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Measurement properties of instruments for assessment of cervical spine function in infants with torticollis: a systematic review.

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Title Page

Measurement properties of instruments for assessment of cervical spine function in infants with torticollis: a systematic review.

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What is known?

- A thorough assessment of the infant presenting with torticollis is essential, in order to correctly diagnose, rule out ‘red flags’ and manage appropriately
- Assessment tools need to have robust measurement properties in order to be of value for clinical practice and research

What is new?

- This systematic review identified two valid and reliable tools for the assessment of cervical spine function in infants with torticollis
- Further research is required to assess the measurement properties of tools already described in the literature and to develop further tools for use in infants with torticollis

Abstract

The aim of this study was to systematically review the measurement properties of instruments which assess cervical spine function in infants with torticollis. Electronic searches were performed in MEDLINE, CINAHL, Embase, Web of Science and the Cochrane Library, combining three constructs (“torticollis”, “cervical spine assessment” and “measurement properties”). Two reviewers independently rated the methodological quality and the quality of measurement properties of identified articles, using both the COSMIN (**CO**nsensus-based **S**tandards for the selection of health status **M**easurement **IN**struments) checklist and quality criteria for measurement properties. Five studies, using six instruments, met the inclusion criteria and were analysed. Included instruments were the goniometer, electronic pendular goniometer, protractor, still photography, Muscle Function Scale and a range of motion limitation scale. All studies assessed reliability and one study also assessed content validity and hypothesis testing. The methodological quality of the studies varied from poor to excellent according to the COSMIN checklist. Two instruments were found to have good measurement properties from high-quality studies: still photography for the assessment of habitual head tilt in supine and the Muscle Function Scale for the assessment of side-flexor muscle function in lateral head righting.

Conclusion This systematic review identified two reliable tools for the assessment of cervical spine function in infants with torticollis. Further research is required to assess the measurement properties of tools already described in the literature and to develop further tools for use in this population.

Abbreviations

CMT Congenital Muscular Torticollis

COSMIN **CO**nsensus-based **S**tandards for the selection of health status **M**easurement **IN**struments

CSp Cervical Spine

HRPRO Health-Related Patient-Reported Outcomes

MIC Minimal Important Change

PoT Postural Torticollis

ROM Range of Motion

Rot Rotation

SCM Sternocleidomastoid muscle

SDC Smallest Detectable Change

SEM Standard Error of Measurement

SF Side flexion

Introduction

Torticollis is a clinical sign of asymmetric neck function which may result from a variety of underlying disorders. The main cause in infancy is muscular in nature [1-3] and can be divided into two clinical subgroups of Congenital Muscular Torticollis or Postural Torticollis [4,5]. Congenital Muscular Torticollis (CMT) results from fibrosis of the sternocleidomastoid (SCM) muscle, which may result in a fibrotic lump or 'pseudo-tumour' [1]. Depending on diagnostic criteria, the incidence of CMT ranges from 1.3% to 3.9% of new-borns [6,7]. Congenital Muscular Torticollis involves shortening or excessive contraction of the SCM muscle with reduced cervical spine (CSp) ipsi-lateral rotation and contra-lateral side flexion, and is associated with breech presentation, difficult labour and hip dysplasia [8]. Postural Torticollis (PoT) results from asymmetry of the cervical musculature, without any morphological changes, but secondary to asymmetric positioning and use. It is commonly associated with plagiocephaly, which is cranial asymmetry due to external pressure [4,9-11]. Again, incidence depends on diagnostic criteria, but has been reported as high as 16% of new-borns [12].

In both CMT and PoT, asymmetric neck function may be the most obvious clinical feature, but this can have subsequent effects on other aspects of the infant's development, such as cranio-facial, musculoskeletal and neuro-developmental effects [8,13-17]. Whilst muscular dysfunction is the most common cause of torticollis in infancy, there are potentially up to 80 causes [18]. Therefore, a thorough assessment of the infant presenting with torticollis is essential to correctly diagnose, rule out 'red flags' and manage appropriately [19,20]. A number of interventions have been found to be effective [20] but further evaluation is recommended, for which robust outcome measures are indicated.

Although a variety of tools exist for the assessment of CSp function in adults [21-28] and in older children [29-34], these are often not suitable for use in infants. Infants are more variable in behaviour and movement, and less able to follow commands or tolerate equipment [20]. Many studies investigating various aspects of torticollis have included CSp range of motion (ROM) as an outcome measure but give limited detail about how the ROM was measured [35-42]. Others give details of the assessment tool used, but measurement properties are not described [10,12,43-46] or are described for an adult population [47].

Assessment techniques need to have robust measurement properties in order to be of value for both research and clinical practice. The aim of this review was to systematically appraise the evidence on measurement properties of assessment tools/techniques for the measurement of CSp function in infants with torticollis.

Methods

This review is reported in accordance with the PRISMA (**P**referred **R**eporting **I**tems for **S**ystematic Reviews and **M**eta-**A**nalysis) guidelines for the reporting of systematic reviews [48].

Data Sources and Search Strategy

A systematic search of the literature was performed by two independent reviewers (HF and AS). Three groups of key words were used - one group relating to "torticollis", the second relating to "cervical spine assessment" and the third relating to "measurement properties". See Appendix 1 for the full list of search terms. The following electronic databases were searched from inception to September 2016: PubMed, CiNAHL (EBSCO), Embase, The Cochrane Library and Web of Science. Google Scholar was searched for additional citations associated with final included studies. Authors of relevant published abstracts were contacted to request a full-text version of the study and/or study data. Reference lists of selected studies were hand searched for additional studies.

Eligibility Criteria

Studies were included if they were original studies, included infants aged 0-3 years, investigated one or more measurement property of an assessment tool, the population of interest included torticollis (diagnosed using recognised criteria) and were available full-text in a peer-reviewed publication.

Studies were excluded if they were case reports, were not written in the English language, involved medical diagnostic tests, focussed solely on another aspect of torticollis (e.g. plagiocephaly) or involved non-muscular causes of torticollis, such as neurological conditions or congenital bony anomalies.

Data Extraction and Assessment of Methodological Quality

Data extraction and assessment of methodological quality were performed by two independent reviewers (AS and HF). Any disagreements about study eligibility, data extracted, or the quality scoring were discussed and, if possible, reconciled by mutual agreement. In the case of disagreements, a third reviewer (DM) was available for arbitration.

The following data were extracted using a standardised data extraction form: study demographics, details of the tool (name, element of CSp function being assessed and protocol for use), details of the subjects and raters, measurement property being assessed and statistical analysis.

The COSMIN (**CO**nsensus-based **S**tandards for the selection of health status **M**easurement **IN**struments) checklist [49,50] was used to rate the methodological quality of the study designs. This tool was developed as a consensus-based checklist to evaluate the methodological quality of studies on measurement properties and has demonstrated high inter-rater agreement [49]. It assesses 12 measurement properties: internal consistency; reliability; measurement error; content validity; construct validity (i.e. structural validity, hypothesis testing and cross-cultural validity); criterion validity; responsiveness; interpretability; item response theory and generalisability. Each measurement property is evaluated in a separate section, employing a number of questions regarding items of design and statistical methodology, in order to assess the extent to which the study meets the standard for good methodological quality. Four response options (excellent, good, fair and poor) are defined for each question, with an overall quality score obtained by taking the lowest rating of any item in a section i.e. “worst score counts” [50]. The COSMIN checklist was initially developed for use on Health-Related Patient-Reported Outcomes (HRPOs), but its authors indicated that it is also appropriate for use with other forms of measurement tools, although some of the scoring criteria (e.g. on sample size) should be used as guidelines and should be at the discretion of the rater [49,50].

The reviewers also attributed a separate rating to the quantitative results for each study, following the quality criteria for measurement properties reported by Terwee et al. [51]. These criteria, derived from existing guidelines and consensus, contain nine sections which apply a rating of positive (+), negative (−), indeterminate (?) or no information available (0) to the measurement properties: content validity; internal consistency; criterion validity; construct validity; reproducibility; longitudinal validity; responsiveness; floor/ceiling effect and interpretability.

Results

The selection process and search yield are shown in Figure 1. Following title and abstract screening, 15 articles remained. Clarification was sought from three corresponding authors. Two authors [52,53] had made reference to unpublished data and they provided further information regarding this data. The pilot study from one author [52] was included, as a full text version was provided as part of a Master of Philosophy thesis [54] and from the other [53] was excluded as it was presented as a poster format, with no full-text available [55]. The third author [29] was contacted for clarification on the age of subjects, and this study was subsequently excluded as it was not clear whether or not the subjects met the age criteria for inclusion.

Of the remaining 12 full text articles assessed for eligibility, eight were subsequently excluded. Four assessed CSp function as an element of a wider condition [56-59] and four described methods of assessment of torticollis but not assessment of their measurement properties [8,10,47,60]. Therefore, a final selection of five studies [54,61-64] was included in this review. These five studies examined measurement properties of six assessment tools for infants with torticollis: electronic pendular goniometer (digital inclinometer) [54]; standard goniometer [61]; large protractor [61]; Muscle Function Scale [62]; still photography [61,63] and ROM limitation scale [64]. Details are described in Table 1 and in Figures 2 – 7.

Of these five studies, all investigated reliability and one also described content validity and hypothesis testing [62]. Therefore, three of the potential 12 COSMIN sections were used to assess methodological quality. Quantitative results of three properties were rated using quality criteria developed by Terwee et al. [51] (see Table 2).

Figure 1

Table 1

Table 2

Figure 2

Figure 3

Figure 4

Figure 5

Figure 6

Figure 7

Rating of Methodological Quality

Reliability

All five studies assessed reliability (see Table 3). Sample sizes of infants ranged from 23 to 109. In two studies the sample was divided into two groups, so the actual sample sizes in those studies were 12+12 [54] and 11+12 [61]. Numbers of raters ranged from one to nine. Sample size is one of the criteria for which the COSMIN checklist may be used more as a guideline and at the discretion of the review author, rather than as one of the criteria on which to determine the "worst score" [49,50]. In addition, the tool does not differentiate between the sample size of the subjects and that of the raters in a reliability study. Therefore, in some systematic reviews of physical measures using the COSMIN checklist, the sample size score has been excluded [65,66] and a similar approach was taken in this review.

Administration was considered to be independent in four of the studies [54,62-64] as the examiners were unaware of the previous assessments or there was an adequate time interval between assessments to minimize risk of recall. In Klackenberg et al. [61] it was considered by the review authors that the administration was not independent for Group 1, as they were re-measured without any time interval, and doubtful for Group 2, as there was only a one-hour time interval and considering that the sample was small, there was a chance that the examiner would recall previous measurements.

In all five studies it was either stated or presumed that patients were stable between test administrations and test conditions were similar for both test administrations. The studies scored '*good*' [54,61] or '*excellent*' [62,63] in relation to reliability statistics, however Murgia et al. [64] scored as '*fair*' due to use of Kappa statistics instead of the preferred weighted Kappa.

Other minor methodological flaws in the design or execution were identified by the review authors. In the study by Chen [54] only the healthy side was assessed, which meant that a limited range was not assessed. Klackenberg et al. [61] did not specify whether the measurements were taken to the nearest 1°, 5° or 10°. However, the illustration of the protractor which had been fabricated (see Figure 3) suggested that it was in 10° intervals (0-90° bilaterally). In two of the studies measuring CSp rotation, the infants' heads were not over the edge of the surface, so true end of range may not have been achieved [61,64]. Murgia et al. [64] used scales with wide intervals for ROM. Range of motion is usually measured in degrees, with means of 110° CSp passive rotation and 70° passive side-flexion reported in infants [67]. In Murgia's scale [64], CSp rotation was divided into four measurement levels and side-flexion into three measurement levels (see Figures 4 and 5). In addition, CMT was one of the exclusion criteria, so only cases of PoT would have been included. There were no important flaws in the other studies [62,63].

One reliability checklist was completed for each element of the reliability study e.g. for inter-rater reliability and for intra-rater reliability (available as Online Resource). The quality scores (using "worst score counts") allocated to the five reliability studies, both including and excluding that for sample size, are described in Table 3.

Content Validity and Hypothesis Testing

One study assessed content validity [62] in which a panel of experts discussed the *Muscle Function Scale* and provided insight, constructive criticism and advice. Most of the items in the checklist were rated as '*excellent*' but the overall score was '*poor*' due to lack of details regarding how this was done (see Table 4). It was also discussed by the review authors that still photographs were used to assess the infants, whereas the MFS is a dynamic measure, as the position needs to be held by the infant for ≥5 seconds in order to obtain a score. Therefore, the infants' head position was being analysed but not the length of time the position was being held.

One study examined hypothesis testing [62]. A number of the items on the hypothesis testing checklist are similar to that of the reliability checklist [49]. Specific hypotheses to be tested should have been formulated and include expected differences between groups or expected correlations between instruments, as well as expected direction and magnitude of the correlations *a priori* (before data collection). This was unclear in the study by Öhman et al. [62], therefore it was given a score of '*poor*' (see Table 4).

Table 4

Rating of Quantitative Results

All five studies reported ICC or Kappa Values >0.7, consistent with a positive reliability rating score, except for the side-flexion component of the ROM limitation scale [64] which had a negative reliability rating (see Table 3). Agreement was reported in one study [61] in which measurement error was expressed as the standard error of measurement (SEM), but minimal important change (MIC) and smallest detectable change (SDC) were not reported. However, the study authors had made a case that ≤6°repeatability was clinically acceptable. Therefore, in that study [61] the goniometer and protractor were given positive reliability rating scores and photography a negative reliability rating score. The one study which reported content validity [62] was found to have a positive rating. See Table 3 for details.

Discussion

This systematic review identified six assessment tools measuring CSp function in infants with torticollis, for which measurement properties had been evaluated: standard goniometer, electronic pendular goniometer, large protractor, a ROM limitation scale, the *Muscle Function Scale* and still photography.

Goniometer

A goniometer is a routinely used tool and is widely available in the clinical setting. Reliability was rated as positive (ICC>0.7) in the one study [61] which used it, assessing intra-rater reliability of passive CSp rotation, but methodological quality rated as poor, primarily due to the short time interval between administrations.

Electronic pendular goniometer

An electronic pendular goniometer (digital inclinometer) is also available for clinical use, although more expensive than a standard goniometer. Reliability was rated as positive (ICC>0.7) in the one study [54] which used it, assessing inter-rater and intra-rater reliability of passive CSp rotation, but methodological quality rated as fair. Two examiners and one assistant were required for the assessment procedure, which may not be clinically feasible.

Protractor

A mat with protractor markings, which was assessed in one study [61] appeared relatively simple to develop. Alternative large joint protractors are available commercially (e.g. Baseline 12-1076 arthrodial goniometer; Reedco arthrodial protractor). Reliability was rated as positive (ICC>0.7), assessing intra-rater reliability of passive CSp side-flexion, but methodological quality rated as poor, again primarily due to the short time interval between administrations.

Range of motion limitation scale

This scale [64] assessed passive CSp rotation/side-flexion and active CSp rotation. The validity of the scale was not tested. Reliability was rated as positive (Kappa>0.7) for rotation and negative (Kappa<0.7) for side-flexion, but methodological quality rated as fair. One of the limitations of this scale identified by the review authors was the wide categories of ROM, which may limit the clinical utility of the scale. However, it should be noted that it was developed as a simple method of detection of limitation of CSp ROM in infants with plagiocephaly (i.e. PoT) and not for precise measurements.

Muscle Function Scale

This scale [62] assessed CSp side-flexor muscle function in lateral head righting, by rating the position of the head when the infant's body was moved from a vertical to a horizontal position. Regarding content validity, very little information was given about the methodology of testing, however a panel of experts provided insight, constructive criticism and advice, with subsequent alterations having been made. The reliability of the scale was rated as positive (Kappa>0.7) and the methodological quality of the study as excellent. This scale is free to use clinically.

Still photography

Still photography (with measurements subsequently assessed using lines drawn on the photograph) was used in two studies [61,63]. Rahlin and Sarmiento [63] used it to assess habitual head deviation from midline (head tilt) in supine. Reliability was rated as positive (ICC>0.7) and the methodological quality of the study as excellent. Klackenberg et al. [61] used it to assess CSp passive rotation and side-flexion, by a photograph taken at the end of range position. In comparison to measurements taken simultaneously with a goniometer/protractor, its reliability was found to be positive (ICC>0.7), but agreement between the two tools was poor and the study quality rated as fair. Still photography is a simple, practical assessment tool. In one study [63] it was performed by one examiner, whereas in the other [61] two assistants were required in addition to the examiner.

Implications for Practice and Research

Torticollis is a relatively common condition of infancy presenting to healthcare professionals [6,7,12]. However, this systematic review has revealed only two measurement tools which have both good measurement properties and study methodological quality. These include still photography for the assessment of habitual head tilt in supine and the *Muscle Function Scale* for the assessment of side-flexor muscle function in lateral head righting. When using an assessment tool, the measurement properties of that tool should be considered, particularly if the tool is being used as a decision-making outcome measure. There has been some inconsistency in the literature [67-73] where reliability values from Klackenburg et al. [61] and Cheng et al. [8,52] (i.e. the pilot study by Chen [54]) are referred to in the measurement of CSp ROM, but the tool used in the subsequent studies does not appear to be the same as the ones in which the measurement properties were originally evaluated. In these subsequent studies [8,52,67-73] an arthrodial protractor is used to measure passive CSp rotation and side-flexion, whereas the original reliability studies were performed on an electronic pendular goniometer [54] and a mat with protractor markings [61]. Based on the COSMIN checklist, both of these studies [54,61] were found to be of poor/fair quality, which raises issues regarding the validity of outcomes assessed in those subsequent studies.

Sample sizes tended to be small, scoring as poor in two [54,61] and fair in one [63] of the studies. The number of raters was not taken into consideration by the COSMIN tool. It had been decided to exclude sample size in the final quality score allocated, which did not change the overall score for two of the studies [61,64] but improved the scores for the other three studies [54,63,68]. A challenge exists in performing reliability studies with infants, as repeated measures and a larger number of raters may cause distress to the infants.

A number of studies were not included in the systematic review but indicate potential for further research [8,12,47,52,57,58,60,64,65,66]. Some of these studies were excluded as CSp function was assessed as a section of an overall assessment for another condition: active CSp rotation in supine and prone were assessed on a six-point scale as part of an Idiopathic Infantile Asymmetry Scale [57] and head tilt in upright was assessed on a four-point scale as part of a Severity Assessment of Plagiocephaly Scale [58]. There is potential for the CSp sections to be analysed separately. The following excluded studies described methods of assessment of torticollis, but not assessment of the measurement properties: Emery [47] described use of a modified goniometer (with a spirit level) but measurement properties were not analysed. However, the reliability of similar instruments had previously been tested in adults [74] and could be tested in infants. Stellwagen & Hubbard [12] used graph paper under the heads of infants to assess passive CSp side-flexion, which could potentially have its measurement properties assessed. Visual estimation was used in a number of studies [64-66], which has been tested for reliability in adults [24,27,75] but not in infants. Studies have reported that over 90% of paediatric physiotherapists use visual estimation to assess cervical function in infants with torticollis [76-78]. Therefore, the measurement properties of visual estimation should be studied further. Cheng et al. [8,52,60] used an overall severity scoring system for torticollis, consisting of six objective and subjective categories, but measurement properties were not described. However, subsequent to this systematic review, a study has been published [81] which examined the reliability of a seven-grade severity system, the Congenital Muscular Torticollis Severity Classification System (CMT-SCS) and found good inter-rater (ICC 0.83) and intra-rater (ICC 0.81) reliability (n=24 infants, 145 raters).

An assessment tool must be clinically practical as well as having adequate measurement properties in order for it to be used, especially in the case of assessing infants [20,79]. One of the studies which had been excluded from this review as it was only in abstract form [55] reported excellent reliability (ICC 0.89-0.97) of 2D video analysis in the measurement of CSp active ROM in infants, with rotation but not side-flexion being comparable to 3D motion analysis (considered the gold standard). However, a subsequent study [53] found the method to be clinically unfeasible in terms of time requirements. In the assessment of infants, it may be practical to have one assistant, as a parent is usually present, but in two of the studies analysed, more assistants were required for use of the electronic pendular goniometer [54] and photography [61].

Of the 12 sections of the COSMIN checklist, reliability was the most common measurement property evaluated, with content validity and hypothesis testing tested in just one study. Other measurement properties such as responsiveness and interpretability should be evaluated in future studies, if such tools are used for intervention studies and clinical decision making.

Strengths and Limitations of the Review

This systematic review adds to other authors' works, which have identified tools used in the assessment of CSp function in infants with torticollis, but not systematically reviewed their methodological quality [20,80]. A strength of this review was the use of the COSMIN checklist for the assessment of methodological quality of studies assessing psychometric properties of measurement tools. Two independent review authors extracted data and assessed methodological quality.

The inclusion/exclusion criteria may have limited the number of articles included in this review, such as those not in the English language. The review was limited to full-text studies, so other relevant studies may have been excluded. Tools tested on infants with other types of torticollis (e.g. ocular) may be applicable to muscular torticollis but were excluded.

The COSMIN checklist was developed for HRPROs, therefore its validity for assessing the methodological quality of these clinometric studies can be questioned. It also generates a floor effect in terms of its "lowest score counts", so that studies which may have scored well on many items will result in a '*poor'* or '*fair'* overall score due to one inadequate item, even when sample size was excluded.

Conclusion

This systematic review has analysed five studies, in which measurement properties of six tools for the assessment of CSp function in infants with torticollis were evaluated. The quality of the studies, assessed using the COSMIN checklist, varied from poor to excellent. The only tools which were found to have good measurement properties from a high-quality study were: still photography for the assessment of habitual head tilt in supine and the *Muscle Function Scale* for the assessment of side-flexor muscle function in lateral head righting. Further high-quality research is required to investigate the measurement properties of assessment tools which have been describedin the literature, but not tested, and also to develop further tools for the assessment of CSp function in this population.

Compliance with Ethical Standards

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Conflict of interest: All authors declare that they have no conflict of interest.

Ethical approval: This article does not contain any studies with human participants or animals performed by any of the authors.

Conflict of interest

The authors declare no conflict of interest

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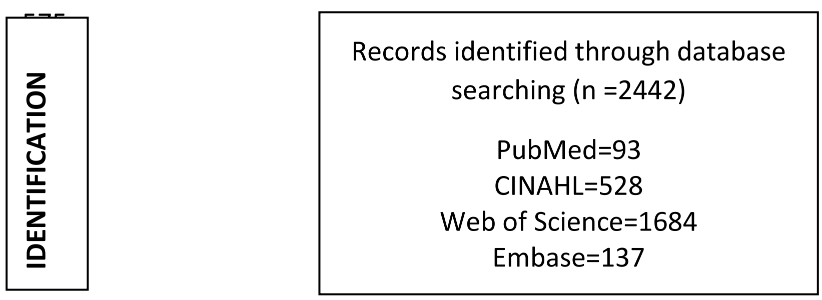


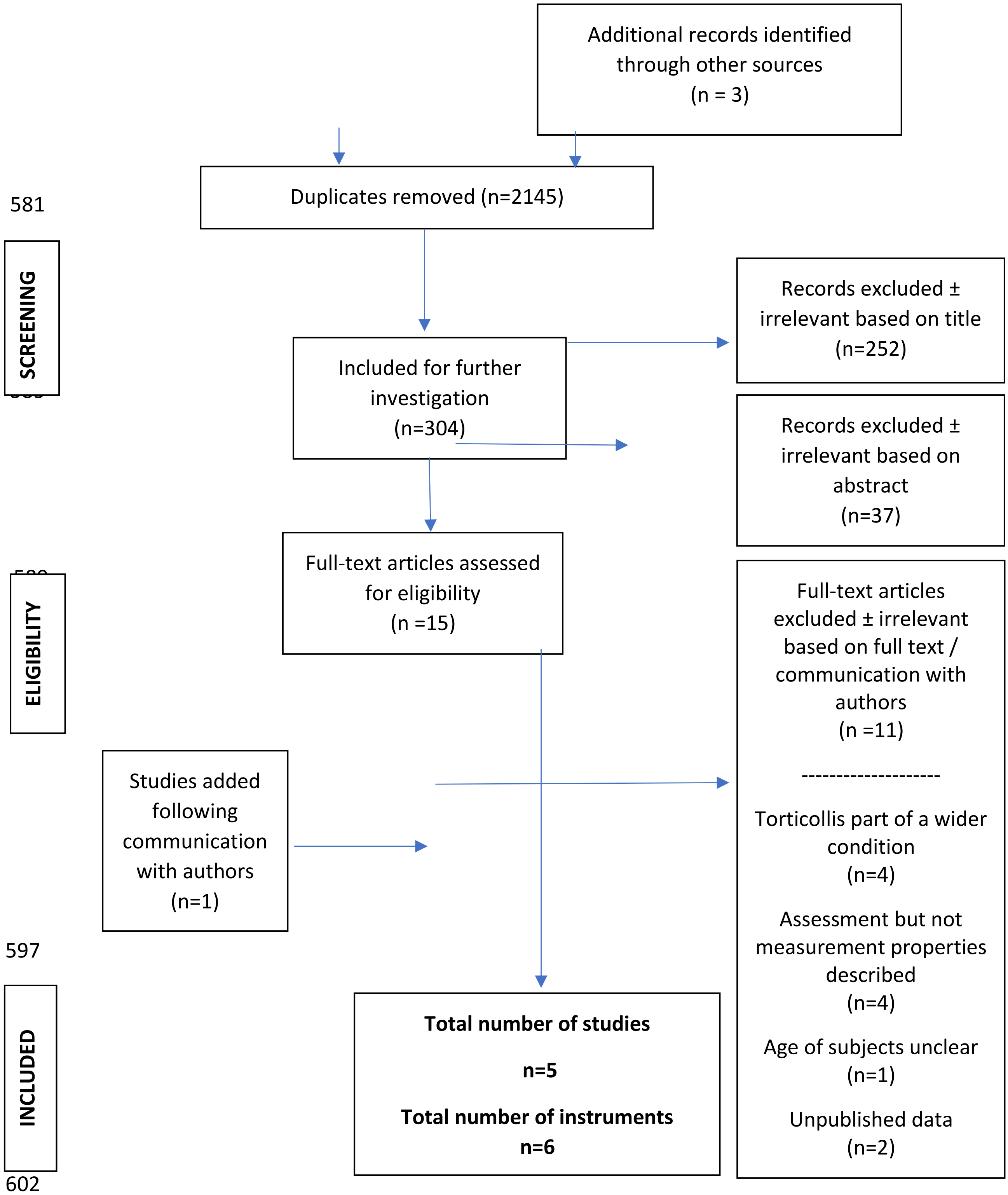


Fig. 1 Flowchart of study selection process and search yield

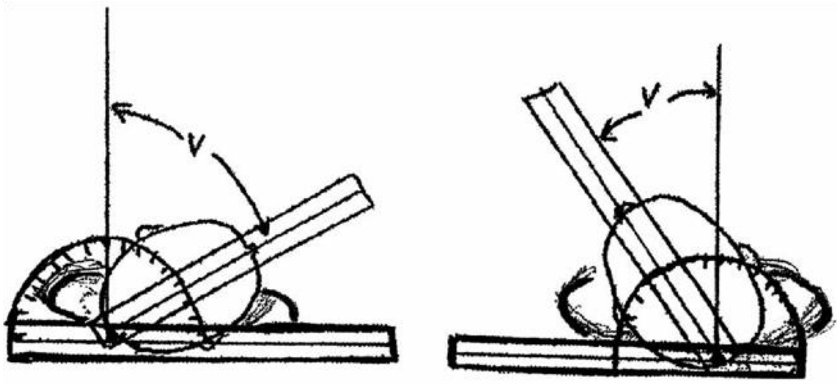


Fig. 2 Cervical rotation measured with goniometer placed above the infant's head. Reproduced from Klackenberg et al. [61] with permission from the publisher (Taylor & Francis Ltd., http://www.tandfonline.com)

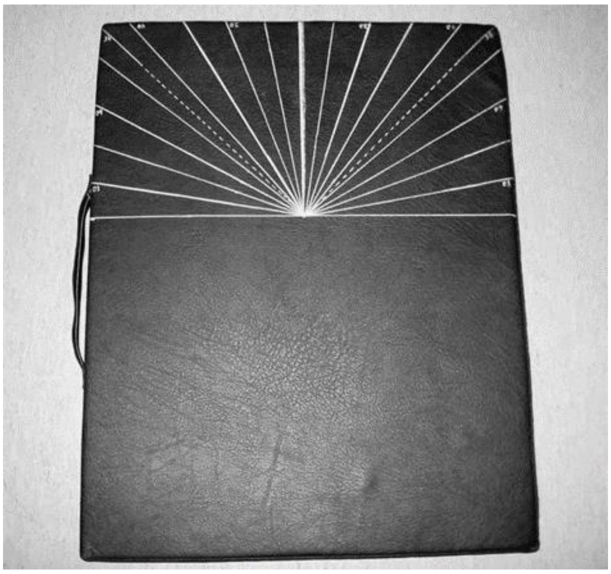


Fig. 3 Protractor used to measure cervical side flexion. Reproduced from Klackenberg et al. [61] with permission from the publisher (Taylor & Francis Ltd., http://www.tandfonline.com)

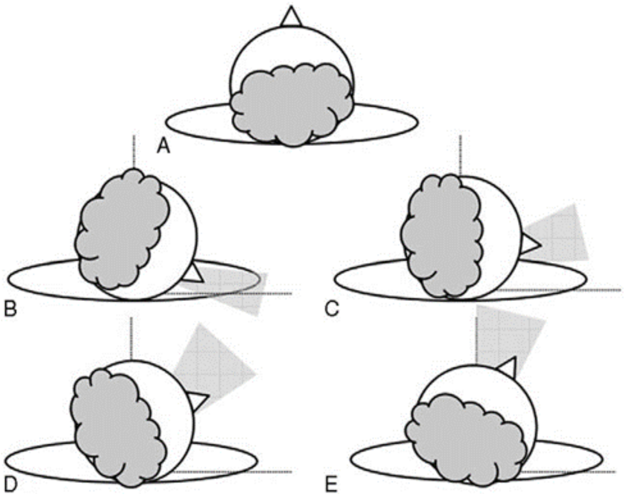


Fig. 4 Range of Motion Limitation Scale - active and passive cervical rotation (A) Neutral position (B) Full ROM (C) Mild limitation (D) Moderate limitation (E) Severe limitation. Reproduced from Murgia et al. [64] with permission from the publisher (Wolters Kluwer Health Inc., https://wolterskluwer.com)

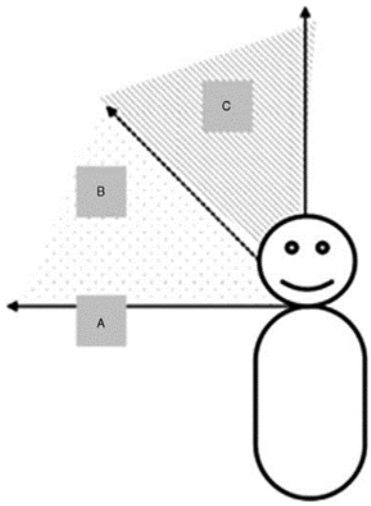


Fig. 5 Range of Motion Limitation Scale - passive cervical side flexion (A) Full range of motion (B) Moderate limitation (C) Severe limitation. Reproduced from Murgia et al. [64] with permission from the publisher (Wolters Kluwer Health Inc., https://wolterskluwer.com)

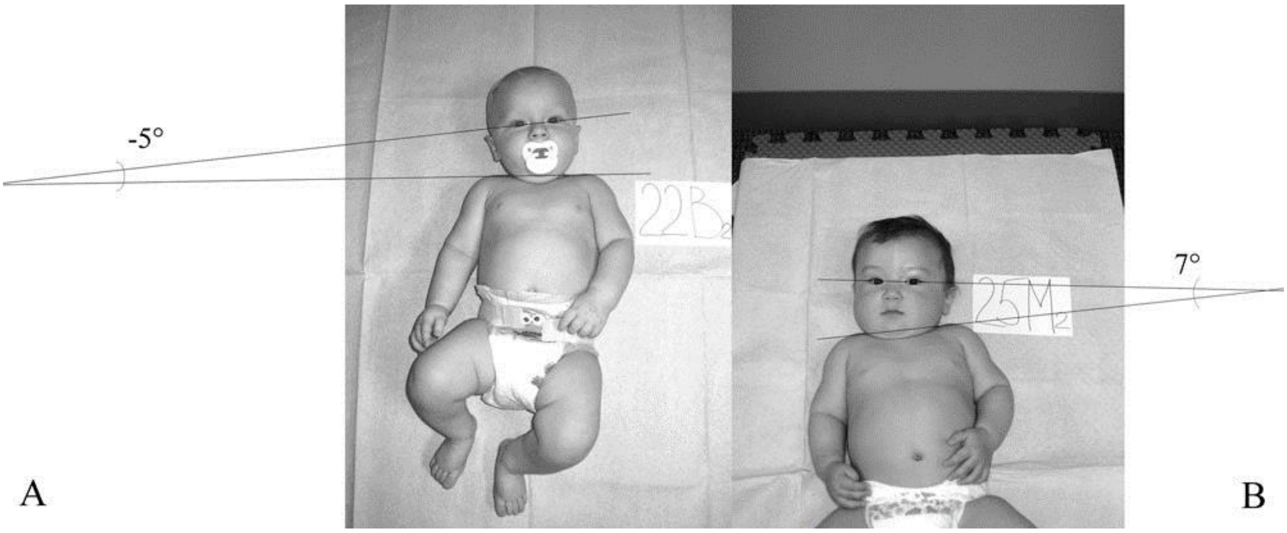
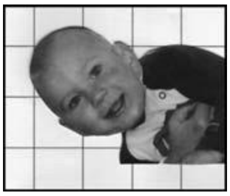


Fig. 6 Still photography measurement procedure, with lines drawn through the eyes and acromion processes using a ruler and a pencil (A: Participant number 22, habitual head-tilt angle measured at −5◦, negative value indicates head tilt to the right; B: Participant number 25, habitual head-tilt angle measured at 7◦, positive value indicates head tilt to the left). Reproduced from Rahlin & Sarmiento [63] with permission from the publisher (Wolters Kluwer Health Inc., https://wolterskluwer.com)

5. >75° (Head more than 75° above the horizontal line).

4. >45°–<75° (Head more than 45° and less than 75° above the horizontal line).



3. >15°–<45° (Head more than 15° and less than 45° above the horizontal line).

2. >0°–<15° (Head more than 0° and less than 15° over the horizontal line).

1. 0° (Head on the horizontal line at 0°).

0. <0° (Head below horizontal line less than 0°).

Fig. 7 Muscle Function Scale (MFS) for Infants (Version II). Reproduced from Öhman et al. [62] with permission from the publisher (Taylor & Francis Ltd., http://www.tandfonline.com)

Table 1: Characteristics of Included Studies

Study	Tool(s)	Measurement Property	Sample size	Diagnosis (Age)	Observers	Measure (protocol)	Time interval	Results
Chen [54]	Electronic pendular goniometer	Reliability -Inter-rater -Intra-rater	Group I: n=12 (inter-rater) Group II: n=12 (intra-rater)	CMT (0-12 mo)	2 PTs (+assistant)	Passive CSp rot (infant supine, HOEOS, body stabilised, head aligned midline & position recorded, movement performed & EOR recorded, used landmarks of interorbital line to 7th spinal process / posterior edge of ears, only non-affected side tested)	1 hour (inter-rater) 2-3 weeks (intra-rater)	ICC 0.87-0.91 (Inter-rater) ICC 0.71-0.79 (Intra-rater)
Klacken-berg et al. [61]	Goniometer and adapted protractor (made from rectangular surface, 75x60x9 cm, with lines marked out in 10° intervals) *See Fig. 2&3*	Reliability -Intra-rater	Group I: n=11 Group II: n=12	CMT (1-5 mo)	1 PT (+assistant)	Passive CSp rot (infant supine, body stabilised, head/body aligned, goniometer above infant's head aligned with nose, rot performed, moveable arm of goniometer realigned with nose & angle recorded) Passive CSp SF (infant supine on protractor, body	Group I: immediately Group II: 1 hour	Group I: ICC 0.97-0.99 SEM 2-5° Group II ICC 0.77-0.95 SEM 4-6°

						stabilised, head/body aligned, SF performed & angle recorded – ? landmarks used)		
	Still photography (plus goniometer)	-Photographs versus the goniometer/ protractor readings taken simultaneously	Groups combined n=23		1 PT (+2 assistants)	Passive CSp rot Passive CSp rot (Digital photographs taken in standardised position at EOR of above, markers on os frontale in line with nose & tip of nose, goniometer/protractor edited out of photograph, angles measured with goniometer on the photograph).		ICC 0.74-0.90 SEM 6-10° Photograph readings 2-3° lower than goniometer/protractor readings performed on infant.
Murgia et al. [64]	ROM limitation scale‡ *See Fig. 4&5*	Reliability -Inter-rater -Intra-rater	n=109	Plagio-cephaly/ being assessed for PoT (0-18 mo)	2 Physicians	Active and passive CSp rot, passive CSp SF (infant supine, body stabilised, infant stimulated to rot CSp actively & position scored, gentle over-pressure given & position scored, passive SF performed & position scored)	1-24 hours (inter-rater) 48 hours (intra-rater)	Kappa 0.80 active rot; 0.83 passive rot; 0.49 passive SF (Inter-rater) Kappa 0.72 active rot; 0.73 passive rot; 0.41 passive SF (Intra-rater)

Rahlin & Sarmiento [63]	Still photography (plus protractor) *See Fig. 6*	Reliability -Inter-rater -Intra-rater	n=30 (2 photo-graphs of each subject taken by 2 investigators = 120 photo-graphs)	CMT (4-16 mo)	2 PTs	Habitual CSp SF (infant supine, visual stimulus at midline, photographs taken from above, lines drawn through lateral corners of eyes/ acromium processes & extended until intersect, then angle measured with protractor)	Weekly intervals x 4	ICC 0.72-0.99; Pearson r 0.72-0.99 (Inter-rater) ICC 0.79-0.84; Pearson r 0.80-0.85 (Intra-rater)
Öhman et al. [62]	Muscle Function Scale† Version I* Version II** *See Fig. 7*	Reliability -Inter-rater -Intra-rater	n=68 photographs	CMT (infants, age not stated)	2 groups of 7 PTs + 2 PTstudents	Active CSp SF (observers shown the photographs of infants with their bodies supported in a horizontal position & their heads in different positions of lateral head righting. One group scored the infants using Version I and the other group using Version II of the scale).	Minimum 1 week (photographs in a different order)	Kappa 0.96; ICC 0.92-0.96 (Inter-rater Version I) Kappa 0.96; ICC 0.90-0.95 (Inter-rater Version II) Kappa 0.96-0.99; ICC 0.94-0.98 (Intra-rater Version I) Kappa 0.96-0.99; ICC 0.93-0.97 (Intra-rater Version II)
		Content validity			Panel of 5 experts	MFS (Versions I & II) discussed and panel provided insight, constructive criticism & advice.		Panel of experts reported validity, recommending a change in scoring system from 5 to 6 levels and

				standardising holding position of infants.

†Muscle Function Scale = an ordinal visual scale that interprets lateral head righting through a categorical scale of 0-5 (previously 0-4).
*Version I: described mainly in words; **Version II: described mainly in degrees

‡ ROM limitation scale: an ordinal visual scale that categorises CSp Rot into full (chin beyond shoulder), mild limitation (chin reaches shoulder), moderate limitation (chin goes beyond mid-clavicle) or severe limitation (chin does not go beyond mid-clavicle); and categorises CSp SF into full (ear to shoulder), moderate limitation (ear goes beyond 45°) or severe limitation (ear does not go beyond 45°).

CMT, Congenital Muscular Torticollis; CSp, cervical spine; EOR, end of range; HOEOS, head over edge of surface; ICC, intra-class coefficient; mo, months; PT, Physiotherapist; PoT, Postural Torticollis; Rot, rotation; SEM, Standard Error of Measurement; SF, side-flexion

Table 2: Quality criteria for rating the results of measurement properties (relevant sections taken from Terwee et al. 2007)

Property	Quality Criteria	Rating
Reliability	ICC or Weighted Kappa ≥ 0.70	+
	ICC or Weighted Kappa < 0.70	-
	Doubtful design or method	?
	No information found on reliability	0
Measurement Error	MIC<SDC or MIC outside LOA or convincing arguments that agreement is acceptable	+
	MIC ≥ SDC or MIC equals or inside LOA	-
	MIC not defined and no convincing arguments that agreement is acceptable	?
	No information found on agreement	0
Content Validity	A clear description is provided of the measurement aim, the target population, the concepts that are being measured, and the item selection AND target population and (investigators OR experts) were involved in item selection	+
	No information found on target population involvement	-
	A clear description of above-mentioned aspects is lacking OR only target population involved OR doubtful design or method	?
	No information found on target population involvement	0

+, positive rating; -, negative rating; ?, indeterminate rating; 0, no information available

ICC, intra-class coefficient; LOA, limits of agreement; MIC, minimal important change; SDC, smallest detectable change; SEM, Standard Error of Measurement.

Table 3: Summary of results of the reliability-related studies (n=5)

Clinical Tool *(Assessing)*	N	**Measurement Property Evaluated**	**Results (quality criteria rating)**				**COSMIN Worst Score**	**Worst Score Item(s)**	**COSMIN Worst Score (excl. sample size)**
			ICC	**Pearson r**	**SEM**	**Kappa**			
Goniometer [61]	11*	Intra-rater reliability	0.98-0.99 (+)		2-3° (+)		Poor	Sample size	Poor
(Passive rot in supine)	12**		0.77-0.79 (+)		5-6° (+)			Independence of administration	
								Time interval between administrations	
Electronic pendular goniometer [54]	12	Inter-rater reliability	0.87-0.91 (+)				Poor	Sample size	Fair
(*Passive rot in supine)*	12	Intra-rater reliability	0.71-0.79 (+)						
Protractor [61]	11*	Intra-rater reliability	0.97-0.98 (+)		5° (+)		Poor	Sample size	Poor
(Passive SF in supine)	12**		0.94-0.95 (+)		4-6° (+)			Independence of administration	
								Time interval between administrations	
Muscle Function Scale† [62]	68	Inter-rater reliability					Good	Sample size	Exc
(Active SF in horizontal)		Version I	0.92-0.96 (+)			0.96 (+)			
		Version II	0.90-0.95 (+)			0.96 (+)			

		Intra-rater reliability							
		Version I	0.94-0.98 (+)			0.96-0.99 (+)			
		Version II	0.93-0.97 (+)			0.96-0.99 (+)			
Still photography + protractor [63]	40	Inter-rater reliability	0.72-0.99 (+)	0.72-0.99 (+)			Fair	Sample size	Exc
		Intra-rater reliability	0.79-0.84 (+)	0.80-0.85 (+)					
(*Postural SF in supine)*									
Still photography + protractor [61]	23	Intra-rater reliability	0.79-0.88 (+)		8-10° (-)		Poor	Sample size	Fair
(*Passive SF in supine)*									
Still photography + goniometer [61]			0.74-0.90 (+)		6-8° (-)				
(*Passive rot in supine)*									
ROM Scale‡ [64]	109	Inter-rater reliability				0.80 A rot. (+) 0.83 P rot. (+) 0.49 P SF. (-)	Fair	Handling of missing items	Fair
(Active rot, Passive rot, Passive SF in supine)		Intra-rater reliability				0.72 A rot. (+) 0.73 P rot. (+) 0.41 P SF. (-)		Flaw in design Kappa not weighted	

* Group I: remeasured immediately ** Group II: remeasured after one hour +: positive rating -: negative rating

†Muscle Function Scale = an ordinal visual scale that interprets lateral head righting through a categorical scale of 0-5

‡ ROM limitation scale: categorised rot into full, mild limitation, moderate limitation or severe limitation; categorised SF into full, moderate limitation or severe limitation.

A, active; Exc, Excellent; excl, excluding; P, passive; rot, CSp rotation; SF, CSp side-flexion

Table 4: Summary of results for validity related and hypothesis testing study (n=1)

Clinical tool (*Assessing)*	N	Measurement property evaluated	Results	Study Quality Rating	COSMIN worst score count	COSMIN worst Score item
Muscle Function Scale† [62] *(Active SF in horizontal)*	68	Content validity	A panel of experts, consisting of five paediatric physiotherapists experienced in the assessment of muscle function in children (mean 23 years, range 12-33 years professional experience), discussed the scale and provided insight, constructive criticism and advice. This included changing the scale from five to six ordered scores and testing both the original version and a revised version (see Table 2 for details)	+	Poor	Flaw in design (lack of detail given)
		Hypothesis testing			Poor	Lack of formation of *a priori* hypotheses Constructs and measurement properties of comparator instruments not described Flaw in design (lack of detail given)

†Muscle Function Scale = an ordinal visual scale that interprets lateral head righting through a categorical scale 0-5

Appendix 1: Search strategy for systematic review using three groups of keywords, “torticollis”, “cervical spine assessment” and “measurement properties”.

TORTICOLLIS		**CERVICAL Sp ASSESSMENT**		**MEASUREMENT PROPERTIES**
Neck (MeSH) **OR** Neck Muscles (MeSH) **OR** Cervical Vertebrae (MeSH) **OR** Neck **OR** Cervical **OR** Cervical spine **OR** Torticollis (MeSH) **OR** Torticollis **OR** Congenital Muscular Torticollis **OR** Fibromatosis Colli **OR** Wryneck **OR** Plagiocephaly, Nonsynostotic (MeSH) **OR** Positional plagiocephaly **OR** Deformational plagiocephaly **OR** Non-synostotic plagiocephaly	**AND**	Range of motion, articular (MeSH) **OR** Joint Flexibility **OR** Joint Range of Motion **OR** Passive Range of Motion **OR** Active Range of Motion **OR** Movement **OR** Muscle Strength (MeSH) **OR** Muscle Strength **OR** Posture (MeSH) **OR** Posture **OR** Symptom assessment (MeSH)	**AND**	Reproducibility of Results (MeSH) **OR** Validation Studies (MeSH) **OR** Outcome Assessment, Health Care (MeSH) **OR** Valid* **OR** Reliab* **OR** Repeatab* **OR** Reproducibility **OR** Psychometric* **OR** Clinimetric*