants using an indirect method for converting heart rate and accelerometry into energy expenditure. The finding that the energy expenditure definition of ≤1.5 METs also applied to typically sedentary postures in individuals with CP is significant because it indicates that researchers need only to measure sitting or lying time and show a reduction in these postures in order to demonstrate increase in energy expenditure. This finding is significant because it may be more feasible for researchers to measure postures through activity monitors rather than energy expenditure directly through indirect calorimetry, especially in daily living situations or population-based studies. However, we do not know if energy expenditures of >1.5 METs has physiological importance in individuals with CP.

My work advances the field in various ways. In the future, a similar process to the initiative carried out by the SBRN for the typically developing population, called the Terminology Consensus Project [2], can be carried out to establish consensus on the definitions for SB and SB related terms in individuals with CP across all ages and functional levels. The scoping review I conducted is analogous to the "literature review" conducted in the Terminology Consensus Project, to establish key terms and definitions in SB research in the CP population. As physical inactivity and SB are still terms that are used interchangeably in the literature, this review adds greater awareness of the need to differentiate between these two constructs in CP research.

Chapter 1 had numerous strengths. The design and reporting of the scoping review were done using rigorous 9-step approach outlined by the Joanna Briggs Institute Reviewer's Manual [10]. A second reviewer (SR) and I conducted all screening independently and in duplicate and completed 20% of the data abstraction independently and then in duplicate. The review had strong expert involvement; the research questions were designed in collaboration with three experts in PA. I discussed the data extraction table with 2 experts in the field of PA in individuals with CP to ensure all the relevant information from included full-text articles were extracted. Furthermore, the title/abstract screening and 20% of data extraction was done independently and then in duplicate with one physiotherapist with expertise in individuals with CP.

It should be acknowledged that the purpose of the scoping review was not to develop an operationalized definition of SB, but rather report on the process of the way in which other studies have attempted to break up the construct into measurable variables. That means that just because many the studies reported sitting and lying as the variables used to measure SB in individuals with CP, this does not indicate that these postures are of physiological importance to measure in individuals with CP. Hence, the limitation is that perhaps the included studies have operationalized SB following the SBRN standards rather than choosing a definition that has physiological significance based on literature. The limitation only highlights the importance of studies in this review that operationalized SB through the physiological components of energy expenditure, muscle activation and oxygen consumption. These studies begin to elucidate the physiological importance of the sitting posture, since, while the mechanism of the deleterious effects of sitting is unknown, it has been hypothesized to be due to differences in muscle metabolism and energy expenditure [11]. Furthermore, one study used “sitting” to represent SB when testing the validity of an instrument but “sitting and lying” to represent SB when testing the reliability of the instrument, without explaining their rationale for counting lying as SB only in the reliability study [12]. This further calls into question the rationale used by studies for constituting a behaviour as “sedentary”. Some included studies did not explicitly define SB. In two studies included in the review, the introduction or discussion says that time spent “sitting/lying” are representative “sedentary behaviours” [13–14]. The studies then states, in the methods, that “sitting and lying” were measured, without further indicating that these two postures constituted SB. While this was not a clear delineation of sitting/lying as a “sedentary” behaviour, it was included in the review because the operationalization of SB was described somewhere in the article. However, this raises the question whether other articles that intend to measure SB time were excluded because they did not include the term “sedentary”. While this may be a limitation with the scoping review, perhaps it highlights a limitation in the field of SB research in individuals with CP, in which the term “sedentary” is not used to describe behaviours which are intended to be described as such. The limitation calls for a standard operationalized definition for SB in individuals with CP so that researchers not only report consistently but also describe their behaviours measured using the term “sedentary”. This reporting would help advance the field by making studies that measure SB easier to search and build upon.

The terms “sedentary” and “inactive” were used by one study interchangeably [15]. This study states, the methods, that the Evenson [16] accelerometer cut points of <100 counts per minute was used to classify inactivity. However, the results section states that ambulatory children with CP spent 8:36 hours inactive, spent 2:38 hours in light activities, and spent 0:44 hours in MVPA. However, the Evenson [16] study classified the cut points of <100 counts per minute as SB. The study further highlights the need for the consistent operationalized definition of SB to be established and promulgated in this field research with individuals with CP.

Another limitation of the review is that the relationship between health indicators and SB was only reported for studies which characterized the behaviour measured as being "sedentary". This further highlights the limitation above, which is that studies measuring the association between health indicators and typically sedentary postures, such as sitting, that were not characterized using the term "sedentary" would not be included. The scoping review rather reports the way in which the studies that explore a relationship with a health indicator, operationalizes SB. This limitation limits the usability of studies from this scoping review for future systematic reviews on exploring the health outcomes of SB in this population.

4.3 ADVANCING OUR UNDERSTANDING OF IMPORTANT HEALTH OUTCOMES FOR PA IN CP

My study in Chapter 2 identified five critical health outcomes (i.e., sleep; nutrition, cardiorespiratory endurance, depression; anxiety) and four important health outcomes (i.e., body size, body composition, blood lipids and glucose, blood pressure). Engaging frontline therapists in the implementation of clinical research is highly valuable, as they are essential in developing, testing and implementing effective management of PA in individuals with CP. Clinicians and researchers can help identify the health outcomes that are relevant to individuals with CP by integrating their clinical expertise and experience working directly with these patients and because they may have a better understanding of the physiological importance of health outcomes towards the overall health of the population. One current contribution of this chapter is in helping to inform the evidence profile for the development of the clinical practice guideline, AACPDM Care Pathway, on PA promotion in individuals with CP. A panel with patients and experts is currently being formed to inform this initiative.

To my knowledge, no previous studies have sought to rate health outcomes as they relate to PA in individuals with CP, as no clinical management plans for PA have been developed specifically for this population. One study has used expert discussion to explore the importance of core outcomes consisting of multimorbidity risk factors in individuals with CP [17]. Another study listed recurring outcomes from key research priorities, such as examining any interventions to improve participation and the quality of life [18]. However, these outcomes were not rated in the context of PA. My study aims to inform the first steps in clinical practice guideline development for PA in individuals with CP, in a similar manner to the initial steps for PA guideline development, including the 24-Hour Movement Guidelines [19], by reporting the rating for nine pre-determined health outcomes in a transparent manner. By using GRADE, an international standard for guideline development, the results can be more easily incorporated in future clinical guideline development in PA management for individuals with CP.

Our intent of identifying important health outcomes of PA that are relevant to individuals with CP comes in light of recognizing that no clinical management guidelines in individuals with CP have been developed. The first step of GRADE calls for the identification of patient-important outcomes, which would impact the evidence profile included to inform guidelines and also impact the rating of the quality of evidence. While the findings of this study provide some important and critical health outcomes for PA in individuals with CP based on the perspective of clinicians and researchers, these outcomes do not have patient input. This is important since ultimately, any future guideline developments for PA management would affect patients.

Ultimately, for future studies, patient input is needed to inform and rate the importance of a more comprehensive list of outcomes [20]. These outcomes can then inform the next step of GRADE, which is conducting a systematic review of PA interventions that measure the patient-important outcomes.

A significant finding from the second chapter is that individuals with CP have many of the same health concerns, based on the experts' opinions, compared to that of the typically developing population. These similarities include cardiorespiratory endurance, body composition, blood lipids and glucose and blood pressure [21,22]. While the 24-Hour Movement guidelines found these outcomes to be critical, most experts in my study rated them as important, with the exception of cardiorespiratory endurance which was rated as critical. One reason could be that outcomes rated as important are substitutes for outcomes that are most important to patients, which are clinical endpoints. The GRADE Approach suggests using surrogate outcomes only when high-quality evidence regarding important outcomes is lacking [20]. However, surrogate outcomes are sometimes necessary important outcomes are relatively infrequent or occur over long periods of time [20]. However, choosing surrogate outcomes would cause evidence to be rated down in further steps in guideline development for directness for measuring outcomes important to patients [23].

The strength of chapter 2 is that the conference-based survey conducted with an international group of respondents representing a large number of specialties. I also had a fairly high number of responders with 55 experts who participated in the voting. The major limitation is that because of the method I used to select the literature, which included a non-systematic search of research on potentially important outcomes in this population, with a final review of the included outcomes by only 2 experts, our findings cannot and should not be interpreted to be an exhaustive list of all important and critical outcomes for PA in this population. Rather, it should indicate the important and critical nature of only the nine pre-determined health outcomes, which has evidence from literature to be important in the population. This limitation prevents direct use of information in this study for future guideline development without more comprehensive studies, with greater expert input and patient input and a systematic literature review, to create exhaustive list of outcomes. Burns et al. [24], in their guideline for the design of self-administered surveys of clinicians, provides suggestions for item generation in a more comprehensive manner, including using “literature reviews, in-depth interviews, focus-group sessions, or a combination of these methods with potential respondents or experts”. An initial systematic search of literature can be done on to examine health indicators that have been reported from PA intervention studies in individuals with CP. Expert and patient input can then help identify additional potentially important outcomes, from literature or their own expertise or experiences, since the GRADE approach specifies that guideline developers must base the choice of outcomes on what is important, not on what outcomes are measured. The focus groups would be charged with the task of reviewing these initial items generated, along with that of generating additional items [25]. Finally, through conducting the groups at the initial stages of the study, a rough list of potentially important health outcomes would be pre-tested indirectly [25].

4.4 CONCLUSIONS

PA and SB are key constructs in the formula for health and well-being in individuals with CP. This thesis addressed several important gaps in the existing literature, which were the need to operationalize SB and report on health outcomes relevant to PA, that are important in individuals with CP. There should be agreement on the operationalization and reporting of SB so that studies can be aggregated to advance this slow-advancing field of study in CP research.

International experts at the 2019 AACPDM/IAACD conference demonstrated keen interest in the promotion of healthy physical behaviours for individuals with CP, across ages and levels of severity. Rigorous research rests upon standardized definitions of terms for consistent reporting. Research that is relevant to stakeholders is conducted through obtaining their input. The results of these projects serve to further the development of standardized and relevant research in the field of SB and PA in individuals with CP.

4.5 REFERENCES

1. Ganz F, Hammam N, Pritchard L. Sedentary behavior and children with physical disabilities: a scoping review. Disabil Rehabil. 2020;1–13.

2. on behalf of SBRN Terminology Consensus Project Participants, Tremblay MS, Aubert S, Barnes JD, Saunders TJ, Carson V, et al. Sedentary Behavior Research Network (SBRN) – Terminology Consensus Project process and outcome. Int J Behav Nutr Phys Act. 2017;14(1):75.

3. Sedentary Behaviour Research Networ. Letter to the Editor: Standardized use of the terms "sedentary" and "sedentary behaviours." Appl Physiol Nutr Metab. 2012;37(3):540–2.

4. Nooijen CF, Slaman J, Stam HJ, Roebroeck ME, Berg-Emons R, Learn2Move Research Group. Inactive and sedentary lifestyles amongst ambulatory adolescents and young adults with cerebral palsy. J NeuroEngineering Rehabil. 2014;11(1):49.

5. Slaman J, Roebroeck M, Dallmijer A, Twisk J, Stam H, van den Berg-Emons R, et al. Can a lifestyle intervention programme improve physical behaviour among adolescents and young adults with spastic cerebral palsy? A randomized controlled trial. Dev Med Child Neurol. 2015;57(2):159–66.

6. Shkedy Rabani A, Harries N, Namoora I, Al-Jarrah MD, Karniel A, Bar-Haim S. Duration and patterns of habitual physical activity in adolescents and young adults with cerebral palsy. Dev Med Child Neurol. 2014;56(7):673–80.

7. Williams SA, McFadden LM, Blackmore AM, Davey P, Gibson N. Do adolescents with cerebral palsy meet recommendations for healthy weight and physical activity behaviours? Disabil Rehabil. 2019;1–6.

8. Tremblay MS, Carson V, Chaput J-P, Connor Gorber S, Dinh T, Duggan M, et al. Canadian 24-Hour Movement Guidelines for Children and Youth: An Integration of Physical Activity, Sedentary Behaviour, and Sleep. Appl Physiol Nutr Metab. 2016;41(6 (Suppl. 3)):S311–27.

9. Verschuren O, Peterson MD, Leferink S, Darrah J. Muscle Activation and Energy-Requirements for Varying Postures in Children and Adolescents with Cerebral Palsy. J Pediatr. 2014;165(5):1011–6.

10. Peters M, Godfrey C, McInerney P, Munn Z, Tricco A, Khalil H. Chapter 11: Scoping reviews. In: JBI Reviewer's Manual [Internet]. 4th ed. JBI; 2019 [cited 2020 Jun 28]. Available from: https://wiki.joannabriggs.org/display/MANUAL/Chapter+11%3A+Scoping+reviews

11. Hamilton MT, Hamilton DG, Zderic TW. Role of Low Energy Expenditure and Sitting in Obesity, Metabolic Syndrome, Type 2 Diabetes, and Cardiovascular Disease. Diabetes. 2007;56(11):2655–67.

12. Bania T. Measuring Physical Activity in Young People with Cerebral Palsy: Validity and Reliability of the ActivPAL™ Monitor: Measuring PA in People with CP. Physiother Res Int. 2014;19(3):186–92.

13. Bania TA, Taylor NF, Baker RJ, Graham HK, Karimi L, Dodd KJ. Gross motor function is an important predictor of daily physical activity in young people with bilateral spastic cerebral palsy. Dev Med Child Neurol. 2014;56(12):1163–71.

14. Bania TA, Dodd KJ, Baker RJ, Graham HK, Taylor NF. The effects of progressive resistance training on daily physical activity in young people with cerebral palsy: a randomised controlled trial. Disabil Rehabil. 2016;38(7):620–6.

15. Mitchell LE, Ziviani J, Boyd RN. Habitual Physical Activity of Independently Ambulant Children and Adolescents With Cerebral Palsy: Are They Doing Enough? Phys Ther. 2015;95(2):202–11.

16. Evenson KR, Catellier DJ, Gill K, Ondrak KS, McMurray RG. Calibration of two objective measures of physical activity for children. J Sports Sci. 2008;26(14):1557–65.

17. Mcphee PG, Claridge EA, Noorduyn SG, Gorter JW. Cardiovascular disease and related risk factors in adults with cerebral palsy: a systematic review. Dev Med Child Neurol. 2018; 61(8):915-923.

18. Gross PH, Bailes AF, Horn SD, Hurvitz EA, Kean J, Shusterman M, et al. Setting a patient-centered research agenda for cerebral palsy: a participatory action research initiative. Dev Med Child Neurol. 2018;60(12):1278–84.

19. Canadian 24-Hour Movement Guidelines : CSEP | SCPE [Internet]. [cited 2020 Jul 5]. Available from: https://csepguidelines.ca/

20. Guyatt GH, Oxman AD, Kunz R, Atkins D, Brozek J, Vist G, et al. GRADE guidelines: 2. Framing the question and deciding on important outcomes. J Clin Epidemiol. 2011;64(4):395–400.

21. Carson V, Lee E-Y, Hewitt L, Jennings C, Hunter S, Kuzik N, et al. Systematic review of the relationships between physical activity and health indicators in the early years (0-4 years). BMC Public Health. 2017;17(S5):854.

22. Poitras VJ, Gray CE, Borghese MM, Carson V, Chaput J-P, Janssen I, et al. Systematic review of the relationships between objectively measured physical activity and health indicators in school-aged children and youth. Appl Physiol Nutr Metab. 2016;41(6 (Suppl. 3)):S197–239.

23. Guyatt GH, Oxman AD, Vist G, Kunz R, Brozek J, Alonso-Coello P, et al. GRADE guidelines: 4. Rating the quality of evidence—study limitations (risk of bias). J Clin Epidemiol. 2011;64(4):407–15.

24. Burns JS, Williams PL, Korrick SA, Hauser R, Sergeyev O, Revich B, et al. Association Between Chlorinated Pesticides in the Serum of Prepubertal Russian Boys and Longitudinal Biomarkers of Metabolic Function. Am J Epidemiol. 2014;180(9):909–19.

25. Nassar-McMillan SC, Borders LD. Use of focus groups in survey item development. Qual Rep. 2002;7(1):1–12.