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REVIEW ARTICLE (META-ANALYSIS)

Past and Current Use of Walking Measures for Children With Spina Bifida: A Systematic Review

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Abstract

Objectives: To describe walking measurement in children with spina bifida and to identify patterns in the use of walking measures in this population.

Data Sources: Seven medical databases—Medline, PubMed, Embase, Scopus, Web of Science, CINAHL, and AMED—were searched from the earliest known record until March 11, 2014. Search terms encompassed 3 themes: (1) children; (2) spina bifida; and (3) walking.

Study Selection: Articles were included if participants were children with spina bifida aged 1 to 17 years and if walking was measured. Articles were excluded if the assessment was restricted to kinematic, kinetic, or electromyographic analysis of walking. A total of 1751 abstracts were screened by 2 authors independently, and 109 articles were included in this review.

Data Extraction: Data were extracted using standardized forms. Extracted data included study and participant characteristics and details about the walking measures used, including psychometric properties. Two authors evaluated the methodological quality of articles using a previously published framework that considers sampling method, study design, and psychometric properties of the measures used.

Data Synthesis: Nineteen walking measures were identified. Ordinal-level rating scales (eg, Hoffer Functional Ambulation Scale) were most commonly used (57% of articles), followed by ratio-level, spatiotemporal measures, such as walking speed (18% of articles). Walking was measured for various reasons relevant to multiple health care disciplines. A machine learning analysis was used to identify patterns in the use of walking measures. The learned classifier predicted whether a spatiotemporal measure was used with 77.1% accuracy. A trend to use spatiotemporal measures in older children and those with lumbar and sacral spinal lesions was identified. Most articles were prospective studies that used samples of convenience and unblinded assessors. Few articles evaluated or considered the psychometric properties of the walking measures used.

Conclusions: Despite a demonstrated need to measure walking in children with spina bifida, few valid, reliable, and responsive measures have been established for this population.

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Spina bifida is one of the most common congenital birth defects, with a current prevalence of about 0.3 to 0.4 per 1000 births in Canada[1] and the United States.[2] With this condition, a neural tube defect results in damage to the spinal cord, brain, and/or meninges. Walking is a challenge for many children with spina bifida. More than half of those with neurological lesions at or below the thoracic level achieve walking at some point in their childhood, with the walking rate increasing as the lesion level decreases.[3-6] As a result of their sensorimotor impairments, many children with spina bifida walk with abnormal gait patterns. This significantly increases the energy cost of walking and reduces their walking endurance.[7-11] A notable proportion of children with spina bifida lose the ability to walk as they age.[4,5]

Achieving and/or improving the ability to walk is an important goal for many children with spina bifida and their families because walking enables greater participation in daily activities and

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recreation and contributes positively to quality of life. As a result, walking is often a focus of the medical management and rehabilitation of those children who have the potential to walk.[3] Considerable effort is spent investigating therapeutic approaches that may lead to improved walking outcomes for children with spina bifida, such as walking training,[11,12] surgical procedures,[13,14] neural prostheses,[15] and orthoses.[16-18]

For researchers and clinicians to accurately evaluate the effects of an intervention on walking and to follow a child's walking ability over time, valid, reliable and responsive measures of walking must be used. Laboratory-based assessments involving sophisticated motion analysis systems can measure kinematic gait characteristics in children with spina bifida.[16,19] Because most clinicians do not have access to such systems, walking measures that can be administered in a clinical setting are required. Furthermore, these measures must have good psychometric properties (ie, validity, reliability, and responsiveness) to be useful. To date, however, there is little guidance for clinicians and researchers as to what walking measures are useful for children with spina bifida.

As a first step toward providing guidance on how to assess walking in children with spina bifida, we performed a systematic review and critical evaluation of the literature. Our objectives were to (1) identify the walking measures used in clinical settings for children with spina bifida; (2) describe the circumstances under which these measures were used; and (3) evaluate these measures with respect to their psychometric properties reported in the literature.

Methods

A systematic review was performed following the Preferred Reporting Items for Systematic Reviews and Meta-Analysis guidelines.[20] There is no protocol for this review. To form the review question, a modified population, interventions or indicators, comparators, outcomes, and a study design framework was used. The population was children with spina bifida aged 1 to 17 years. The indicator was a measure of walking that could be used in a clinical setting. There were no comparators. The outcome of interest was walking ability, and there were no restrictions on the study design.

Search strategy

A literature search was completed in consultation with an information specialist at the University of Saskatchewan. Seven databases—Medline, PubMed, Embase, Scopus, Web of Science, CINAHL, and AMED—were searched from the earliest known record until March 11, 2014. The search terms included keywords and controlled vocabulary (where applicable) and focused on the following themes: (1) children; (2) spina bifida; and (3) walking (see appendix 1 for search example). No restrictions were placed on the language, date, or type of publication.

Duplicate records were removed using the research management tool REFworks.[a] All abstracts were then independently reviewed by 2 authors (D.L.B. and K.E.M.) to select the articles for full-text review. The inclusion criteria were as follows: (1) Study participants were children with spina bifida (diagnoses included spinal dysraphism, neural tube defects, meningomyelocele, congenital or nontraumatic spinal cord injuries, spina bifida, myelomeningocele, lipomyelomeningocele, lipomeningocele, and spinal cord malformation). (2) Functional walking capacity, defined as "the ability to ambulate daily using reciprocal steps overground for short distances" with or without assistive devices,[21] was assessed or reported in some way. This definition was chosen because it reflects walking that is clinically relevant.

Exclusion criteria included: (1) Studies in which the participants were exclusively infants (ie, younger than 1y) or exclusively adults (ie, 18y and older). (2) Studies that did not include any individuals with spina bifida. (3) Studies that did not examine walking function (eg, examined gross motor function or wheeled mobility). (4) Studies in which walking was evaluated only through kinematic, kinetic, or electromyographic analysis (ie, measures not routinely used in clinical settings). (5) Studies in which walking ability was given a dichotomous classification of ambulatory or nonambulatory. (6) Animal studies, modeling studies, narrative review articles, conference proceedings, editorials, or letters to the editor.

Data extraction

The following data were extracted from the included full-text articles using a standard data extraction form: patient population tested, type of neural tube defect, level of lesion, age of participants, number of child participants with spina bifida, method of participant recruitment, method of walking assessment, purpose of walking assessment, psychometric properties of the walking measures used, results of walking measurement, and type of study. For articles that were not in English, individuals proficient in the language and/or Google Translate assisted with translation, and in one case the author of the article was contacted for the information in English.

Methodological quality of the included articles was assessed by adapting the methods of Dobson et al.[22] Nine methodological characteristics were assessed, such as the sampling method and psychometric properties of the walking measures used (table 1). These categories were rated as adequate/inadequate or stated/not stated depending on the question. Methodological quality was evaluated by a pair of reviewers independently and the final decisions reached through consensus.

Data synthesis

Descriptive summaries

Extracted data from all included articles were summarized to describe the use of walking measures in children with spina bifida. Because this is a descriptive review, there is no principal summary measure. The following analyses were completed: (1) The number of times a given measure was used was counted and expressed as a percentage of the total number of times a walking measure was used in the included articles. (2) The number of times a given study purpose was reported was counted. For example, possible study purposes included investigating the relation between walking ability and another variable or examining the effects of a surgical or orthotic intervention. (3) For each walking measure identified, information concerning its validity, reliability, responsiveness, and/or interpretability (eg, interpretation of scores relative to normative data, cutoff values) was aggregated.

Statistical machine learning analysis

To further describe how walking measures are used in children with spina bifida, we determined whether there was any pattern to the selection of a measure. For example, are categorical rating scales chosen because of participant age and/or because of the range of lesion levels? If such patterns exist, they would inform future measurement guidelines. The included studies were