Review

Psychometric properties of measures of gait quality and walking performance in young people with Cerebral Palsy: A systematic review

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ABSTRACT

Availability of outcome measures (OMs) with robust psychometric properties is an essential prerequisite for the evaluation of interventions designed to address gait deterioration in young people with Cerebral Palsy (CP). This review evaluates evidence for the reliability, validity and responsiveness of outcome measures of gait quality and walking performance in young people with CP. A systematic search was performed in MEDLINE, CINAHL, PubMed and Scopus. Articles that met the eligibility criteria were selected. Methodological quality of studies was independently rated by two raters using the modified COnsensus-based Standard for the selection of health status Measurement INstruments checklist. Strength of evidence was rated using standardised guidelines. Best evidence synthesis was scored according to Cochrane criteria. Fifty-one articles reporting on 18 distinct OMs were included for review. Best evidence synthesis indicated a moderate to strong evidence for the reliability for OMs of walking performance but conflicting evidence for the reliability of OMs of gait quality. The evidence for responsiveness for all OMs included in this review was rated as 'unknown'. The limitations of using the modified COSMIN scoring for small sample sizes are acknowledged. Future studies of high methodological quality are needed to explore the responsiveness of OMs assessing gait quality and walking performance in young people with CP.

1. Introduction

Even in those children and adolescents with Cerebral Palsy (CP) which are ambulatory, walking ability is often affected. Not surprisingly, considerable research effort has been therefore directed towards improving or maintaining the young person's walking ability for example through surgery [1], orthotic interventions [2], botulinum toxin [3] selective dorsal rhizotomy [4] and exercise programmes [5].

Crucial for this research, is the use of reliable, valid and responsive outcome measures to meaningfully evaluate the success of these interventions [6]. Psychometric properties have been defined as the elements that contribute to the statistical adequacy of a measurement instrument in terms of reliability, validity, measurement error and internal consistency [6]. Critically evaluating the psychometric properties of available OMs can provide essential knowledge and evidence for clinicians and researchers, allowing the selection of the most appropriate OM(s) for a specific clinical or research question. Two different groups previously reviewed the measurement properties of measures of gait function and performance in neuro-paediatrics. The first review focused on the reliability and responsiveness of outcomes of

gait function such as the Functional Mobility Score (FMS) and Gross Motor Function Measure (GMFM) [7]. Interestingly, in this review, little consideration was given to outcomes of gait quality (e.g. gait kinematics). Furthermore, these authors only reported on the measurement reliability and responsiveness with no explicit consideration of validity of the OMs. The second review by Rathinam and colleagues [8] critically appraised reliability and validity of measures of gait quality but only those derived from observational gait assessment (OGA) tools used in paediatrics. Furthermore, the methodological quality of the outcome measures was not assessed using a standardised checklist such as the Consensus-based Standards for the selection of health status Measurement Instruments (COSMIN) checklist [9]). As a result, it was not possible to assess the relative strength of the evidence provided to support their recommendations. A recent review [10] appraised the OMs of walking ability in CP using the COSMIN checklist, however this review focused only on OMs that are simple and quick to perform.

The COSMIN checklist, recommended for use in systematic reviews of measurement properties [9], has been used previously to explore the psychometric properties of gait function [7] and other OMs, such as balance, aerobic capacity and habitual physical activity in children with

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CP [11–13]. Considering the increasing number of studies reporting the results of gait analysis, ranging from visual observation scores to computerised three-dimensional gait analysis (3DGA), a review of the psychometric properties of outcomes of both gait quality (i.e. gait characteristics) and gait performance using a standardised quality checklist appears warranted. Therefore, the aim of this study was to evaluate the methodological quality and the strength of the evidence of studies that reported an evaluation of the psychometric properties of OMs of gait quality and walking performance in young people with CP.

2. Methods

2.1. Search strategies

The MEDLINE, CINAHL, PubMed and Scopus databases were searched using main search categories, which were 'cerebral palsy', 'gait', 'outcome measure' and 'measurement properties' up to 14th January 2016. For the PubMed database, the same search strategy with a published additional sensitive search and exclusion filter for measurement properties [14] was applied. The details of search strategies are provided in Appendix A in Supplementary material. Finally, the reference lists of all the primary identified studies were manually searched and examined for studies meeting the inclusion criteria.

2.2. Inclusion and exclusion criteria

The aim of this review was to explore and summarise the evidence presented in studies reporting the measurement properties of OMs for gait and walking performance in children and adolescents with CP. Only papers dealing with children and young adults (3 to 25 years) with Cerebral Palsy were included. This lower age limit was chosen for neurophysiological reasons and practical reasons (e.g. compliance/understanding). This review focused on five psychometric properties including reliability (test retest reliability, inter-rater reliability, and intra-rater reliability), measurement error, construct validity, criterion validity and responsiveness. The full list of study inclusion and exclusion criteria is shown in Table 1.

2.3. Study selection process

After removing duplicates, two reviewers (AZ and MvdL) independently screened the titles and abstracts of the studies resulting from the literature search based on the inclusion and exclusion criteria. In case of disagreement or uncertainty, the full paper was reviewed. A third reviewer (KJ or TM) was available if no consensus could be reached.

2.4. Quality assessment process

Full articles that met the inclusion criteria were independently rated by two reviewers (AZ as main and MvdL or KJ). In the case of disagreement, there was a discussion to reach consensus. Each study was rated to determine (i) the overall methodological quality of studies investigating specific psychometric properties and (ii) the quality of the psychometric properties.

2.5. Evaluation of overall methodological quality scores

To determine the methodological quality of the studies, the COSMIN checklist [9] was used. The COSMIN checklist consists of nine boxes (internal consistency, reliability, measurement error, content validity, structural validity, construct validity, cross-structural validity, criterion validity, and responsiveness) with 5–18 items each checking the methodological standards of the paper in terms of its design and statistical approach. Each item was scored on a four-point rating scale ('poor', 'fair', 'good' or 'excellent'. The overall methodological quality score was based on the lowest rating of any items ticked in any box.

In the original COSMIN criteria, developed to assess psychometric properties of self-reported questionnaires and studies with a sample size of less than 30 are given the methodological rating of 'poor'. It was anticipated that studies on the psychometric properties of gait quality and performance measures in young people with CP would often have less than 30 participants, and the application of the original criteria would exclude studies with otherwise good or excellent methodological quality. Consequently, in this review, we did not use the sample size item for the rating of any of the psychometric properties. Instead, sample size was accounted for at the best evidence synthesis stage. This approach, which was first described by Dobson et al. [15] and was agreed by the COSMIN developers, was subsequently adopted in several other COSMIN reviews of outcomes used in the CP population [7,10,11–13].

As the definitions and terminology of certain psychometric properties adopted by COSMIN may not always be similar to those used by authors of the articles reviewed, we applied the COSMIN taxonomy instead of the terms used in the articles. As recommended by COSMIN, small modifications can be made to each scoring system to suit the purpose of the review or characteristics of the outcome measures [16]. We therefore developed 'rules' within the COSMIN rating to minimise differences between reviewers in interpretations of checklist items. The items relating to 'missing items' were not scored if the outcome measure was not a questionnaire or a test battery as we regarded 'items' as questions in a questionnaire or parts of a test battery. With respect to the 'time interval appropriate' item, we regarded the time interval of two weeks or less to assess the test-retest reliability as appropriate. For questionnaires we regarded the minimum time interval for no recall as one week. Finally, in those studies assessing the validity of the observational gait analysis, three dimensional gait analysis was accepted as the gold standard.

2.6. Evaluation of the quality of the psychometric properties

The quality of the psychometric properties i.e. the strength of the evidence of the studies included in the review was assessed using the quality criteria developed by Terwee and colleagues [17], which have been subsequently slightly revised by the author [18] as shown in Table 2. These guidelines were developed to score the quality of studies in terms of their design, methods and outcome on the development and evaluation of the particular instruments. All OMs were rated with 'positive' (+), 'indeterminate' (?), 'negative' (−), depending on the results of the studies. If no information was available, a zero ('0') was recorded.

Of note, some of the OMs, for example Edinburgh Visual Gait Score (EVGS), may be given two ratings (i.e. 'positive' and 'negative'). This is because these OMs have more than one component for which the strength of evidence can be rated, for example 'knee angle at initial contact' and 'hip angle in stance'. If both 'negative' and 'positive' ratings are given for the different components, the evidence derived from these OMs will be rated as 'conflicting' in the synthesis of best evidence, described below.

Table 1
Inclusion and exclusion criteria of the systematic review.

Inclusion criteria
- Study population: children, adolescents and young adults (3–25) with CP
- Studies reporting on psychometric properties (reliability, validity and responsiveness) of OMs of gait quality or walking performance.
- Studies published in English
- Full text original article

Exclusion criteria
- Studies that validate translated versions of the OMs
- Studies in which the primary aim is not to assess psychometric properties (e.g. intervention studies or cohort studies exploring correlations between OMs)