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Review

Systematic review on gait classifications in children with cerebral palsy: An update

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ABSTRACT

Background: Gait classification systems (GCSs) aim to aid clinicians and researchers in categorizing the gait of pathological populations, with the intent to improve the communication between them, to support treatment planning and enable the evaluation of patients over time. Throughout the years, various GCSs have been defined for children with cerebral palsy (CP), which were first summarized in a systematic review published in 2007.
Research question: The current systematic review aimed to: a) identify GCSs that have been more recently developed, b) appraise their methodological quality and c) specify the most commonly used multiple joint gait patterns for children with CP reported in literature.
Methods: Four databases (Medline, EMBASE, CINAHL, Web of Science) were searched until July 2017. Several forms of validity and the reliability of these studies were assessed according to the principles of the consensus-based standards for the selection of health measurement instruments checklist or criteria defined in the original review. All published GCSs were also scrutinized in order to identify multiple joint patterns that have reached a predefined level of consensus.
Results: Thirty-six studies were considered in this review, 15 of them being GCSs that were not included in the original review. The validity, reliability and clinical applicability of all GCSs was reported, including 3 studies from the original review. Six multiple joint patterns for children with CP reached a consensus in literature.
Conclusion: Since the previous review, obvious progress has been made in the field of GCSs for CP, resulting in improved methodological quality of the majority of published GCSs. This encouraged the applicability of GCSs in clinical or research settings. The six reliable, valid and commonly used multiple joint patterns, emerging from this systematic review, may aid clinical and research applications and create a common language among healthcare providers.

1. Introduction

Cerebral palsy (CP) constitutes the most prevalent cause leading to physical impairment during childhood [1]. With approximately 70% of CP children being ambulatory [2], gait analysis has become an indispensable tool in the assessment of their gait deviations. Specifically, three dimensional gait analysis (3DGA) is regarded as the "golden standard" in CP gait evaluation [3,4] and, consequently, several gait classification systems (GCSs) have been formed, based on data acquired through 3DGA [5–20]. GCSs have been reported to aid in clinical decision making [5,17], in improving communication among healthcare providers [5,17,20] and in comparing gait between or within specific patients [20]. On the other hand, it has been argued that a "hard" gait pattern assignment may over-simplify the real gait deviations of patients [13,21]. Furthermore, GCSs may include the description of deviations occurring in one or multiple lower limb joints simultaneously (single or multiple joint patterns, respectively), thus challenging the establishment of commonly accepted definitions in the clinical community of CP or the distinction between GCSs and gait patterns.

In 2007, Dobson et al. published a systematic review on eighteen previously published GCSs for children with CP, including studies as recent as March 2005 [5]. The included GCSs were divided into either

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qualitatively or quantitatively constructed ones, based on whether they were formed by clinical expertise or objectively measured data, respectively. The study concluded that several challenges with regards to classification construction, validity, patient inclusion, and reliability remained to be resolved. Moreover, methodological limitations of previously published GCSs hindered their applicability in clinical practice or research [5].

The aim of the current study was three-fold. Firstly, to review the study characteristics of GCSs published since March 2005. Secondly, to include all reliability or validation studies, so as to form a detailed overview of the strengths or limitations of all available GCSs. Thirdly, to identify and describe the most commonly used multiple joint patterns described in literature, providing clinicians with a comprehensive inspection of the most commonly described multiple joint patterns.

2. Method

2.1. Search strategy

Two reviewers (AN and EP) conducted an extensive systematic search for all publications up to July 2017, following the guidelines of the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) [22]. Four online databases were searched, namely: Pubmed, Embase, Cinahl, and Web of Science. Key words included the following search terms: (1) cerebral palsy or diplegia or hemiplegia or quadriplegia or Little's Disease, (2) gait analysis or locomotion or walking or gait, (3) classification or taxonomy or cluster analysis or gait pattern or clinical classification or pattern recognition or classification algorithm or machine learning or neural network or connectionist model or support vector machine or fuzzy logic. Subsequently, these terms were combined into (1) AND (2) AND (3). "All fields", MeSH terms and wild card symbols were used so as to ensure all key word variations. When possible, relevant filters were implemented such as humans, age range, and language (English). Finally, references of relevant articles were screened to ensure that additional records that might have been accidentally overlooked by the electronic searches would be included (snowballing method).

2.2. Inclusion criteria

After removing duplicate studies, two reviewers (EP and AN or EP and IV) independently screened each reference based on title and abstract. Eligibility of full-text papers was assessed based on the following inclusion criteria: a) studies focused primarily on children with spastic CP (aged between 0 and 18 years, i.e. minimally 80% of the study population being patients with spastic CP under 18 years of age), b) gait assessment (kinematic, kinetic, spatiotemporal or electromyographic parameters), c) allocation to groups or distinct categories defined by gait analysis, d) published after March 2005 or not included in the review by Dobson et al. [5]. Original GCSs as well as subsequent validity or reliability studies of these classifications were included. Studies were excluded if a) their primary focus was on children with dystonia or ataxia, b) no full paper was available, c) the main focus was on gait indices, summary measures or functional classifications, d) in case of reviews, protocols or case series and, finally, e) the study was not available in English.

2.3. Data extraction

Two independent reviewers (EP and IV) extracted important study characteristics of all new GCSs. They used the data extraction form that was created and applied during the systematic review by Dobson et al. [5], so as to maintain as much consistency as possible between the two reviews. The characteristic "reliability test results" was not retained due to the currently applied investigation of reliability.

In case of disagreement or lack of clarity, a third reviewer (KD) was consulted in order to reach a consensus. Extracted information consisted of the type of patients that were considered in each study, which included their anatomical distribution, along with typically developing (TD) subjects, if they were also taken into consideration during the development of each GCS. Per study, also the number of included subjects per subject type, their ages, the applied measurement tools and the investigated gait variables were recorded. Furthermore, the type of scale each GCS was built upon and the number of classes per GCS were also part of the extraction form. Finally, the anatomical levels that were considered in each study and the applied overall approach to construct the classification (quantitative or qualitative) were reported.

2.4. Psychometric properties

The principles of the COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) checklist were applied to thoroughly review the quality of the scrutinized GCSs [23–25]. This checklist allows researchers to investigate the methodological quality of each study, by establishing quality scores in each separate measurement property, such as reliability and various types of validity. Each measurement property is represented in a box with several items, which allows for their individual scoring. Researchers identify which boxes and items apply to their study according to their research questions [23–25]. A 4-point scoring system has been established with scores varying between "excellent", "good", "fair" or "poor", the lowest score in each box defining the final score of the total box [25].

The psychometric properties were investigated for all studies that were identified in this review, as well as for the GCSs previously reviewed by Dobson et al. [5], including subsequent validity or reliability studies of previously published GCSs up to July 2017. All information was extracted by EP and IV and KD was consulted in case of disagreement. Previously, Dobson et al. had synthesized a customized quality assessment checklist due to the lack of published standardized or validated ones [5]. In the current review, some of the characteristics of this customized quality assessment checklist were maintained, on one hand to ensure some extent of homogeneity and comparability between the two reviews, and on the other hand because these parameters were considered to be indispensable when describing GCSs. These characteristics included the quality of the classification construction and more specifically the planes of motion, the description of the construction method (i.e. detailed explanation about the entire construction process) and, finally, the individual characteristics of the classification groups, enabling their precise definition and the differentiation between groups. The classification construction was subsequently given a total score, in line with the COSMIN principles.

For the remaining aspects of validity, as well as for the reliability of GCSs, the COSMIN checklist was adjusted by the reviewers, such that all relevant boxes and items would be investigated (Supplementary tables 1 and 2). In addition, the used sample in each study was reported and judged relatively to the number of the defined patterns that stemmed from this particular sample size, as well as the generalizability of these results. The latter refers to whether the employed sample was described in detail and refers to the following criteria: age, sex, disease characteristics, setting of study and patient selection method. Moreover, 6 additional types of validity where identified as important for this study:

1) Validation of a GCS against a different dataset (than the one where the initial GCS was based on),
2) Automatic classification algorithm (with possible comparison to clinicians' classification),
3) Validation against a previous GCS (direct comparison to patterns of a previous GCS),
4) Face validity (comparison of kinematic data to video reports or categorization of patients based on consensus or expert opinion),
5) Validation against a valid clinical scale (comparison of gait patterns to scales such as the modified Ashworth scale (MAS) [26] for

spasticity) and
6) Responsiveness (classification of the same patients in at least two instances to establish whether their gait patterns have changed over time or after treatment).

In the previous review, the utility of each GCS was reported with a criterion of whether the authors of each study referred to the utility of their study or not [5]. In the present study, the introduction of the term "clinical applicability" was considered more relevant, for which specific criteria were formulated (Supplementary table 1). Moreover, the score for every original GCS was based on the description of clinical applicability in that publication as well as in any subsequent reliability or validity studies that applied the original GCS. The original studies were thereby labelled as "main". Similar to the overall COSMIN scale, the lowest item score indicated the final score.

Regarding the reliability of these GCSs, studies were analyzed based on the recruited sample size, whether the raters were blind to patient specific characteristics, whether the intra-rater reliability was also tested and the applied statistics.

2.5. Commonly used multiple joint patterns

All the available GCSs identified in the review of Dobson et al. [5] and in the current update were scrutinized so as to identify the multiple joint patterns that stem from these GCSs and are most prevalently reported in literature, or the patterns upon which several researchers agree. A panel of 4 reviewers (KD, EO, AVC and EP) defined the following criteria for these commonly used patterns where agreement has been reached by the scientific community: i) their classification groups were adequately defined, according to Table 2 and Dobson et al. [5], ii) the original GCSs displayed either an "excellent" or a "good" clinical applicability (Table 2 and Supplementary table 1), and iii) at least 2 independent research groups have cited the derived patterns in their publications since the original introduction of the GCS. These citations should refer to the GCS as a whole or to some of its defined patterns and not only to the techniques or methodologies applied to derive them. Similar in- and exclusion criteria regarding abstracts, commentaries, full paper availability and articles published in English, as described in section 2.2 were applied. Self-citations by the same researchers or research groups were not considered. In order to create a summary of all multiple joint patterns that displayed a relevant consensus, each specific multiple joint pattern of a GCS was judged based on: i) whether 3 or more GCSs have also used the same multiple joint pattern name or its characteristics and/or ii) reliability studies have supported the definition of this particular pattern. Subtle differences in definitions among the various studies were considered possible, however, the most prevalent description of each term, with the best psychometric properties has been considered as the main one.

3. Results

3.1. Search strategy yield

The electronic search resulted in 5045 studies and, via the snowballing method, 18 references were additionally identified. After the removal of duplicates, 4057 records were screened, from which 3916 were excluded based on title and abstract alone. Eligibility of 141 full-text articles was assessed according to the inclusion/exclusion criteria mentioned in 2.2. In the end, 33 studies were considered to be eligible for inclusion in this study (Fig. 1). Three additional studies [27–29] – previously included in [5] – were also considered in the current update due to the fact that either reliability or validity studies had been published during the period of search, further investigating the psychometric properties of these GCSs. This selection procedure finally resulted in 36 included studies.

3.2. Study characteristics of new GCSs

Out of the 36 studies, 15 were newly introduced GCSs, with quite heterogeneous participant characteristics and sample sizes (Table 1). Seven studies incorporated TD children as control subjects [7–10,12,13,18], 5 studies focused only on patients with spastic diplegia [7,9,11,12,19], whereas 2 studies did not specify how many of the included patients presented with a spastic motor disorder or another type [13,19]. The sample size varied widely, from 19 children [8] to approximately 1537 children with CP [13], while 3 GCSs, which were formed based on expert opinions, did not use concrete patient data for the development of their definitions [11,17,20]. The ages of the patients that were used were also diverse (2 to 30 years of age) [9,16].

The majority of the studies used or established rules based on the analysis of data acquired through 3DGA (n = 12) [6,7,9,10,12–16,18–20]. Other methods included the interpretation of surface electromyography (EMG) [8], video recordings [11], or expert opinion [17]. In all but 4 studies, the description of kinematic parameters was the primary focus [6,10–13,15–20] and in 2 studies, only spatiotemporal parameters were taken into consideration [7,9].

Ten GCSs were constructed on a nominal scale [6,10–12,15–20], while 2 studies used mixed features of both nominal and ordinal scales [7,14]. GCSs defined multiple groups, ranging from 4 multiple joint patterns [11] to 71 possible combinations of single joint deviations [19], while two GCSs investigated the deviations on a single joint level (one joint [8] or one joint at a time [20]). Distal deviations, i.e. at the ankle or foot, were most frequently classified (n = 11) [6,8,10,11,13,15–20]. The knee and the hip joint motion was described in 9 out of the 15 studies [6,10–13,16,17,19,20] and the motion of the pelvis, the trunk or the upper limb during gait was described in 6 [11–13,17,19,20], 2 [11,17] and 1 [11] GCSs, respectively. Lastly, 9 GCSs were constructed in a quantitative way [7–10,12,13,15,16,18], employing mostly clustering techniques (n = 6). The remaining GCSs included multiple or single joint patterns that were constructed in a qualitative way, where patterns were defined by a certain expert decision making process. In those cases, decisions were based either on clinical experience and expertise of a (local) group of researchers [6,11,14,17,19] or a survey in an international group of experts [20].

3.3. Validity

Different levels of validity were evaluated across all studies (Table 2). Detailed information on the analytical sub-scores, which compile the total COSMIN score for each category, can be found in the Supplementary tables 3a-c.

The studies in which GCSs were originally introduced, were judged for their construction method (i.e. explanation about the entire construction process - Supplementary table 1). Eight GCSs were judged as displaying an "excellent" classification construction method [7–9,11,14,16,18,20], while three were labelled as "good" because they only described gait patterns across one [15,19] or two [17] planes of motion. GCSs were characterized as "fair", because they either did not display an adequate explanation or detailed description of the classification groups that were formed [10,12,13], or they did not provide any information on how their classes were constructed [6,27,29]. Only one GCS was judged as displaying a "poor" classification construction [28].

The sample size was characterized by indicating the number of patients / limbs / trials / strides measured relative to the number of derived patterns (Supplementary table 1). Most of the 36 included studies referred to the number of patients (either CP or with specific, topographical description, namely spastic hemiplegia or diplegia, n = 23) [6,7,9,11,12,18,27,29–44]. In the current review, 25 studies had either an "excellent" or a "good" sample size definition [6–11,13–16,27,29,31–36,38–42,45,46], whereas the remaining studies were labelled as "fair" [18,19,30,44], "poor" [12,37,43,47] or "non-applicable" [17,20,28]. Nineteen studies displayed an "excellent"

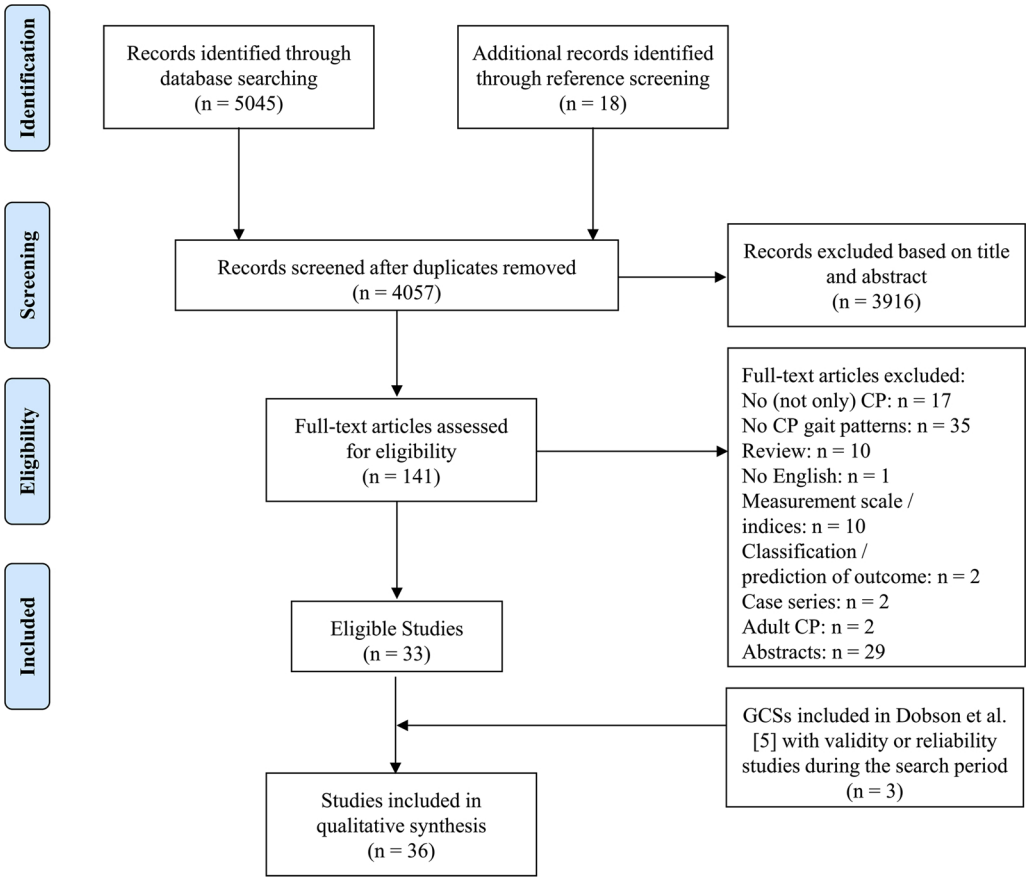

Fig. 1. Flowchart of the search strategy.

score for generalizability (5 out of 5 items) [6,7,11,16,18,27,30–34,38,40–45,47]. In three studies this category was "non-applicable" [17,20,28], 3 studies had a quite low generalizability (2 out of 5 items) [12,13,39] and the remaining ones addressed either 4 [10,14,15,19,29,36] or 3 [8,9,34,37,46] of the 5 items.

From the additional types of validity, 20 studies validated a GCS "against a different dataset" from the one used in the original GCS [7,9,11–13,15,16,19,31–34,36,39–41,44–47], with 6 of the studies having scored "excellent" [7,32,34,40,44,45]. The least reported form of validity was "against a previous GCS" (n = 1) [10], while "responsiveness" was tested in 4 studies [7,9,29,31] and validity 'against a valid clinical scale" was tested in 9 studies [13,16,31,33,36,39–41,44]. Eight studies focused on either of the remaining validity forms, namely: the development of an "automatic classification algorithm" [8,12,15,18,34,39,46] and "face validity" [17,20,30,33,38,41,42,47]. An extra validation sample was used in 7 studies, with the sample varying from 2 patients (representing 4 limbs) [9] to 1152 limbs of CP children [39]. Finally, four studies did not employ any form of extra validation [6,14,27,28].

3.4. Clinical applicability and reliability

An overview of the clinical applicability of all reviewed studies is shown in Table 2, while the utility scored according to [5] is displayed in Supplementary table 3c. The GCSs that were constructed in a quantitative way, displayed a "fair" clinical applicability [7,9,10,12–14,16,18]. Eleven studies were judged as demonstrating "excellent" [6,11,19,27–29,40,41,43–45] and the remaining ones a "good" clinical applicability [8,15,17,20,30–39,42,46,47]. The final scores for all the "main" studies (i.e. those where additional reliability or validity studies were also reviewed) [11,20,27–29], except for one study [6], were characterized as displaying a "good" clinical applicability.

Reliability was examined in 6 studies [29,30,37,38,42,43], of which only one had an "excellent" score [43] (Table 3). Three studies assessed only inter-rater reliability [30,38,42], with one of them not using a weighted kappa for the reliability evaluation, despite the fact that it concerned a GCS built on an ordinal scale [38]. Finally, the last two studies [29,37] were judged as "fair" due to the small sample size that was used to investigate the reliability of the GCS in question.

3.5. Agreement on commonly used multiple joint patterns

All GCS were inspected according to the criteria specified in 2.5. The number of total citations, along with the number of different research groups that cited these GCSs are reported in Supplementary table 4. Nine GCSs [27,29,48–54], incorporated in [5], had an "adequate" description of classification groups. However, 5 of them were constructed based only on quantitative approaches [49,51–54], hence, their clinical applicability was characterized as "fair", in compliance to Supplementary table 1. These GCSs were thus not further considered in this investigation. The clinical applicability of the studies of Simon et al. [48] and Hullin et al. [50] was judged as "good" and "excellent", respectively. The applicability of all other studies can be found in Table 2. Five studies were excluded because they did not satisfy the criterion of having at least 2 citations by other research groups [8,11,15,17,20].

Consensus was reached for six multiple joint patterns, namely "genu recurvatum", "drop foot", "true equinus", "jump gait", "apparent equinus" and "crouch gait" (Table 4, Fig. 2). Table 4 contains the name of each pattern, specifies the authors that originally introduced or established each term, gives a description of the patterns' characteristics, reports other GCSs that have referred to the specific patterns or their

Table 1
Identified studies[a] on classification of gait deviations in children with cerebral palsy – study characteristics.

Study	Subjects Type	Sample size per type	Age range (mean ± STD)	Measurement tool used	Type of gait variables used	Scale type	Number of groups	Anatomical levels classified	Method of classification construction
Wren et al. [6]	SD, SQ, SH, ST, athetosis	291, 76, 115, 5, 5	3.1-24.4 (9.6 ± 4y)	3DGA (Vicon)	Kinematic	Nominal	14	H, K, A, F, HF, FF	Qualitative pattern recognition using quantitative data
Vaughan et al. [7]	SD, TD	78, 669	5-20 (10y8m), 5-98	3DGA (Vicon)	Leg length, stride length, cadence	Mixed nominal[e]	5	N / A	Quantitative using fuzzy clustering
Schmidt-Rohlfing et al. [8]	SD, SH, TD	15, 4, 50[d]	3-12 (7.8y)	Surface EMG	EMG of TA, G, S	Ordinal	3	A	Quantitative using a fuzzy expert system based on expertise
Xu et al. [9]	SD, TD	88, 68	2-20 (9.9y), 2-13 (7.1y)	3DGA (Vicon)	Stride length, cadence, leg length, age	Ratio	5	N / A	Quantitative using k-means and hierarchical clustering
Toro et al. [10]	SH, SD, SQ, SM, Dystonia, Ataxia, TD	25, 24, 4, 1, 1, 1, 11	5-16 (9y)	3DGA (CAST)	Kinematic	Nominal	13	H, K, A	Quantitative using hierarchical cluster analysis and incorporation of qualitative clinical judgement
Ferrari et al. [11]	SD	N/A	N/A	Video	Kinematic	Nominal	4	Head, UL, T, P, H, K, A, F	Qualitative pattern recognition
Carriero et al. [12]	SD, TD	20, 20	5-12 (9y), 6-13 (8.7y)	3DGA (Vicon)	Kinematic, sp-t	Nominal	9	P, H, K	Quantitative using PCA and fuzzy c-mean cluster analysis
Rozumalski and Schwartz [13]	CP[b], TD	71.2% of 2159	N/A	3DGA	Kinematic	Ordinal	5	P, H, K, A	Quantitative using k-means cluster analysis
Williams et al. [14]	SD, SH	59, 15	7-19 (13.4y)	3DGA (Vicon)	Kinetic	Mixed ordinalf	8	N / A	Qualitative pattern recognition using quantitative data
Van Gestel et al. [15]	SH, SD, ST	58, 80, 1	5-12 (8y10m ± 2y2m)	3DGA (Vicon)	Kinematic, kinetic	Nominal	19	K, A	Quantitative using probabilistic classification through BN based on experts' opinions
Bonnefoy-Mazure et al. [16]	SH, SD	40, 82	3-30 (14.2 ± 7.5)	3DGA (Vicon)	Kinematic, sp-t	Nominal	4	H, K, A	Quantitative using fuzzy window coding and MCA
Davids and Bagley [17]	CP	N/A	N/A	Expert opinion	Kinematic, kinetic	Nominal	16	T, P, H, K, A, F	Qualitative pattern recognition
Krzak et al. [18]	SH, TD	24, 20	12.0 ± 4.1, 11.8 ± 2.7	3DGA (Vicon)	Kinematic, walking speed, age	Nominal	5	HF, FF	Quantitative using PCA and k-means cluster analysis
Simon et al. [19]	SD	188	4.25-25 (11.7 ± 0.2)	3DGA (Vicon), videotapes	Kinematic	Nominal	71	P, H, A, F	Qualitative pattern recognition using quantitative data
Nieuwenhuys et al. [20]	sCP[c]	N/A	N/A	Expert consensus, 3DGA	Kinematic, kinetic	Nominal	49	P, H, K, A, F	Qualitative pattern recognition using quantitative data

Abbreviations: SD: spastic diplegia; SQ: spastic quadriplegia; SH: spastic hemiplegia; ST: spastic triplegia; TD: Typically developping children; (s)CP: (spastic) cerebral palsy; SH: spastic hemiplegia; SM: monoplegia; N/A: non-applicable; STD: standard deviation; y: years; m: months; 3DGA: three dimensional gait analysis; EMG: electromyography; TA: tibialis anterior muscle; G: gastrocnemius muscle; S: soleus muscle; sp-t: spatiotemporal; H: hip; K: knee; A: ankle; F: foot; HF: hindfoot; FF: forefoot; UL: upper limb; T: trunk; P: pelvis; PCA: principal component analysis; MCA: multiple correspondence analysis; BN: Bayesian networks.
[a]Included studies were identified according to the outlined search strategy in Section 2.1 of this article.
[b]CP with excessive knee flexion at initial contact (> 20°).
[c]Spastic CP (without foot deformities).
[d]50 healthy gait cycles, sample size not mentioned.
[e]Mixed nominal: mostly resembles a nomial scale but also has some characteristics of an ordinal one.
[f]Mixed ordinal: mostly resembles an ordinal scale but not all classes are relative to each other.

Table 2
Validity, utility and clinical applicability of gait classification systems in children with cerebral palsy, scored according to the adapted version of the COSMIN scoring criteria [25].

Study	Validity										Clinical applicability	
	Classification construction				Used sample	Sample Size	Generali-zability	Other validity forms		Extra validation sample		
	Planes of motion	Construction method	Classification groups	Cosmin score				Form	Cosmin score		Individual studies	Main study
Winters et al. [27]	1	Not stated	Adequate	Fair	46 SH	Good	5/5	/			Excellent	Good
Dobson et al. [30]					34 SH	Fair	5/5	4	Fair		Good	
Riad et al. [31]					112 SH	Excellent	5/5	1	Good		Good	
								5	Fair			
								6	Good			
McDowell et al. [32]					91 SH	Good	5/5	1	Excellent		Good	
Dobson et al. [33]					58 SH	Good	5/5	1	Good		Good	
								4	Fair			
								5	Excellent			
Agostini et al. [34]					38 SH	Excellent	5/5	1	Excellent		Good	
Padilla et al. [35]					83 SH	Good	3/5	2	Fair	25% of 83 patients	Good	
Sutherland and Davids [28]	1	Not stated	Partial	Poor	N/A	N/A	N/A	/			Excellent	Good
de Morais Filho et al. [36]					1805 SD	Excellent	4/5	1	Good		Good	
								5	Good			
Rodda et al. [29]	1	Not stated	Adequate	Fair	174 SD	Excellent	4/5	6	Fair		Excellent	Good
Stott et al. [37]					20 SD	Poor	3/5	/			Good	
Kim et al. [38]					91 SD	Good	5/5	4	Good		Good	
Sangeux et al. [39]					100 SD	Excellent	2/5	1	Fair	1552 limbs	Good	
								2	Fair			
								5	Fair			
Wren et al. [6]	3	Not stated	Adequate	Fair	492 CP	Excellent	5/5	/			Excellent	Excellent
Rethlefsen et al. [40]					1005 CP	Excellent	5/5	1	Excellent		Excellent	
								5	Excellent			
Vaughan et al. [7]	N/A	Adequate	Adequate	Excellent	78 SD	Good	5/5	1	Excellent	4 patients	Fair	
								6	Good			
Schmidt-Rohlfing et al. [8]	N/A	Adequate	Adequate	Excellent	38 limbs	Excellent	3/5	2	Good		Good	
Xu et al. [9]	N/A	Adequate	Adequate	Excellent	88 SD	Excellent	3/5	1	Fair	2 patients	Fair	
								6	Fair			
Toro et al. [10]	1	Adequate	Partial	Fair	132 limbs	Good	4/5	3	Good		Fair	
Ferrari et al. [11]	3	Adequate	Adequate	Excellent	213 SD	Excellent	5/5	1	Good		Excellent	Good
Cioni et al. [41]					213 SD	Excellent	5/5	1	Good		Excellent	
								4	Fair			
								5	Good			
Pascale et al. [42]					50 SD	Good	5/5	4	Excellent		Good	
Benedetti et al. [47]					36 limbs	Poor	5/5	1	Fair		Good	
								4	Good			
Carriero et al. [12]	3	Adequate	Partial	Fair	20 SD	Poor	2/5	1	Fair	27 patients	Fair	
								2	Fair			
Rozumalski and Schwartz [13]	1	Adequate	Partial	Fair	2956 strides	Excellent	2/5	1	Fair	83 normal subjects	Fair	
								2	Fair			
								5	Fair			
Williams et al. [14]	N/A	Adequate	Adequate	Excellent	133 trials	Good	4/5	/			Fair	
Van Gestel et al. [15]	1	Adequate	Adequate	Good	651 trials	Good	4/5	1	Good	20% of data	Good	
								2	Good			
Bonnefoy-Mazure et al. [16]	3	Adequate	Adequate	Excellent	204 limbs	Excellent	5/5	1	Good		Fair	
								5	Excellent			
Davids and Bagley [17]	2	Adequate	Adequate	Good	N/A	N/A	N/A	4	Excellent		Good	

(*continued on next page*)

Table 2 (*continued*)

Study	Validity Classification construction			Cosmin score	Used sample	Sample Size	Generalizability	Other validity forms		Extra validation sample	Clinical applicability	
	Planes of motion	Construction method	Classification groups					Form	Cosmin score		Individual studies	Main study
Krzak et al. [18]	3	Adequate	Adequate	Excellent	24 SH, 20 TD	Fair	5/5	2	Excellent		Fair	
Simon et al. [19]	1	Adequate	Adequate	Good	376 limbs	Fair	4/5	1	Fair		Excellent	
Nieuwenhuys et al. [20]	3	Adequate	Adequate	Excellent	N/A	N/A	N/A	4	Excellent		Good	Good
Nieuwenhuys et al. [45]					1719 trials, 154 trials TD	Good	5/5	1	Excellent		Excellent	
Nieuwenhuys et al. [43]					82 CP	Poor	5/5	/			Excellent	
Nieuwenhuys et al. [44]					286 CP	Fair	5/5	1	Excellent		Excellent	
								5	Excellent			
De Laet et al. [46]					1719 trials	Good	3/5	1	Good		Good	
								2	Good			

In bold: original studies. The adapted COSMIN scoring criteria [25] can be found in Supplementary table 1.
Other forms of validity: 1) Validation of a GCS against a different dataset; 2) Automatic algorithm in comparison to clinicians' classification; 3) Validation against a previous GCS; 4) Face validity; 5) Validation against a valid clinical scale; and 6) Responsiveness.
Abbreviations: N/A: non-applicable; SH: spastic hemiplegia; SD: spastic diplegia; CP: cerebral palsy; TD: typically developing children.

characteristics and indicates whether these patterns have been tested for reliability. Fig. 2 offers a schematic representation of the most characteristic deviations during a gait cycle per multiple joint pattern. Four of these multiple joint patterns comprise the GCS published by Rodda et al. [29]. These patterns have been tested for reliability and several forms of validity, and their definitions are frequently adopted in other GCSs [10–13,16,17,27–29,50,55–57]. The pattern "apparent equinus" is the only one of the 4 that has limited reliability or validity [37–39], but its description has been used in other GCSs [11,17,50]. In addition, the patterns "genu recurvatum" and "drop foot" were considered, because their definitions dictate that a pathological motion in 1 or 2 joints is accompanied by minor gait disturbances in the remaining lower limb joints [27,48]. Specifically, the former refers to knee motion in recurvatum with plantar flexion or decreased dorsiflexion motion [48]. This is similar to the "true equinus" pattern defined by Rodda et al., however, according to Rodda et al., an ankle equinus motion is the main deviation and it may also be combined with a normal knee motion [29]. Finally, "drop foot", as originally described by Winters et al. [27], consists not only of increased plantar flexion or lack of dorsiflexion during the swing phase, but also by increased knee and hip flexion during various parts of the gait cycle.

4. Discussion

The three-fold aim of this review differentiates it from the previous review that aimed to critically evaluate the GCSs that were published between 1966 and 2005 [5]. Even though the search period of the present update spanned only 12 years, 15 new GCSs have emerged [6–20], in comparison to 18 included in the previous review [5]. This difference in number is quite remarkable, especially taking into consideration the stricter inclusion criteria applied in this study (for example the exclusion of abstracts). This clearly indicates that researchers are continuously trying to characterize gait, in order to facilitate clinical practice [5,17,20,58] or to assist in the evaluation of inter- or intra-patient differences or treatment outcome [17,20,59]. The methodological shortcomings identified in the previous review and the lack of consensus among researchers necessitated the inclusion of reliability or validation studies (in case these would be available), which was not exhaustively covered previously [5]. The current study of these psychometric properties additionally illustrated the strengths or limitations of all GCSs, and supported the derivation of commonly used patterns in literature, by providing objective criteria.

4.1. Classification construction

In the present study, out of the 15 newly identified GCSs, 6 evaluated gait deviations across all planes of motion [6,11,12,16,18,20], whereas 4 studies were labelled as "non-applicable" because they either assessed only kinetic [14], spatiotemporal [7,9] or EMG data [8]. From the remaining studies (n = 5), 3 described gait patterns in the sagittal plane [10,13,15], 1 in both sagittal and transverse planes [17] and 1 only in the transverse plane [19]. The utility and need to evaluate the deviations of CP gait in non-sagittal planes of motion - and especially in the transverse plane - have been increasingly documented [12,15,17,19,29,58,59]. This need might explain the improvement in comparison to Dobson et al. [5], where only 1 study evaluated impairments in all 3 planes [49]. According to Davids and Bagley, deviations in the coronal plane are "sagittal and transverse plane deviations seen out of plane" and their clinical significance is unclear [17]. Contradictory to this, a more recent study based on international experts' consensus established through a Delphi process, considered coronal plane deviations to be important when evaluating CP patients' gait [20].

The construction method of all GCSs was adequately described, apart from one [6]. This might be due to the fact that Wren et al. did not intend to publish a GCS, but to study the prevalence of gait deviations

Table 3
Studies reporting reliability of gait classification systems in children with cerebral palsy, scored according to the adapted version of the COSMIN scoring criteria [25].

Study	Sample size	Blind raters	Intra-rater reliability assessed	Time interval stated	Time interval appropriate	ICC calculated	Kappa calculated	Weighted kappa	Weighting scheme	Cosmin
Winters et al. [27]										
Dobson et al. [30]	Excellent	Excellent	Fair	N/A	N/A	N/A	Excellent	Excellent	Excellent	Fair
Rodda et al. [29]	Fair	Excellent	Excellent	Excellent	Excellent	N/A	Excellent	Excellent	Excellent	Fair
Stott et al. [37]	Fair	Excellent	Excellent	Excellent	Excellent	N/A	Excellent	Excellent	Good	Fair
Kim et al. [38]	Excellent	Excellent	Fair	N/A	N/A	N/A	Excellent	Fair	N/A	Fair
Ferrari et al. [11]										
Pascale et al. [42]	Excellent	Excellent	Fair	N/A	N/A	Excellent	Excellent	N/A	N/A	Fair
Nieuwenhuys et al. [20]										
Nieuwenhuys et al. [43]	Excellent	Excellent	Excellent	Excellent	Excellent	N/A	Excellent	N/A	N/A	Excellent

In bold: original studies. The adapted COSMIN scoring criteria [25] can be found in Supplementary table 2.
Abbreviations: N/A: non-applicable ; ICC: intra-class correlation coefficient.

in children with CP [6]. In the process, though, they referred to deviations which had either not been previously mentioned in other GCSs or they defined patterns differently [6], which led to the study's inclusion in this review. Construction methods have been better described in more recent studies, in contrast to [5], where half of the studies had no details on the applied construction methods. The detailed description of a construction method may be dependent on whether a GCS is built based on a quantitative or a qualitative technique. In the review of Dobson et al., GCSs built in a qualitative way were never judged as displaying an adequate or partial description [5]. In the present review, on the contrary, most of the studies specifically referred to how their patterns were defined, based on observations or various levels of expertise [11,14,17,19,20].

For the category "classification groups", it was judged whether the gait deviations were adequately described such that the derived groups could be adequately differentiated between one another, similar to the criteria found in [5]. The vast majority of the studies (n = 12) provided sufficient information on the various classes that resulted from the formation of each GCS, whereas in 3 studies, it was stated that the defined groups were not completely independent from each other [10,12,13]. Another attention drawing aspect is whether a GCS is always able to classify the population for whom it is intended. Ferrari et al. stated that, with their GCS, 91% of the considered patients could be successfully assigned to a gait pattern, indicating that merely 9% of their patient sample remained unclassified [11]. This, however, was not the case for other GCSs. When re-implementing the GCS proposed by Winters et al. [27], several studies indicated an inability to classify their patients using the reported definitions, resulting in the proposal of an extra, less involved class [30–32]. The prevalence for this extra class reached up to 42% of the recruited patients in a study performed on a population-derived sample [32]. This substantial number of unclassified patients may be explained by the fact that the exact definitions provided by the authors of the original study may not have been precisely applied [30,31]. The inability to classify all patients has also been presented in the studies of Stott et al. who applied the GCS of Rodda et al. [29] where 3 patients could not be classified [37], de Morais-Filho, who could not classify 13% of their patients (n = 228) [36] when using the GCS of Sutherland and Davids [28], and, more recently, in a reliability study of a GCS where 4.7% of all gait trials could not be classified [43] according to the rules suggested by Nieuwenhuys et al. [20]. Failure of a GCS to assign observed gait patterns to one of the predefined classes might on one hand be dependent on its

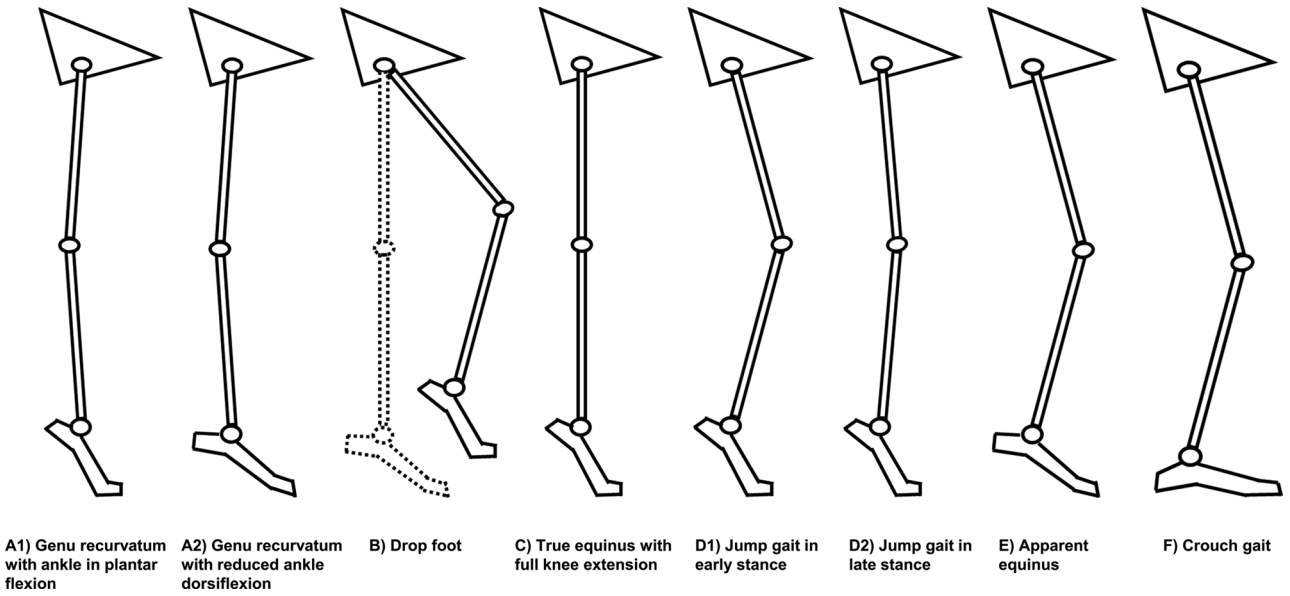

Fig. 2. Commonly used multiple joint patterns. The detailed descriptions of each pattern are provided in Table 4. A1) genu recurvatum with ankle in plantar flexion; A2) genu recurvatum with reduced ankle dorsiflexion; B) drop foot; C) true equinus with full knee extension; D1) jump gait in early stance; D2) jump gait it late stance; E) apparent equinus; F) crouch gait. The bold black lines indicate the deviations comprising the pathological multiple joint patterns, the dashed lines in B represent the contralateral lower limb.

Table 4
Overview of commonly used multiple joint patterns.

Pattern	Originally introduced by	Description	Reference to original study or pattern name in other studies	Reliability
Genu recurvatum	Simon et al. [48]	Full knee Ext or HE during stance, with almost normal hip motion during stance and impaired ankle motor control, resulting in PF or reduced DFL	[10 - Toro et al., SH / SD / SQ / SM / Dystonia / Ataxia]: Cluster 6 (equinus gait types) showed PF throughout the GC, full knee Ext or HE and hip Ext possible in single stance [17 - Davids and Bagley, CP]: True equinus with (hyper)extended knee and/or hip [27 - Winters et al., SH]: Group II = Persistent PF throughout the GC, full knee Ext or HE during stance [28 - Sutherland and Davids, SD]: Recurvatum knee gait = Increased knee Ext to HE in mid- and late stance, with variable motion in the swing and equinus type pattern with PF at foot strike, loss of early stance DFL and increased PF in mid- and late stance [48 - Simon et al., SH / SD]: Genu recurvatum = Full knee Ext or HE during stance with almost normal hip motion during stance and impaired ankle motor control. Early recurvatum = Full knee Ext or HE during stance with (neutral or) PF of the ankle and forward trunk lean. Late recurvatum = Full knee Ext or HE during stance with (neutral or) DFL on the ankle and backward trunk lean [50 - Hullin et al., SH]: Group IV = knee HE with tibial arrest or Group V = knee HE with persistent ankle DFL [55 - Lin et al., SD]: Patients with recurvatum present also with ankle PF throughout stance and (slightly) decreased hip ROM [56 - Yokochi et al., SD]: Patients that had knee Ext of equal or more than 0° during stance, always combined with ankle PF and trunk 'bending' [57 - O'Byrne et al., SH / SD]: Mild recurvatum (mild genu recurvatum combined with possible ankle DFL also during swing) and severe recurvatum (marked genu recurvatum, slight hip FL and ankle PF)	[41,43]
Drop foot	Winters et al. [27]	Dropfoot during swing but adequate DFL ROM, increased knee FL at TSw, IC and loading response, hip HFL during swing and increased lordosis throughout the GC	[11 - Ferrari et al., SD]: Dropfoot used to describe PF during swing, combined with DFL during push-off, increased knee amd hip FL during stance in group 2 or with hip and knee FL and possible placing reaction in group 4 [16 - Bonnefoy-Mazure et al., SH / SD]: RB group described as similar to a "true equinus profile gait ". It showed PF during swing, reduced DFL in comparison to controls during stance (-1StD just below 0o), increased knee FL in TSw and IC, hip increased FL in swing [27 - Winters et al., SH]: Dropfoot during swing but adequate DFL ROM, aMaxDF > 0° during stance, increased knee FL at TSw, IC and loading response, hip HFL during swing and increased lordosis throughout the GC [50 - Hullin et al., SH]: Group I = dropfoot in swing with normal stance kinematics [57 - O'Byrne et al., SH / SD]: Drop foot pattern = ankle in PF during swing with hip and knee in FL	[43]
True equinus	Rodda et al. [29]	Ankle in equinus during stance, full knee Ext, full hip Ext, pelvis within normal ROM or anterior tilt	[10 - Toro et al., SH / SD / SQ / SM / Dystonia / Ataxia]: Clusters 5 and 8 (equinus gait types) showed PF throughout the GC, full knee Ext and hip Ext possible in single stance [12 - Carriero et al., SD]: Area 1 covered CP children with an equinus pattern (toe-walking) [17 - Davids and Bagley, CP]: True equinus with normal knee and/or hip motion [29 - Rodda et al., SD]: Ankle in equinus, full knee Ext, full hip Ext, pelvis within normal ROM or anterior tilt	[29,37,38]

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Table 4 (*continued*)

Pattern	Originally introduced by	Description	Reference to original study or pattern name in other studies	Reliability
Jump gait	Rodda et al. [29]	Ankle in equinus, particularly in late stance. Knee and hip in HFL in early stance, followed by Ext to a variable degree in late stance, pelvis within normal ROM or anterior tilt	[11 - Ferrari et al., SD]: Patterns 1 and 3: continuous ankle PF with increased hip + knee flexion [12 - Carriero et al., SD]: Area 2 covered CP children with a jump knee pattern (excessive knee flexion at initial contact followed by extension later in stance). [13 - Rozumalski and Schwartz, CP]: refers to the term "jump gait" as "problem of increased knee flexion at initial contact" / resembles pattern "moderate crouch with equinus" [16 - Bonnefoy-Mazure et al., SH / SD]: LB gait profile described as similar to a "jump knee gait profile" and is characterized by an excessive ankle PF and reduced ROM for the hip and knee [17 - Davids and Bagley, CP]: true equinus with flexed knee/hip indicates PF with both other joints in flexion [27 - Winters et al., SH]: Group IV = ankle PF, reduced knee and hip range of motion, increased pelvic lordosis during terminal stance [28 - Sutherland and Davids, SD]: jump knee refers to increased knee and hip flexion, with reduced ROM with (near) normal knee extension in stance being possible and dorsiflexion in initial stance [29 - Rodda et al., SD]: Ankle in equinus, particularly in late stance. Knee and hip in HFL in early stance, followed by Ext to a variable degree in late stance, pelvis within normal ROM or anterior tilt [55 - Lin et al., SD]: "jump group" with increased KF at IC and decreased knee ROM, but PF only in swing and good hip ROM [56 - Yokochi et al., SD]: FP group: knee in flexion, ankle in PF, lumbar lordosis	[29,37,38]
Apparent equinus	Rodda et al. [29]	Ankle normal ROM, knee and hip in HFL throughout stance, pelvis within normal ROM or anterior tilt	[11 - Ferrari et al., SD]: Patterns 2 and 4 display PF at IC, but not throughout the GC, along with hip and knee flexion [13 - Rozumalski and Schwartz, CP]: Pattern "moderate crouch with anterior pelvic tilt" [16 - Bonnefoy-Mazure et al., SH / SD]: RT gait profile described as similar to an apparent equinus profile gait, showing a slightly higher flexion at the knee and hip joints [17 - Davids and Bagley, CP]: Apparent equinus with foot normally aligned relative to the tibia (not in PF) and excessive knee and/or hip flexion [29 - Rodda et al., SD]: Ankle normal ROM, knee and hip in HFL throughout stance, pelvis within normal ROM or anterior tilt [50 - Hullin et al., SH]: Group III = Persistent hip and knee flexion : IC with toe or foot flat contact, knee flexion throughout stance, no hip extension [57 - O'Byrne et al., SH / SD]: Proximally flexed ankle walkers = hip and knee joints in flexion with normal ankle movement and ROM	[29,37,38]

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Table 4 (*continued*)

Pattern	Originally introduced by	Description	Reference to original study or pattern name in other studies	Reliability
Crouch gait	Rodda et al. [29]	Ankle in excessive DFL throughout stance, knee and hip in HFL, pelvis in normal ROM, anterior or posterior tilt	[10 - Toro et al., SH / SD / SQ / SM / Dystonia / Ataxia]: 4 crouch gait type clusters, all clusters show increased knee flexion, decreased hip ROM (clusters 3 and 4 also always > 0°), ankle motion variable [12 - Carriero et al., SD]: Area 3 covered CP children with a crouch knee pattern (knee flexion throughout stance ≥30°) [13 - Rozumalski and Schwartz, CP]: Pattern "severe crouch" is similar [16 - Bonnefoy-Mazure et al., SH / SD]: LT gait profile described as similar to a crouch gait profile, described by a triple FL (hip, knee in FL and ankle in DF) and a reduction of knee and hip ROM [17 - Davids and Bagley, CP]: crouch gait characterized by flat-foot or calcaneal contact pattern and increased knee flexion throughout stance, tertiary deviations at the hip (increased flexion) or pelvis (increased anterior tilt) are possible [28 - Sutherland and Davids, SD]: crouch knee = increased knee flexion (≥30°) throughout stance and increased ankle DF, increased hip flexion throughout stance in case of iliopsoas contracture [29 - Rodda et al., SD]: Ankle in excessive DFL throughout stance, knee and hip in HFL, pelvis in normal ROM, anterior or posterior tilt [55 - Lin et al., SD]: Crouch group showed increased ankle DF and knee and hip flexion throughout stance [56 - Yokochi et al., SD]: Patients with knee FL, ankle DF and pelvic lordosis [57 - O'Byrne et al., SH / SD]: Severe crouch = hip and knee joints diminished ROM and in FL, ankle in DFL	[29,37,38]

Abbreviations: Ext: extension; HE: hyperextension; PF: plantar flexion; DFL: dorsiflexion; ROM: range of motion; FL: flexion; TSw: terminal swing; IC: initial contact; HFL: hyperflexion; GC: gait cycle; SH: spastic hemiplegia; SD: spastic diplegia; StD: standard deviation; SQ: spastic quadriplegia; SM: spastic monoplegia; CP: cerebral palsy.

inability to cover all possible gait deviations in the researched patient sample. On the other hand, it has been argued that CP patients' gait lies along a continuum [7,17], hence forcing it in distinct or "hard" classes might be inaccurate. This was the case in the study of Vaughan et al. where they allowed for variable membership of each patient to the defined groups [7]. Moreover, Carriero et al. also found an overlap between their CP groups, highlighting the possible existence of simultaneous gait deviations [12], while Van Gestel et al. tested their algorithm both for distinct and for mixed patterns in CP gait [15]. Overall, this review found an improvement in the description of the classification groups in comparison to [5], with 21 studies being judged as having "adequately" defined groups.

4.2. Sample and generalizability

As far as the included patients in each study are concerned, there was quite some heterogeneity in applied patient inclusion criteria. First, there is still no common language between studies, as the majority referred to patients (n = 23) [6,7,9,11,12,18,27,29–44]. Others referred to limbs (n = 5) [8,10,16,19,47], trials (n = 4) [14,15,45,46] or even strides (n = 1) [13]. Another remaining issue is the extent of information taken into consideration when studying the gait of CP children. Several researchers chose to average the gait cycles of their patients, creating a "representative trial" from each patient or limb [7,10,16,31,36,38,40]. Others selected one representative trial out of the available ones [14,19,29,32,43,44], or included three [18] or all available trials [8,15,35,37,45,46].

In this review, it was decided to report the number of observations (patients / limbs / trials / strides) based on where emphasis was put on in the results of each study. The sample size was subsequently labelled according to the number of observations per pattern. Hence, in studies where joint patterns were defined or validated, the pattern discrimination capacity against the characterization of their sample size could be evaluated.

Detailed characteristics of the patient sample were sometimes not available, which is illustrated by the generalizability scoring. This was more obvious in studies where the development of an algorithm was the main purpose of the study [8,9,12,13,35,39,46]. It is important to note that some GCSs, such as Winters et al. or Rodda et al., were originally designed only for hemiplegic or diplegic patients, respectively, influencing their generalizability score. Additionally, in studies where generalizability was "non-applicable" [17,20], specific patients were actually not classified in the original publications; these GCSs, though, were built with the intention to be able to describe gait deviations in all children with spastic CP.

4.3. Other forms of validity

In the considered studies, either one of the 6 "other" forms of validity was frequently examined, resulting in an almost equal distribution of excellent, good and fair scores. The lack of thorough descriptions of applied methods and included patients were mostly responsible for "good" scores, whereas "fair scores" were mostly assigned due to the limited description of patients. The methods were considered as "excellent" only when specific criteria were stated in each study, such as who classified the trials and how the study was conducted. The results were reported in detail in most of the cases and appropriate statistics to the purposes of each exploration were usually applied. In order to ensure the incorporation of GCSs in clinical decision making and treatment outcome evaluation, GCSs should relate to clinical symptoms. So far, only few studies have evaluated the association between the defined gait patterns and the symptoms of spasticity [13,16,31,39,44], weakness [13,16,44] or loss of selective motor control [13]. Finally, an extra validation sample was used in only 7 studies, with a sample size ranging from only 2 patients [9] to more than 750 CP children [39].

4.4. Clinical applicability and reliability

For clinical applicability, the studies that scored the lowest ("fair") were GCSs built upon automatic classification algorithms. In those cases, minimal clinical input was taken into consideration in the development of the patterns. An example of clinical input in GCSs built upon automatic classification algorithms is the a priori choice of important gait parameters to be incorporated in the algorithms [7,9,10,12,16,18]. The studies that developed automatic classification algorithms based upon previously published and validated GCSs were considered to be "driven" by qualitative rules and were labelled as displaying a "good" clinical applicability [35,39,46].

Since the previous review [5], the reliability of GCSs has been evaluated more frequently. In total, six reliability studies have been conducted [29,30,37,38,42,43], 5 of which had not been included in the previous review. Inter-rater reliability was assessed in all studies, in 3 of which intra-rater reliability was additionally established [29,37,43].

4.5. Multiple joint patterns with consensus

The third aim of this review was to evaluate whether consensus exists over certain multiple joint patterns. This can be performed by several techniques including the Delphi consensus approach to establish clinical experts' agreement [60] or by conducting a thorough analysis of the available literature, as in this review. Several GCSs were initially intended to be applicable to a specific population, either hemiplegic [27,50] or diplegic [11,19,28,29] CP children. Based on this review, however, it was apparent that the multiple joint patterns where agreement was reached according to the criteria described in Section 2.5, have been observed in both populations, further establishing their use in all ambulant children with CP [8,9,16–19,27–29,48,50,55–57] (Table 4).

The literature review revealed that six multiple joint patterns displayed a relevant consensus (Fig. 2, Table 4). Establishing an agreement over the entire GCSs instead of individual gait patterns would be more difficult or even virtually impossible based on the performed systematic review. Interestingly, 4 out of these 6 commonly used multiple joint patterns (Table 4) were described in the GCS established by Rodda et al. [29], who introduced the "true equinus" and "apparent equinus" patterns and extended two patterns originally introduced by Sutherland et al. [28]. The patterns "jump gait", "apparent equinus" and "crouch" can mainly be differentiated from one another based on the position of the ankle joint [29]. In contrast to other studies that have defined "crouch" as a pattern of increased knee flexion during stance [6,13], according to [29], knee motion on its own is not sufficient to distinguish crouch from other patterns. Despite the variability in definitions of these 4 multiple joint patterns, the characteristics presented by Rodda et al. were considered as the most exhaustive ones, always including information about the co-occurring deviations across all lower limb joints, while stemming from the GCS with the most established validity and reliability [29,35–37].

Apart from these 4 patterns established by Rodda et al. [29], the literature review also highlighted two additional patterns, namely "drop foot" and "genu recurvatum". While the name of these two patterns, just like the true equinus pattern, suggests strong emphasis on one specific joint movement deviation, they are multiple joint patterns since they are combined with specific descriptions of the motions in the other lower limb joints, such as compensations and/or overall minor gait deviations. While "drop foot" may appear as a swing phase pattern, its other gait features in stance (i.e. normal dorsiflexion), as well as minor deviations in other joints (i.e. increased knee flexion at initial contact, during loading response and terminal swing, increased hip flexion during swing and pelvic lordosis throughout the gait cycle) show that this is not the case [27]. Hence, this definition does not allow to combine "drop foot" with other multiple joint patterns that appear

mainly as stance phase patterns. The multiple joint patterns that were highlighted in this review do not enable a simultaneous inspection of the pathological motions occurring during the stance and swing phases of gait. The patterns “genu recurvatum” and “true equinus” differ in the main focus of the included deviations, the knee or the ankle joint motion, respectively. The former describes a knee motion towards or in hyperextension with reduced dorsiflexion and possibly -but not necessarily- plantarflexion in the ankle. The latter refers to a constantly plantarflexed ankle during stance, that may be combined with a normal or increased knee extension and a possibility for a complete extension at the level of the hip [29,48]. This review has concluded that the original pattern definitions may cause confusion in case a patient suffers simultaneously from plantarflexion and knee hyperextension during stance. According to Davids and Bagley [9], a distinction should be made between a true equinus movement with a normal knee motion or a knee in hyperxtension. For reasons of clarity, it was decided to adopt the same reasoning and support the classification of patients who exhibit both pathological motions in the “genu recurvatum” pattern, while keeping the “true equinus” pattern for equinus combined with normal knee motion. Future classification of actual patient data might prove whether both of these two patterns are prevalent and should be considered independently or not. It is likely that a multiple joint pattern that describes mild deviations across all lower limb joints is still missing from this overview of commonly used multiple joint patterns, as suggested in previous studies [15,17,20,31]. Lastly, all 6 commonly used multiple joint patterns describe sagittal plane deviations. Multiple joint deviations in the coronal or the transverse plane have hardly been defined. Apart from the study of Simon et al. [19], other GCSs have not described the potential co-occurrence of rotational deviations across different joints in children with CP, and in that study, no specific multiple joint patterns arose. The strict criteria of this review as to the inclusion of GCSs that have been cited by at least two other research groups might pose a bias to more recent GCSs. For the latter, there may not have been sufficient time to be discovered or replicated by other researchers. Three such studies were identified [17,19,20]. Davids and Bagley introduced a GCS based on expertise, the defined patterns of which resemble the sagittal plane patterns of Rodda et al. [29]. Simon et al. and Nieuwenhuys et al. described an extended series of distinct single joint patterns [19,20]. The combinations of these single joint patterns have not yet been thoroughly explored to declare them valid to be implemented as multiple joint patterns in clinical practice.

4.6. Limitations

As in the previous systematic review by Dobson et al. [5], a meta-analysis was not appropriate for this review, since outcomes in the form of effect sizes cannot be extracted from this type of studies [60]. Therefore, the COSMIN checklist was used and adapted, thereby ensuring a “best evidence synthesis”. By using the COSMIN, however, the lowest score of one category defined the category’s complete score, which may be considered as potentially too strict. Further, the selection of the four online databases that were searched and the inclusion criteria might have been restrictive. It is also possible that additional search terms could have resulted in the inclusion of more studies. In addition, with regards to the commonly used multiple joint patterns, considering the number of times a GCS has been cited by a different research group might be biased for recently developed GCSs. However, for the majority of the evaluated GCS, this criterion could establish whether a GCS and its patterns appeal to the scientific community or not. Finally, aiming to encourage a common language among clinicians or researchers, (minor) differences in definitions might have been overlooked. For example all patterns that have been described as ‘crouch’ were grouped, irrespective of whether they referred only to the pathological motion of the knee joint or to that of the other lower limb joints as well.

5. Conclusion

The present study aimed to report and critically appraise the GCSs published in regards to CP gait and to list the multiple joint patterns, with reported reliability and validity, which have reached a certain consensus in literature. The methodological quality of studies has improved by increasingly reporting on various levels of validity or reliability of GCSs, further establishing the clinical applicability of GCSs. On the other hand, specific limitations remain, such as the lack of association of gait patterns to clinical symptoms, or the inclusion of all 3 planes in the assessment of gait. The most common drawbacks of the current GCSs are still that the majority of them only or mainly considered kinematic parameters, without the simultaneous incorporation of EMG or kinetics, as well as the fact that qualitative and quantitative data are seldom combined, which would support the establishment of clinically valid GCSs. Lastly, highlighting the existence of 6 commonly used multiple joint patterns may aid clinical and research applications and create a common language among healthcare providers. Future validation studies could focus on exploring whether these 6 commonly used multiple joint patterns cover all potential patterns in children with spastic CP. Currently, there is no agreement over the co-occurrence of multiple joint patterns describing independent deviations of the stance and swing phases or in the other planes of motion. Finally, the relations of multiple joint patterns with clinical symptoms of spasticity, weakness and selectivity remain to be established.

Author statement

This study was designed by EP, AN, AVC, EO, and KD. EP, AN, and IV were responsible for the systematic searches, the quality assessment criteria establishment and the assessment of all included articles. All authors have had complete access to the study data, spanning the study duration. Every author contributed to the critical appraisal of the results and was subsequently involved in the revision of the manuscript that was written by EP. All authors approve the final version of the manuscript and agree to be accountable for the content of the work.

Conflict of interest statement

The authors declare that they have no conflict of interest.

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Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:https://doi.org/10.1016/j.gaitpost.2019.01.038.

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