Wearable sensors, cerebral palsy and gait assessment in everyday environments: is it a reality? – A systematic review

Ana Francisca Rozin Kleiner[a]
Anderson Belgamo[b]
Aline Souza Pagnussat[c]
Anselmo de Athayde Costa e Silva[d]
Chiarella Sforza[e]
Nelci Adriana Cicuto Ferreira Rocha[a]

[a] Department of Physiotherapy, Neuropediatrics Section, Federal University of São Carlos, São Carlos, Brazil
[b]Federal Institute of São Paulo (IFSP), Piracicaba, Brazil
[c]Rehabilitation Sciences Graduate Program, Universidade Federal de Ciencias da Saude de Porto Alegre (UFCSPA),Porto Alegre, Brazil
[d]Physical Education Faculty – Pará Federal University, Belém, Pará, Brazil
[e]Department of Biomedical Sciences for Health, The University of Milan, Milan, Italy

Correspondence to: Ana Francisca Rozin Kleiner
E-mail: anafrkleiner@gmail.com

Summary

This systematic review aimed to investigate emerging methods used to quantify gait parameters in children with cerebral palsy (CP) in everyday environments.
The StArt computational tool automatically screened the following databases: ACM, Engineering Village, IEEE, PubMed, Scopus and Web of Science from inception to June 2018. Studies reporting the use of wearable sensors to assess gait in daily settings in children with CP were included.
Data regarding 1563 studies were extracted, but only three studies could be included on the basis of the inclusion/exclusion criteria. These studies proposed wearable technologies based on the use of signals provided by triaxial accelerometers and force resistive pressure sensors. These are able to track levels of activity and detect falls, gait deviations and gait symmetry in children with CP in their daily environments. To date, only two types of sensors have been tested in this population and it remains to be clarified how wearable sensors, used to quantify activity level, might benefit children with CP.

KEY WORDS: cerebral palsy, gait, inertial measurement unit, walking, wearable sensor.

Introduction

The International Classification of Functioning, Disability and Health (ICF) (WHO, 2001) states that the functional and disability processes in children with cerebral palsy (CP) are strongly influenced by the contexts in which they conduct their daily lives. However, much of the available evidence about movement and mobility in this population focuses mainly on the components of body structure and function that contribute to and/or influence their functional activities (Kleiner et al., 2015). There is a lack of research concerning contextual factors that may affect the mobility (Palisano, 2006) of children with CP. But such factors are very important, as adolescents with CP have been reported to walk less and to be less physically active than their peers without CP (Bjornson et al., 2007).
From a biomechanical point of view, walking ability is usually evaluated through gait analysis (GA). GA is quantitative, multifactorial, three-dimensional computerized evaluation of walking (Perry and Burnfield, 2010) performed by means of technology such as specialized, computer-interfaced video cameras to measure patient motion, electrodes placed on the surface of the skin to detect muscle activity, and force platforms embedded in a walkway to monitor the forces and torques produced between the ambulatory patient and the ground (Perry and Burnfield, 2010). GA is an important method of quantifying gait that provides information able to shed light on the etiology of gait abnormalities and help guide treatment decision making. However, understanding CP locomotion in an ecological context is also necessary in order to reach more effective rehabilitation goals (Kleiner et al., 2015). Researchers carry out GA in a motion analysis laboratory, which means they do not evaluate children in an ecological context. The examination is performed in an optimized environment (e.g., on a flat surface), and patients need to wear electrodes and anatomical markers to allow their walking to be assessed. However, their performances in the laboratory do not accurately reflect their behavior in the community. Furthermore, self-report methods that are available for measuring behavior in the community are subject to several different types of bias (Hiratuka et al., 2010; Palisano et al., 1997; Graham et al., 2004; Harvey et al., 2007).
Recently, interest has shifted towards the possibility of conducting gait assessments in everyday environments, facilitating long-term monitoring (Bonato, 2010). This is made possible by the use of wearable technologies rather than laboratory-based equipment (Iosa et al., 2016). Recent advances in wearable sensors, especially inertial body sensors, have opened up a promising future for GA. Not only are these sensors easier to adopt in clinical diagnosis and treatment procedures than their current counterparts, they also make it possible to mon-

itor gait continuously outside clinics, thereby allowing seamless patient analysis from clinics to free-living environments (Iosa et al., 2016; Piwek et al., 2016; Majumder et al., 2017).
The purpose of this paper was to provide a systematic review of emerging methods used to quantify the gait of children with CP in everyday environments. More specifically, this systematic review aimed to answer the following questions:
RQ1. What types of wearable sensors are used to monitor the gait of children with CP in day-to-day settings?
RQ2. What are these wearable sensors able to track?

Materials and methods

This systematic review is reported according to the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) guidelines. It was decided to perform this study as a systematic literature review according to the guidelines proposed by Kitchenham (2004), given that these offer the possibility of using the StArt computational tool (Fabbri et al., 2016), which provides resources to assist the researcher throughout the process of conducting the systematic review, from the creation of the research protocol through to data mining and reporting.
The use of the StArt tool forces a researcher to follow the steps of a systematic review: (1) planning; (2) execution and (3) summarization/reporting. In the first phase, investigators must identify the need for a review and create a review protocol containing important information about the systematic review. In the second phase, they should identify and select relevant primary studies, and extract and synthesize data. Finally, in the third phase, they should summarize and report the results of the systematic review to relevant communities.

The planning phase

Identification and selection of studies

A literature search was carried out from the earliest date of publication, referring to the subject of interest, up to April 20th 2018, using the following databases: ACM Digital Library, Engineering Village, IEEE, PubMed, Scopus and Web of Science. There were no additional records identified through other sources.
The keywords used in the survey were: *cerebral palsy*, *inertial measurement unit*, *smartphone*, *wearable sensor*, *gait, walking*. An initial study was conducted to identify an adequate combination of these keywords in order to conduct the research. The following search strings were then defined:

Search string 1 (SS1): («cerebral palsy» OR CP) AND (Inertial Measurement Unit (IMU) or smartphone or «wearable sensor») AND (gait or walking)

Search string 2 (SS2): («inertial measurement unit» OR «IMU») AND («cerebral palsy» OR CP) AND gait

Search string 3 (SS3): («cerebral palsy» OR CP) AND wearables AND gait

Search string 4 (SS4): («cerebral palsy» OR CP) AND smartphone AND gait.

The four search strings were applied to all the databases used and all results were imported into the StArt tool. We chose to develop four strings in order to increase the number of papers detected relevant to this systematic review.

Inclusion and exclusion criteria

The articles identified by the search and imported into the StArt tool were evaluated by reading their titles and abstracts. Two independent researchers reviewed the titles and abstracts of the papers to determine their eligibility for inclusion. In the event of doubt, the article was read in full before a decision was rendered. To be included, manuscripts had to be methodological studies, written in English, and relate to CP monitoring through the use of wearable sensors.
Articles were excluded if they met the following criteria: conference proceedings introduction; only abstract available; monitoring performed in movement analysis laboratories or clinical settings; manuscript not related to CP; article dealing with robotic systems or exoskeletons. In accordance with Cochrane's recommendations for the formulation of systematic reviews, case studies and reviews were also excluded (Higgins and Green, 2006).

Features of the studies and quality assessment

An analysis of the selected articles was performed, recording the following data: (1) participant characteristics: participants' CP diagnosis and presence or absence of a control group of typical children or adults; sample size; participants' age; CP gait type; and use of classification systems such as the Gross Motor Function Classification System (GMFCS); (2) wearable system type, body placement and sensor type; (3) type of monitoring.
The methodological quality of studies was assessed using a rating checklist adapted from previous systematic reviews (Costa et al., 2013; Soh et al., 2011). The questions were selected, according to the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) guidelines (Von Elm et al., 2007), from the literature on the development of quality criteria described in the Cochrane Handbook for Systematic Reviews, and from the Critical Appraisal Skills Programme, developed by the Oxford Regional Health Authority (Milne et al., 1995). The checklist covered the following aspects: (1) presentation of study objectives; (2) rationale for study hypotheses; (3) use of appropriate design to meet objectives; (4) participant delineation, (5) inclusion criteria proposed by the study; (6) description of volunteer recruitment; (7) description of sampling type; (8) ethical aspects; (9) volunteers not participating in or excluded from the study; (10) sample computation for volunteer selection; (11) description of variables; (12) use of appropriate statistical methods to analyze the results; (13) descriptive measures of precision or variability of study results; (14) the study's external validity; (15) findings presented in a clear, objective manner; and (16) the study's limitations. The score assigned for each item indicates the clarity in the description of the study data, viz., 1 for a study that meets the requirement and 0 for one that does not address it; the scores thus add up to a maximum of 16 points.

A study that scores from 12 to 16 points has few methodological limitations and is considered to be of good quality; a study scoring 7 to 11 points presents moderate methodological limitations and is considered fair; and a study scoring fewer than 7 points demonstrates significant methodological limitations, thus its quality is deemed poor.

Results

The execution phase

In total, 1563 articles were found. Table I shows the number of articles found in each database when applying each of the search strings (SS1, SS2, SS3 and SS4).

Of these 1563 articles, 422 articles were automatically classified as duplicates by the StArt tool. Thus, there remained 1141 unclassified articles which were analyzed, considering the title and abstract and applying the inclusion and exclusion criteria presented previously. Of these 1141 articles, 1133 were rejected in the article selection phase on the basis of the exclusion criteria. The exclusion criteria most frequently met were: «manuscript not related to cerebral palsy» (840 times), followed by «conference proceedings introduction» (101 times) and «clinical monitoring» (85 times).

Consequently, 8 articles were selected to be read in full: Smith and Bagley (2010), Mancinelli et al. (2012), Chong and Yunus (2012), Strohrmann et al. (2013), Zhang et al. (2014), Leite and Postolache (2017), Carcreff et al. (2018), and Hegde et al. (2018). Note that the oldest article selected was written in 2010, and 50% of the articles selected were published in the last five years: 2018 (n=2); 2017 (n=1); 2014 (n=1).

Finally, 5 studies were rejected: 2 because they did not involve children with CP (Chong and Yunus, 2012; Leite and Postolache, 2017); 1 because the subjects were evaluated using a delimited environment that was not part of the children's day-to-day settings (Strohrmann et al., 2013); 1 paper because the data were collected in a laboratory environment (Carcreff et al., 2018). Furthermore, as the studies of Zhang et al. (2014) and Hegde et al. (2018) come from the same research group, and Zhang et al. (2014) is a feasibility study in which the instrument is tested in just 3 children with CP, we also decided to exclude this study.

Figure 1 shows the PRISMA flowchart illustrating the study selection process, and the three selected articles

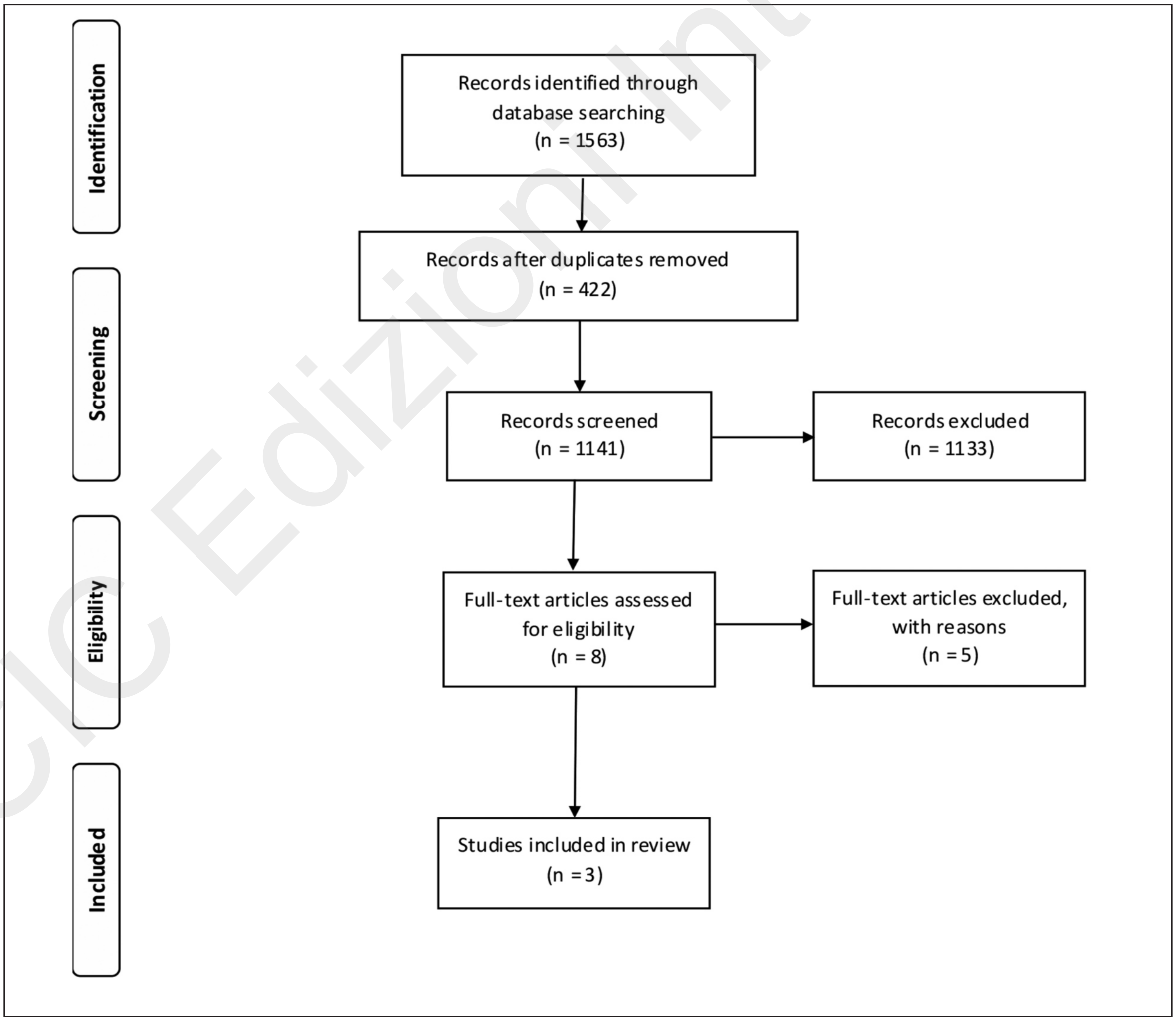

Figure 1 - PRISMA flowchart.

Table I - Numbers and percentages of papers uploaded by StArt from each database.

Strings	ACM	Engineering Village	IEEE	PubMed	Scopus	Web of Science	Total
SS1	669	61	2	3	13	12	760
SS2	0	56	0	2	6	7	71
SS3	1	66	239	0	39	0	345
SS4	1	0	381	1	2	2	387
Total	671	183	622	6	60	21	1563
%	42.9%	11.7%	39.8%	0.3%	4%	1.3%	100%

are detailed in Table II. Table III reports the methodological quality of the studies under review, considering all the rated sub-items.

Discussion

The summarization/reporting phase

The objective of this systematic review was to search for emerging methods used to quantify the gait of subjects with CP in everyday environments. More specifically, it aimed to identify the type of wearable sensors used to monitor this population in day-to-day settings, as well as to identify what these devices are able to track. We found three articles, spanning the period 2010 to 2018. Two of them had good methodological quality (Mancinelli et al., 2012; Hedge et al., 2018). The remaining study (Smith and Bagley, 2010) showed moderate limitations, and was therefore of average methodological quality.

Smith and Bagley (2010) developed a miniature, wearable activity/fall monitor positioned inside a fanny pack. The wireless monitors digitally streamed tri-axial acceleration data to the hard drive of a base station laptop computer. The researchers attached fanny packs to children's lower backs, and amplitude, impact, and orientation information in the accelerometer signals determined their level of activity (i.e. standing, lying down, walking, running, jumping) and detected falling. This wearable automatically logs 1-minute updates of a child's activity level and detects and logs when a child falls down; it works automatically for two weeks, 24 hours a day.

Mancinelli et al. (2012), presented ActiveGait, a novel sensorized shoe-based system for monitoring gait deviations. The ActiveGait system was specifically designed to gather gait data from children with CP in their home and in community settings. The wearable sensor consists of an instrumented shoe with 15 insole sensors (iSole), one ankle angle sensor, a data collection unit, as well as external download hardware and data processing software. The iSole was made up of 15 force sensitive resistors positioned within a custom-made insole. Eleven of the sensors were 0.5" in length, while the other 4 sensors were 1.5" in length.

The short sensors were placed in the metatarsal region of the insole. The long sensors were placed in the heel region. Placing more sensors towards the forefoot allowed greater accuracy of the center of pressure trajectory estimates, especially in children who showed a toe-walking pattern. The preliminary results presented in this paper showed that the ActiveGait system may be a useful tool for the longitudinal monitoring, in day-to-day settings, of gait deviations and toe-walking severity in children with CP.

Hegde et al. (2018) developed the pediatric SmartShoe, a wearable sensor system for performing ambulatory monitoring of physical activity and gait in children with CP. This wearable consisted of resistive pressure sensors and one 3-D accelerometer.

The five pressure sensors were embedded in the insole and were placed under the heel, the heads of the metatarsal bones and the hallux. The 3-D accelerometer and Bluetooth wireless electronics were placed in a box attached to the back of the shoe. The set of sensors utilized made it possible to monitor important phases of the gait cycle, such as heel strike, stance phase and toe-off. Pressure and acceleration data were sent via Bluetooth communication to a Windows smartphone. The phone had a program with a user-friendly interface, allowing the user to select sensors and sampling rate, and to start or stop the data-collecting process. The system was light, minimally obtrusive and low cost in terms of power consumption.

The sensor data were accumulated on the internal storage of the smartphone and later transferred to a personal computer for processing. This wearable sensor system is able to measure gait symmetry in children with CP in the community environment. The system was based on unobtrusive shoe sensors which are convenient for everyday real-life usage.

With regard to RQ1, two types of wearable sensors were found: a fanny pack (Smith and Bagley, 2010) and shoes (Mancinelli et al., 2012; Hegde et al., 2018). These wearable technologies were based on triaxial accelerometer signals and/or force resistive pressure sensors. In answer to RQ2, these sensors are able to track the level of activity, and detect falls and gait deviations, as well as gait symmetry, in children with CP in their daily environments.

Study limitations

Even though there are many types of wearables that could provide important information about the activity levels of children with CP (Bonato, 2010; Iosa et al., 2016; Piwek et al., 2016; Majumder et al., 2017), only two types have been tested in this population so far. Thus, it remains unclear how wearable sensors, providing an ever-increasing stream of behavioral and physiological feedback, may benefit children with CP. Moreover, we adopted excessively strict inclusion and exclusion criteria in this study, conducted in order to observe and understand what type of wearable devices are being developed specifically to assess the daily living of

Table II – Description of subjects and study design.

Study	Groups	Sample Size	Age	Gait Type	GMFCS	Wearable Type	Body placement	Sensor	Monitoring
Smith and Bagley (2010)	CP TD	35 51	2-14 years	-	- -	A miniature, battery-powered microcontroller-based activity/fall monitor worn by the child in a small fanny pack during everyday living.	Lower back	Triaxial accelerometer signals	Algorithms for automatic real-time processing of the accelerometer signals to monitor a child's level of activity and to detect falls. The monitor continuously logs 1-minute activity levels and the occurrence and characteristics of each fall over two-week recording sessions.
Mancinelli et al. (2012)	CP	11	12.6± 3 years	Equinus and crouch gait patterns with disability ranging from very mild to moderate or severe.		Instrumented shoe with 15 insole sensors (iSole).	Foot	A sensorized shoe-based system for monitoring gait deviations.	The data was collected under supervised and unsupervised conditions, for example: laboratory setting, outdoor settings, and home settings. The methodology derives from severity measures based on features extracted from CoP trajectories.
Hegde et al. (2018)	CP TD	10 11	4-9 years	A variety of types of lower extremity impairment found among the children with CP.	(6) GMFCS II (4) GMFCS I	Wearable shoe sensor system (SmartShoe).	Foot	Sensors embedded in the shoe included 5 force resistive pressure sensors and one 3-D accelerometer.	This wearable sensor system is used for monitoring physical activity and gait. Novel data processing techniques were developed to remove the effect of orthotics on the sensor signals. Machine learning models were developed to automatically classify activities of daily living.

Abbreviations: CP, cerebral palsy; TD, typically developing; GMFCS, Gross Motor Function Classification System levels; CoP, center of pressure

Table III - Methodological quality of the studies under review.

Sub-items	Objective		Study Design		Partici-pants		Methodological/ Statistical analysis					Results		Discussion			Score	Quality
	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	Total	Classification
Smith and Bagley (2010)	1	1	1	1	0	0	0	1	0	0	1	1	1	1	1	1	11	Moderate
Mancinelli et al. (2012)	1	1	1	1	1	1	1	1	0	0	1	1	1	1	1	1	14	Good
Hegde et al. (2018)	1	1	1	1	1	0	1	1	0	0	1	1	1	1	1	1	13	Good

children with CP. We agree that several types of wearable sensors are being developed to asses this population, but they are still being tested in the optimal controlled conditions typical of a laboratory, a setting that was not the focus of this study. These considerations explain why just 3 papers (out of 1563 articles) met the inclusion/exclusion criteria.

Concluding remarks

Since measurements of activity levels and gait patterns are important aspects of CP rehabilitation programs, wearable sensors are a promising technology in this field. Studies that aim to develop wearable sensors for monitoring the gait of children with CP may help physicians and therapists to understand each patient's profile, and thus to quantify the impact of rehabilitation programs in terms of children's activity levels. Additionally, the detection of children's gait patterns in daily life may help in determining the nature of the impairments to be treated, and in assessing the effects of treatment on function in truly ecological conditions.

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