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Review

Identifying methods for quantifying lower limb changes in children with idiopathic toe walking: A systematic review

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ABSTRACT

Background: Idiopathic toe walking (ITW) is a diagnosis of exclusion for children walking on their toes with no medical cause. This systematic review aimed to identify and evaluate the clinical utility, validity and reliability of the outcome measures and tools used to quantify lower limb changes within studies that included children with ITW.

Methods: The following databases were searched from inception until March 2018: Ovid MEDLINE, EBESCO, Embase, CINAHL Plus, PubMed. Inclusion criteria were studies including children with ITW diagnosis, reporting use of measurement tools or methods describing lower limb characteristics, published in peer-reviewed journals, and in English. The relevant psychometric properties of measurement tools were extracted, and assessed for reported reliability and validity. Included articles were assessed for risk of bias using McMaster quality assessment tool. Results were descriptively synthesized and logistic regression used to determine associations between common assessments.

Results: From 3164 retrieved studies, 37 full texts were screened and 27 full texts included. There were 27 different measurement tools described across joint range of motion measurement, gait analysis, electromyography, accelerometer, strength, neurological or radiology assessment. Interventional studies were more likely to report range of motion and gait analysis outcomes, than observational studies. Alvarez classification tool in conjunction with Vicon motion system appeared the contemporary choice for describing ITW gait. There was no significant association between the use of range of motion and gait analysis outcomes and any other outcome tool or assessment in all studies ($p > 0.05$).There was limited reliability and validity reporting for many outcome measures.

Significance: This review highlighted that a consensus statement should be considered to guide clinicians and researchers in the choice of the most important outcome measures for this population. Having a standard set of measures will enable future treatment trials to collect similar measures thus allowing future systematic reviews to compare results.

1. Introduction

Toe walking is commonly described as the lack of heel strike at the initial contact phase of the gait cycle. Although toe walking is regarded as a normal variation in gait development in children up to the age of three years [1], the maturation to ankle dorsiflexion at heel strike is usually completed by the age of five years [2,3]. Toe walking may be a consequence of a disease process, trauma or neurogenic influences [4]. Conditions known to cause or be associated with this gait type include cerebral palsy, muscular dystrophy, autism spectrum disorders, global developmental delays, lower limb injury or tumours [4].

Where there is an unknown medical cause for the gait pattern, this condition is defined as idiopathic toe walking (ITW) [5]. The estimated prevalence of ITW is approximately 5% in healthy children [6], It typically affects boys more than girls, but presents in both genders [7]. ITW is most commonly associated with ankle equinus [8] and often considered an effect of the toe walking gait over time if left untreated [9]. Consequences of equinus can result in lower limb and/or foot pain [10], and poor performance in sport and low exercise levels [11]. Many treatment studies for children diagnosed with ITW such as surgery [12,13] or serial casting [14,15] use the resolution or reduction of ankle equinus as the primary outcome measure. However, the impact of ITW

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Table 1
Search terms for systematic review of the literature.

"idiopathic toe walking"	"Lower limb"	Test*
"toe walking"	Leg*	Assessment*
Equinus	Foot	Equipment
"Ankle equinus"	Ankle	Device*
"Tip toe"	Hip	Tool*
"Habitual toe walking"	Knee	Measur*
"equinovarus"	Gait	Dynamometer
	Walk*	Characteristic*
	Locomot*	
	Ambulat*	

on lower limb characteristics beyond the ankle is unknown. It is also unknown if there is consistency between studies on the type of assessments that researchers are using to measure outcomes in this cohort.

The primary aim of this systematic review was to identify what lower limb characteristics were measured in children displaying ITW. Secondary aims were to determine which measurement tools were used in assessing these characteristics, and explore the validity and reliability of the identified measurement tools.

2. Methodology

This systematic review was reported and performed in compliance with the Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) [16]. The PICO (Population, Intervention Comparison and Outcomes) model was used to establish the search terms used [17]. The following databases were accessed: Ovid MEDLINE, EBESCO, Embase, CINAHL Plus, Pubmed from date of inception until March 2018. The key word search terms used in the review and strategy were customised for each database (Table 1). Targeted hand searching of reference lists and citations of included articles supplemented the search strategy.

Studies were included if they met the following criteria: children up to the age of 18 who were diagnosed with idiopathic toe walking, a measurement tool describing lower limb characteristics, published in peer review journals and available in English. Studies that reported a single case design or protocol for a prospective study were excluded. Studies where participants were toe walking as a result of described medical conditions were also excluded. The first author screened titles and abstracts. The full text of remaining papers were obtained and reviewed by two authors (AC/PM or AC/CW) with inclusion based on inclusion and exclusion criteria after agreement by both raters. Conflicting decisions discussed in person or with the third author until a consensus was reached.

2.1. Data extraction and quality appraisal

Article methodology was assessed for the level of evidence using the NHMRC Evidence hierarchy [18]. The reported reliability and validity of the tools used within studies were extracted. The McMaster quality assessment tool [19] was applied to assess the risk of bias and quality of the included studies. The overall score for each measurement property determined by a 'worse score counts' approach. Scores were allocated 1 for yes, 0 for no or not addressed, and the item deducted from the overall score for not applicable on these guidelines. Two assessors (AC and CW) individually performed data extraction and assessment of methodological quality. In the case of any uncertainty or if conflict arose, the authors resolved this through discussion with a third author (PM).

2.2. Data analysis

Due to the heterogeneity of the included studies, a descriptive synthesis of the results was undertaken. Analysis was performed with Stata 13 [20]. Frequencies of outcome assessment groups were calculated and logistic regression analysis used to explore if there were associations between the two most commonly used outcome assessment groupings and any of the other outcome assessment groupings identified. Statistical significance was set at $p < 0.05$.

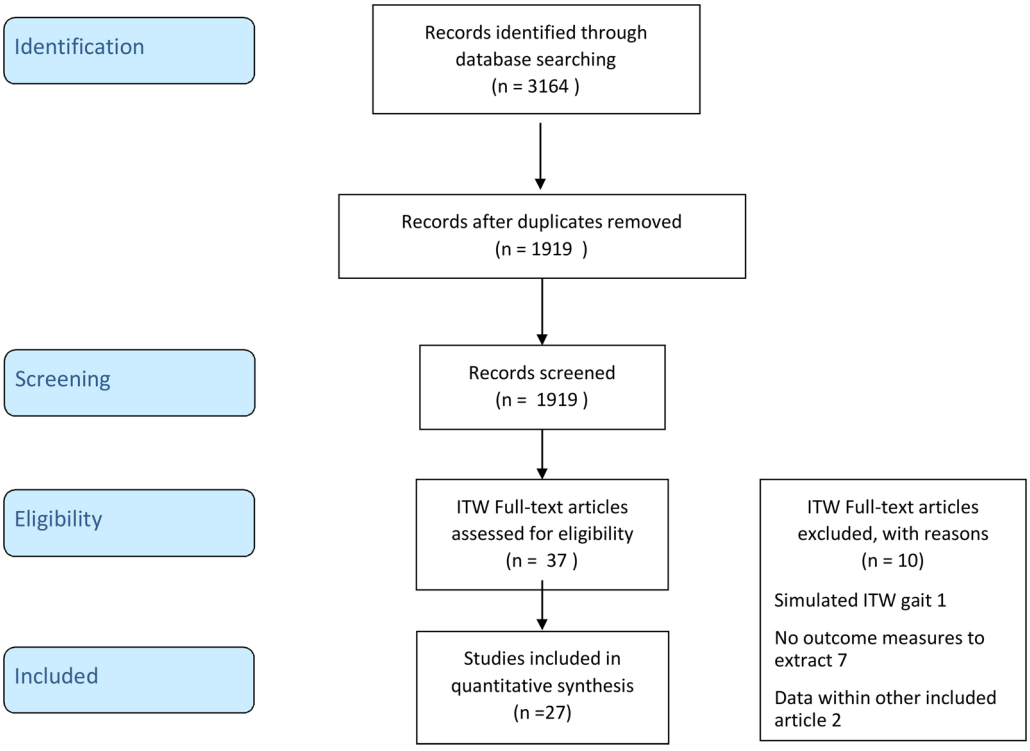

Fig. 1. Prisma Flow chart.

Table 2
Table of Included ITW studies.

AUTHOR	STUDY DESCRIPTION	LEVEL OF EVIDENCE	LOCATION	AGE RANGE (YRS)	AGE MEAN (SD IF REPORTED) (YRS)	SAMPLE SIZE
Beneditti [35]	Prospective Case Control (intervention)	III3	Italy	Not reported	7.6*	9
Christensen [36]	Prospective Cohort Study (Observation	III3	USA	3 to 17	4.8	45
Christensen [37]	Prospective Cohort Study (Observation	III3	USA	7 to10	7.7	30
Clark [24]	Prospective Case Series (intervention)	IV	USA	2.9 to 5.4	4.2 (1.2)	5
Davis [28]	Retrospective Cohort Study (Intervention)	III3	Canada	Group 1: 4.9 to13.0 Group 2: 4.3 to12.2	Group 1: 8.7 (2.7) Group 2: 7.1(2.2)	Group 1: 20 Group 2: 24
Engelbert [7]	Cross sectional (observation)	III2	Netherlands	Not reported	Equinus 15.2 (2) No Equinus: Not reported	Equinus:31 No Equinus: 9
Engstrom [14]	RCT (intervention)	III1II	Sweden	5 to 14.5	9.4	47
Fanchiang [32] [31]	Prospective Case Control (intervention)	III2	USA	Not reported	6.8 (1.6)	15
Fox [15]	Prospective Case Series (intervention)	IV	UK	2 to 14.4	6.1(3.5)	44
Hemo [3]	Retrospective Case Series (intervention)	III-3	USA	4.2 to 13.1	9.0	15
Herrin [38]	RCT(intervention)	II	USA	Not reported	Group 1: 5.49 Group 2: 4.58	Group 1: 9 Group 2: 9
Kelly [39]	Prospective Case Control (Observation)	III-3	Ireland	4 to 14	7.0	22
McMulkin [33]	Retrospective Case Series (intervention)	IV	USA	6.4 to11.3	9.0	8
Murphy [40]	Prospective Case Series(intervention)	III3	Australia	Not reported	5.93 (1.83)	15
O'Sullivan [25]	Retrospective Cross Sectional (Observation)	IV	Ireland	4 to 16	Not reported	81
Pendharkar [11]	Prospective Case series (Observation)	III3	Australia	5 to 12	7.7	5
Pendharkar [41]	Prospective Case series (Observation)	III2	Australia		8.5 (3.1)	10
Policy [30]	Prospective Case Control (Observation	III	USA	3 to10	6.8(2)	8
Pomarino [26]	Prospective Case Control(Observation)	II2	Germany	2to 13	Not reported	0
Rose [29]	Prospective Case control (Observation)	III2	USA	3 to 10	5.1(2.4)	8
Satila [22]	RCT (Intervention)	II	Finland	2 to 9 2.5 to8	Group 1: 4.9 Group 2: 5.3	Group 1: 14 16
Sinclair [27]	Retrospective Cross Sectional (Observation)	III1	USA	6 to14	8.7	22
Stricker [42]	Retrospective Cohort Study (Intervention)	IV	USA	All groups: 2-13	Group 1: 3.2 Group 2: 4.2 Group 3: 3.9	Group 1: 48 Group 2: 17 15
Van Bemmel [43]	Prospective Case Series (Intervention)	IV	Netherlands	6 to 16	11.5	55
Williams [8]	Prospective Case control (Observation)	III1	Australia	4 to 8	6 (1.4)	30
Williams [44]	Prospective Case Control (Intervention)	III3	Australia	Not reported	5.93 (1.83)	15

* Only ITW population described.

3. Results

There were initially 3164 studies screened for inclusion (Fig. 1). Of these, 37 full texts were assessed and 27 were included in the review. Fig. 1 describes the process of study retrieval and selection and Table 2 describes the study characteristics. The participants' ages ranged from 2 [21,22] to 19.5 [23]years. Sample size ranged from five participants [24] to 81 [25]. There were a variety of study designs; however, only two studies were randomised comparative effectiveness trial [14,22].

Outcome measures were categorised and tabulated (Table 3 and Supplementary data 1). These measures predominantly focused on identification of any impairment associated with the ITW gait pattern. The number of outcome measures reported by studies varied from a single outcome measure category [26,27] through to four concurrent different outcome measure categories [28,29]. The two outcome categories with the greatest frequency of use were range of motion (ROM) testing (n = 17, 63%) and gait analysis (n = 17, 63%). When ROM was captured as an outcome measure, gait analysis was also used 10 times (59%), accelerometery was also used once (6%), electromyography (EMG), strength and neurological measures were each concurrently used three times (18%). Where gait analysis was used, ROM was also used 10 times (59%), accelerometery and strength measures were also used twice (12%), EMG was used once (6%). There were no studies capturing both gait analysis outcomes and neurology assessment/outcome measures as displayed in Table 3 and Fig. 2. Despite there being equal use of range of motion and gait analysis outcomes, there was no significant association between the use of range of motion and gait analysis outcomes and any other outcome tool or assessment in all studies ($p > 0.05$).

There were 15 interventional studies, of which 12 studies used ROM as an outcome measure and of these 12, the majority used goniometry as a measurement tool (n = 8, 53% of 15 studies), 14 used Gait Analysis and within this group, almost half used the Vicon (n = 7, 47% of the 15 studies). The Alvarez classification system was used in four of these 15 interventional studies, in combination with Vicon in three studies and with plain video in one study. Of note, there was only one interventional study that measured lower limb strength, and one study that utilized EMG. No interventional study collected accelerometer data.

There were 12 observational studies of which four collected ROM data with goniometry (n = 4, 33% of the 12 studies), and five provided instrumented gait analysis data with the GaitRite® (n = 2, 17% of the 12), Coda-3 motion analysis system (n = 2, 17% of the 12) or simple video recording (n = 1, 8% of the 12). Accelerometry (n = 4, 33% of the 12) was exclusively used in the observational studies.

The reliability and validity of the actual measurement tools used within the studies were captured within the McMaster quality assessment within questions 5 and 6 respectively (Table 4). Of the 16 measurement tools identified, only six reported reliability data and two reported complete validity data within the article. Reliability data was not provided or referenced for any studies utilising measures of accelerometry or EMG. The two studies each that used measures of strength [14] and tone [29,30] both provided evidence of reliability. Reliability and validity for the ROM measure, the weight bearing lunge test was referenced [8], however these results were obtained using an adult cohort, not children. Table 4 provides scores assessing the risk of bias relating to the study design using the McMaster quantitative critical

Table 3
Outcome measures included within studies.

Study	Range of motion	Gait Analysis		Accelerometer	EMG	Strength	Neurology	Radiology
		Equipment	Classification					
Beneditti [35]	X	X						
Christensen [36]		X		X				
Christensen [37]		X		X				
Clark [24]	X	X						
Davis [28]	X	X	X		X			
Engelbert [7]	X					X		
Engstrom [14]	X	X				X		
Fanchiang [31,32][30		X	X					
Fanchiang [32] [31]		X	X					
Fox [15]	X	X						
Hemo [3]		X						
Herrin [38]	X	X						
Kelly [39]		X						
McMulkin [33]	X	X	X			X		
Murphy [40]	X	X						
O'Sullivan [25]		X					X	
Pendharkar [11]				X				
Pendharkar [41]				X				
Policy [30]	X				X		X	
Pomarino [26]	X							
Rose [29]	X		X		X		X	
Satila [22]	X		X					
Sinclair [27]								X
Stricker [42]	X							
Van Bemmel [43]	X							
Williams [8]	X	X						
Williams [44]	X	X						

appraisal tool [34] which ranged from 6/11 to 13/13. All domains were scored for included articles. The majority of studies did not justify a sample size (n = 16, 59%) in addition to poor scores for documenting reliability (n = 14, 52%) and validity (n = 17, 63%) of outcome measures or tools used.

4. Discussion

This review identified a large number of measurement tools utilised in descriptions of ITW in both observational, and interventional studies. No prior systematic review has explored the outcome measures and assessment tools utilised in this population group, nor subjected the tools to critique for their validity and reliability. Understanding any similarities in studies and which are the most valid and reliable measures allows researchers to plan measures appropriate to the outcomes of interest. It also enables health professionals to consider which measures should be used within the clinical setting, ensuring best practice recommendations are rapidly translated and adopted.

Most studies included in this review varied widely in selection of assessments and measures used during diagnosis, descriptive and intervention outcomes. More recently (2015 onwards) the Alvarez classification in conjunction with Vicon motion analysis are suggestive of increasing use [31–33], although two studies from similar research groups utilized the GaitRite®. Of note, instrumented computerised motion analysis systems are expensive and may not be readily accessible for many health professionals in clinical practice. Although Vicon and GaitRite® systems require gait laboratory conditions, the Alvarez classification has the potential to be used by health professionals with relatively basic video software in the clinical setting [28].

ITW is an exclusionary diagnosis eliminating orthopaedic concerns and neurological diagnoses. The use of some neurological tests commonly used within the diagnostic suite of outcome measures was seen

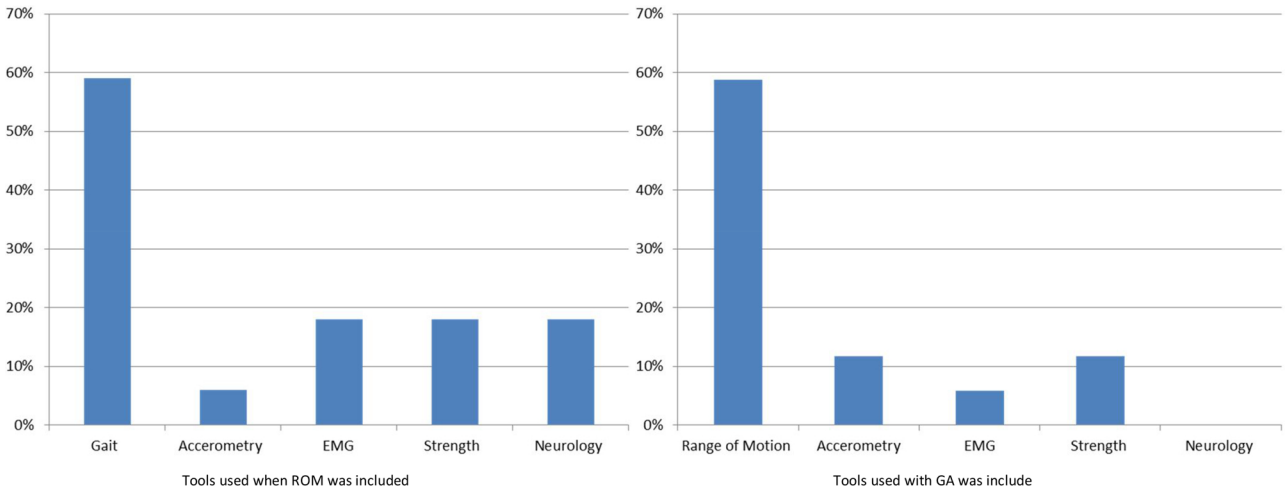

Fig. 2. Additional measurement tools used when range of motion (ROM) and Gait Analysis were included.

Table 4
McMaster Quality Assessment Scores.

Study	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10	Q11	Q12	Q13	Q14	Q15	TOTAL
Beneditti [35]	Y	Y	Y	N	N	N	Y	NA	N	Y	Y	Y	NA	N	Y	8/13
Christensen [36]	Y	Y	Y	N	Y	Y	NA	NA	NA	Y	Y	Y	NA	NA	Y	9/10
Christensen [37]	Y	Y	Y	Y	Y	Y	NA	NA	NA	Y	Y	Y	NA	N/A	Y	10/10
Clark [24]	Y	Y	N	N	Y	Y	Y	Y	Y	Y	Y	Y	NA	Y	Y	12/14
Davis [28]	Y	Y	Y	Y	N	N	N	N	N	Y	Y	Y	Y	NA	Y	9/14
Engelbert [7]	Y	Y	Y	N	Y	Y	NA	NA	NA	Y	Y	Y	Y	NA	Y	10/11
Engstrom [14]	Y	Y	Y	Y	N	N	Y	Y	N	Y	Y	Y	Y	Y	Y	12/15
Fanchiang [31,32,30]	Y	Y	Y	N	N	N	Y	Y	Y	Y	Y	Y	Y	N/A	Y	11/14
Fanchiang [32] [31]	Y	Y	Y	N	Y	Y	Y	Y	Y	Y	Y	Y	Y	N/A	Y	13/14
Fox [15]	Y	Y	Y	N	N	Y	Y	Y	Y	Y	Y	Y	NA	N	Y	11/14
Hemo [3]	Y	Y	Y	N	N	N	Y	N	Y	Y	Y	Y	NA	NA	Y	9/13
Herrin [38]	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	15/15
Kelly [39]	Y	Y	Y	N	N	Y	NA	NA	NA	N	N	Y	Y	NA	Y	7/11
McMulkin [33]	Y	Y	Y	N	Y	Y	N	N	N	Y	Y	Y	NA	NA	Y	9/13
Murphy [40]	Y	Y	Y	N	Y	Y	Y	Y	Y	Y	Y	Y	NA	NA	Y	12/13
O'Sullivan [25]	Y	Y	N	NA	N	Y	NA	NA	NA	Y	Y	Y	Y	NA	Y	9/10
Pendharkar [11]	Y	Y	Y	N	Y	Y	NA	NA	NA	Y	Y	Y	NA	NA	Y	9/10
Pendharkar [41]	Y	Y	Y	N	NA	NA	NA	Y	Y	Y	Y	Y	Y	NA	Y	10/11
Policy [30]	Y	Y	N	N	Y	Y	NA	N	Y	Y	Y	Y	Y	NA	Y	10/13
Pomarino [26]	Y	Y	N	N	N	N	NA	NA	NA	Y	N	Y	Y	NA	N	6/11
Rose [29]	Y	Y	Y	N	N	N	NA	NA	NA	Y	Y	Y	Y	NA	Y	9/12
Satila [22]	Y	Y	Y	Y	N	N	Y	Y	N	Y	Y	Y	Y	Y	Y	12/15
Sinclair [27]	Y	Y	N	NA	Y	Y	Y	N	N	Y	Y	Y	Y	NA	Y	10/13
Stricker [42]	Y	Y	Y	NA	N	N	N	Y	Y	Y	Y	Y	Y	N/A	Y	10/13
Van Bemmel [43]	Y	Y	Y	Y	Y	Y	NA	NA	NA	Y	Y	Y	Y	NA	Y	11/11
Williams [8]	Y	Y	Y	Y	Y	Y	NA	NA	NA	Y	Y	Y	Y	NA	Y	11/11
Williams [44]	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	NA	N/A	Y	13/13

McMaster Quality Assessment Questions: Q1 Was the purpose of the study clearly stated?: Q2 Was relevant background literature reviewed? Q3 Was the sample described in detail? Q4Was sample size justified? Q5 Were the outcome measures reliable? Q6 Were the outcome measures valid? Q7 Was Intervention was described in detail? Q8 Was Contamination avoided? Q9 Was Cointervention avoided? Q10 Results were reported in terms of statistical significance? Q11 Were the analysis method/s appropriate? Q12 Clinical importance was reported? Q13 Were differences between groups clinically meaningful? Q14 Drop-outs were reported? Q15 Conclusions were appropriate, given study methods and results?

in three studies in this review [25,29,30]. The methodology of these was not reported in a way that leads to confidence that these measures should be used to identify outcomes in any interventional research for children with ITW. Typically the presentation of neurological signs would lead to a referral for further investigation and not a diagnosis of idiopathic toe walking.

The breadth of measurement outcomes and variability of assessments in idiopathic toe walking research is challenging, particularly the use of tools without established reliability and validity. It also is problematic as it precludes meta-analysis of outcomes due to the heterogeneity of results. Many researchers in other foot and leg disorders are advocating for a consensus of measures to be used in observational and outcome based research [34]. This present review highlights the potential for those working in ITW research to apply a similar consensus framework for outcome measure selection.

This review has a number of limitations. Firstly, this review was limited to research published in English. Additionally, many of the included studies did not cite the validity or reliability of the tools despite the tools being widely used and considered 'accepted practice'. The reference list of included studies were title searched for validity and reliability articles associated with the included tools or outcomes. These were not found for many of the gait laboratory tools. This study did not extract the protocol used for individual outcome measures unless this was specified as a separate outcome. It is acknowledged that many authors may not cite references for commonly utilized gait laboratory protocols and tools used due to limitations on word limits and reference numbers imposed by a journal.

In this review, the McMaster quality assessment tool was used primarily to identify risk of bias relating to study design, with no specific quality tool applied to the extracted reliability or validity of the tools. Hence, health professionals should consider this limitation when selecting assessment tools and outcome measures in the clinical setting. We would encourage consideration of adopting improved or more reliable measures as they become available to clinicians.

5. Conclusion

Measurement of range of motion and gait pattern are most frequently reported in studies that include children with ITW. Although this review did not aim to provide a recommendation on outcome measures, protocols or equipment for future ITW research, it highlights the potential for consensus regarding the outcome measures for both interventional and observational studies in this population. It also identifies the potential for a standard suite of tools, procedures and protocols for data collection to be identified to enable future treatment trials to embed similar measures, thus facilitating pooling of results across clinical research groups.

Conflict of interest management

CW had two articles included within this review. To manage this conflict during screening and extraction, AC and PM reviewed and extracted all research published by CW that had potential for inclusion according to the inclusion and exclusion criteria.

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