

SYSTEMATIC REVIEW

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Efficacy of non-surgical, non-pharmacological treatments for congenital muscular torticollis: a systematic review and meta-analysis

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Abstract

Background Congenital Muscular Torticollis (CMT) is the third most common musculoskeletal condition in infancy, and if untreated can lead to significant disability. While a range of conservative treatments are commonly used in the management of CMT, an understanding of their efficacy and safety is limited. This systematic review and meta-analysis, without language or discipline restriction, was conducted to address this knowledge gap.

Methods Electronic searches of CENTRAL, PubMed, 22 other electronic databases, three trials registers and Google Scholar, were conducted for randomised controlled trials, which examined any non-surgical, non-pharmacological interventions, including but not limited to manual treatments, movement therapy, acupuncture, adjunctive therapies and physical support, in children aged 0 to 5 years with CMT. Two reviewers independently assessed the risk of bias of the included studies using the Cochrane Risk of bias 1 tool, rated their certainty of evidence using grading of recommendations assessment, development and evaluation (GRADE) framework, and performed random-effects meta-analyses.

Results One hundred studies (80 from China) involving 8125 participants published between 1990 and 2023 were included. Adding manual therapy to an active control resulted in short-term improvements in passive cervical rotation (odds ratio (OR) 9.79, 95%CI 4.26,22.50), passive cervical lateroflexion (OR 2.66, 95%CI 1.17,6.04), active cervical rotation (OR 3.94, 95%CI 1.08,14.35), symmetric head posture (OR 4.55, 95%CI 2.57,8.05), sternocleidomastoid tumour thickness (mean difference (MD) -2.12 mm, 95%CI -2.98,-1.26) and development of symmetrical movement (standardised MD -0.70, 95%CI -0.95,-0.45). The addition of an electrophysical agent to an active control reduced sternocleidomastoid tumour thickness (MD -2.03 mm, 95%CI -2.67,-1.39) and optimised Tuina reduced tumour thickness more than traditional Tuina (MD -1.20 mm, 95%CI -1.80,-0.59). Adverse events were uncommon but poorly reported, with 71 (71%) of studies providing no data. Study heterogeneity limited pooling of data for meta-analysis, and there was very low to low certainty evidence for all results, due to high risk of bias, small sample sizes and study heterogeneity.

Conclusions This review found that non-surgical, non-pharmacological treatments may be effective for CMT, but the certainty of evidence is very low to low. These findings are important in informing clinical guidelines

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and management for CMT and highlight an urgent need for large definitive trials that address the limitations of current studies.

Protocol registration Cochrane Database of Systematic Reviews (No.: CD012987).

Keywords Conservative treatment, Safety, Infant, Manual therapy, Electrophysical agents, Traditional Chinese Massage

Background

Congenital Muscular Torticollis (CMT) is the third most common musculoskeletal condition in infancy. It is characterised by tightness or weakness of one sternocleidomastoid (SCM) muscle, a unilaterally impaired lateral head-righting reflex, and in some cases a palpable SCM tumour. It has a prevalence of up to 16% in those aged 0 to 12 months, [1] and there has been an increased incidence of infant referrals for CMT over the last 30 years, which may be attributable to parents avoiding prone positioning in response to the American Academy of Pediatrics 'Safe to Sleep' campaign. Avoidance of positioning in prone is believed to delay resolution of some types of CMT [2–4]. CMT may also be associated with hip dysplasia [5, 6] and be a part of a larger syndrome of postural asymmetry involving spinal convexity. [7, 8] The condition can lead to reduced range of movement, [9] craniofacial asymmetries, [2] secondary cervicothoracic scoliosis, [10] and motor developmental delays [11, 12]. A range of non-surgical and non-pharmacological interventions, such as manual treatments, movement therapy, acupuncture, adjunctive therapies and physical support, are available for the management of CMT, [7, 8, 13–20] each with potential benefits and harms [15, 17, 21, 22]. However, the efficacy and safety of these treatments for CMT is unclear.

Clinical guidelines for the treatment of CMT provide recommendations for both first choice and supplemental treatments [17]. While these guidelines are based on nine randomised controlled trials (RCTs) and a number of observational studies, they are not informed by meta-analyses and are limited to English-language studies from a single discipline (physical therapy). A number of systematic reviews have also examined the evidence for non-surgical and non-pharmacological treatments for CMT, however these have focussed on a specific intervention across professions, [23–25] one or more interventions within a profession, [26–31] or have considered interventions across two or more professions, but restricted searches to English-language databases [15, 32]. Moreover, RCTs examining the use of high-velocity, low-force manipulation for CMT have reported differing results, with one trial finding no improvements in a range of outcomes, [16] resulting in clinical guidelines and reviews suggesting there is no evidence of efficacy, [17,

33] whereas recent studies have found a range of benefits for individuals with CMT [34–37]. Moreover, of the systematic reviews examining conservative treatments for CMT, only one has conducted a meta-analysis and this examined Tuina and stretching [27]. There is no systematic review with meta-analyses that has comprehensively examined the efficacy and safety of non-surgical and non-pharmacological treatments for CMT. The aim of this review was to address this gap in our knowledge.

Methods

This systematic review examining the safety and efficacy of non-surgical, non-pharmacological therapies for CMT was conducted according to the PRISMA 2020 guidelines, [38] with the protocol registered in the Cochrane Database of Systematic Reviews (No.: CD012987). [39] One reviewer (one of EC, HL, JA, MA) searched CENTRAL, including the Cochrane Developmental, Psychosocial and Learning Problems Specialised Register, PubMed, Embase, 22 other electronic databases and three trials registers from inception to August 2023 (Additional file 1). Reference lists, Google and Google Scholar, were searched and researchers were contacted to obtain unpublished data. Keywords included terms and synonyms for torticollis, paediatrics and RCTs. Searches were developed by three University Health Science Librarians and are provided in Additional file 1. Search results were exported to Covidence for screening [40].

Eligibility criteria

Studies that met the following criteria were included:

- (1) RCTs, quasi-RCTs and cluster -RCTs, irrespective of publication status or language, that recruited participants from birth to five years of age with CMT, but not other forms of torticollis. CMT is present at birth, usually diagnosed in the first months of life, and may result from intrauterine positioning and/or development. In contrast, studies that included participants with non-muscular congenital causes of torticollis, acquired torticollis (which can develop at any age after birth and secondary to conditions such as skeletal anomalies, trauma or infections), neurological torticollis, ocular torticollis and vestibular torticollis were excluded. [41] Participants were

assessed by a clinician as having any of three categories of CMT, (i) SCM tumour, [14] (ii) muscular torticollis, [14] (iii) SCM imbalance or postural CMT, which may be a part of a postural asymmetry [2, 14] and may also be described as positional preference; [16, 42–44]

- (2) Any non-surgical and non-pharmacological intervention for CMT including but not limited to (i) manual therapies (stretching hypertonic muscles, massage including soft tissue mobilisation or Tuina (Traditional Chinese medicine massage), traction, spinal mobilisation and (high-velocity, low-amplitude thrust) manipulation), [13, 14, 16, 17, 23, 35, 43, 45–49] (ii) movement therapies (exercises, positioning, handling and environmental adaptations that aim to strengthen weak neck and trunk muscles), [13, 14, 16, 17, 20, 47, 48] (iii) acupuncture, [50–53] (iv) adjunctive therapies such as electrotherapy (including microcurrent, ultrasound and heat therapy) and taping, [18, 54, 55] and (v) physical support (including soft collar, tubular orthosis and pillow); [46, 56]
- (3) Individual outcomes including range of neck motion, symmetric head posture, adverse events, lateral head-righting, thickness of SCM tumour, early motor skills, development of symmetrical movement, craniofacial asymmetry, parental stress and compliance to exercises. Examples of validated outcome measures are Still Photography for symmetric head posture (scale 0–25 degrees) and the Muscle Function Scale for lateral head-righting (scale 0 – 5). [57, 58] Studies were included regardless of the outcomes they selected. However, where studies reported data that was amalgamated from more than one outcome, the results for these studies were included, but the amalgamated data from analyses were excluded.

Screening and data extraction

Covidence software was used to remove duplicate records (JA) [40]; then two reviewers, including at least one who was fluent in the language of the original paper, independently screened titles and abstracts against the inclusion and exclusion criteria and resolved disagreements through discussion (two of EC, JA, JK, NJC, NTNC, TM, YC) (see Appendix 2A in Additional file 2). Working in pairs, reviewers obtained full-text reports and assessed them independently against the review criteria, resolving disagreements through a third reviewer (DU or MJ) when necessary. Two reviewers (two of DWLW, EC, HA, HJH, HL, LR, JA, JK, JP, JS, JX, ML, NTNC, NJC, PE, PV, RB, TM, YC) independently extracted data onto a pre-prepared standardised form in Covidence (Appendix 2B in Additional file 2). [40] Where additional information was required, attempts were made to contact study authors.

It was recorded if study data was not reported, or no reply was received to requests for further information. Disagreements between data extractors were resolved by discussion, or by arbitration from a third reviewer (DU or MJ) as needed. One review author (JA) exported the extracted data from Covidence into Review Manager 5. [59]

Risk of bias assessment

Two reviewers (two of DWLW, EC, HA, HL, JA, LR, ML, NTNC, PV, RB, TM, YC) independently used the Cochrane Risk of bias (RoB) 1 tool with domains for selection, performance, detection, attrition, reporting and other sources of bias, [60] and resolved any differences in interpretation through consensus. Where required, a third reviewer provided arbitration (DU or MJ). The overall RoB for a study was recorded to be the same as the least favourable assessment made for any individual domain of bias in the study, using the methodology from the RoB 2 tool [61].

Data synthesis and statistical analyses

Review Manager 5.4 software was used for data synthesis [59]. Interventions were categorised into combinations because this is the way that they were investigated in RCTs; that is, interventions typically comprised between two and five component treatments. Where it was possible to calculate an effect size, mean differences (MDs) or risk ratios (RRs) with 95% Wald confidence intervals (CIs) were reported for summary effects. Where different scales were used to measure similar outcomes, standardised MDs (SMDs) were estimated. In order to report the improvement in range of neck motion, symmetric head posture and lateral head-righting, stratification of the analysis by timing of measurement was performed and all estimates of improvement were converted into odds ratios (ORs). To convert continuous measurements into ORs, one author (MJ) first calculated SMDs and then used the method of Hasselblad and Hedges. [62] Summary SMD from one meta-analysis was interpreted using Cohen's effect sizes (small, medium, large). [63] The aim was to present results from a network meta-analysis as summary relative effect sizes (RR or MD) for each possible pair of treatments; however, there was an insufficient number of studies in each intervention pair to permit reliable results. Where multiple outcomes were used to measure a construct, all outcomes were recorded to avoid losing potentially useful data; and for outcomes measured at multiple follow-up time-points, data were extracted from every available time-point. Where data permitted, random-effects meta-analyses (inverse variance method) were conducted because of potential clinical and methodological diversity in the studies included

in the meta-analysis, but the fixed-effect model was also used in a sensitivity analysis. Otherwise, a narrative summary of the results was provided. Due to insufficient numbers of poolable studies and/or inadequate demographic and descriptive detail reported in the included studies, a subgroup analysis by infant age, type or severity of CMT was unable to be performed as planned.

Certainty of evidence

Two reviewers (JA and NTNC) independently used GRADEpro GDT software to rate the certainty of the evidence as high, moderate, low, or very low, based on the five criteria outlined in the grading of recommendations assessment, development and evaluation (GRADE) framework (risk of bias, inconsistency, indirectness, imprecision and publication bias). [64, 65] Where consensus was not reached, a third reviewer arbitrated (DU or MJ). The evidence was summarised for each outcome by comparison (see Additional file 3).

Results

The search strategy yielded 5364 records (Fig. 1). After removing 1833 duplicates and following title, abstract and full text screening, 87 RCTs and 13 quasi-RCTs were identified as suitable for inclusion; these studies involved 8125 participants with CMT. The number of studies excluded and the reasons for exclusion are presented in Fig. 1.

Primary study characteristics are shown in Additional file 4 and provided in detail in Additional file 5. The predominant study locations were the People's Republic of China (eighty studies (80%)) followed by Europe (11 studies (11%)). The number of participants per study ranged from 20 [36, 37] to 310 [66] and 25 studies (25%) had 100 participants or more. Of the 100 studies, 40 (40%) identified a single category or type of CMT and either excluded all other types or made no reference to them, 38 (38%) investigated two to three categories, and 22 studies (22%) did not identify the type of CMT being investigated.

The interventions were divided into 10 categories (see Additional file 6). The control arm for each trial is described in Supplementary Table 5A, see Additional file 5. On examining the interventions and controls, 70 studies (70%) were found to use an additive interventional design, where an active control was compared with the experimental intervention added to an active control, 28 studies (28%) were substitutive, in which an experimental intervention was compared to an active control, and two studies (2%) used an experimental intervention compared with a placebo or no treatment. [67, 68] Trials were grouped according to their intervention, control and design, observing considerable heterogeneity, and consequently 24 distinct groups or comparisons were

formulated for analysis. The effects of each intervention broken down by comparison are detailed in Additional file 7.

Ten outcomes were reported, each of which were measured by one or more instruments (see Additional file 4). Range of neck motion was evaluated in 38 studies (38%), and symmetric head posture in 16 studies (16%), each commonly measured by an arthrodial protractor; lateral head-righting was assessed in 9 studies (9%), typically using the Muscle Function Scale; thickness of SCM tumour (32 studies (32%)) was usually measured using colour ultrasonography; early motor skills was assessed in 5 studies (5%), with the Alberta Infantile Motor Scale being used by some studies, and the development of symmetrical movement was measured in 7 studies (7%), with some trials using the Idiopathic Infantile Asymmetry Scale. Changes to craniofacial asymmetry were reported in 11 studies (11%), often assessed using items taken from the Severity Assessment for Plagiocephaly; parental stress was scored using the Parental Stress Index in 1 study (1%), and compliance to home exercises was evaluated by a parental questionnaire in 1 study (1%). The presence or absence of adverse events was reported in 29 studies (29%) and are described in Table 1.

Quality assessment

The overall RoB was judged to be high for 51 studies and unclear for the remaining 49 studies. In general, study reports lacked the information required to determine either 'high' or 'low' RoB, and hence were rated 'unclear'. Regarding individual domains for RoB, the number of studies determined to be 'unclear' ranged from 41 studies that did not provide sufficient information about random sequence generation to 83 studies without sufficient detail to determine selective reporting. The domains most commonly determined to have a 'low' RoB were random sequence generation (48 studies) and other sources of bias (47 studies) (Fig. 2 and Additional file 5).

Meta-analysis

Five of the ten outcomes are reported in meta-analyses: range of neck motion, symmetric head posture, lateral head-righting, thickness of SCM tumour and the development of symmetrical movement. Table 2 gives a summary of evidence from meta-analyses across professions presented by comparison and outcome, along with directions of effect, overall risks of bias and certainty of evidence. Of the 24 comparisons, 74 (74%) studies were assigned to five comparisons, and of these five, three incorporated meta-analyses formed from

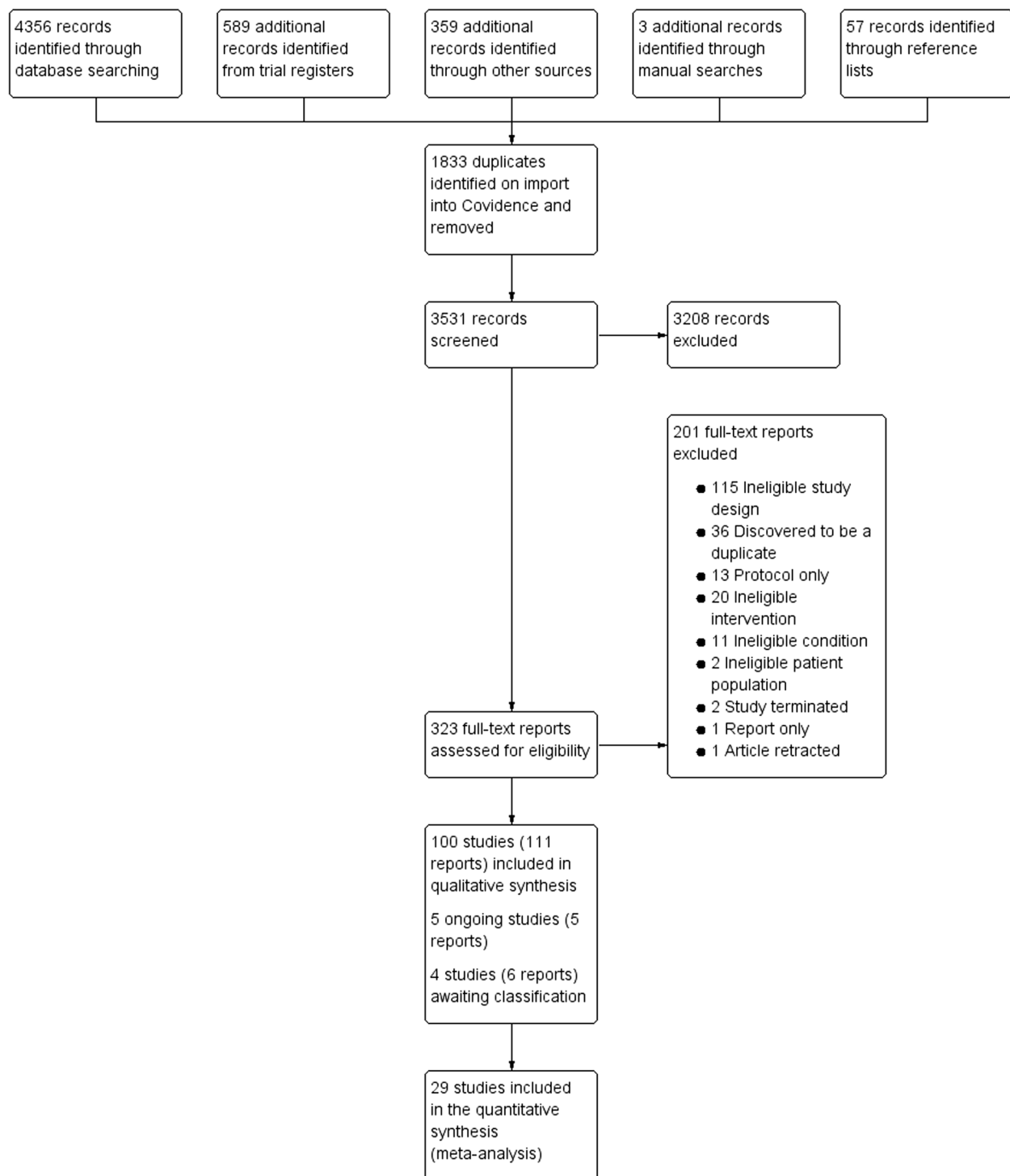


Fig. 1 Preferred Reporting Items for Systematic Reviews and Meta-analyses flowchart of included and excluded studies

24 (24%) studies. There were no meta-analyses in the remaining 21 comparisons. The results of investigations of heterogeneity and sensitivity analyses are provided in Additional file 7.

Adding manual therapy to a control intervention

A total of 33 studies (33%) examined adding manual therapy to a control intervention, in which the control may contain a manual therapy component. Twenty

Table 1 Adverse events by comparison

Studies that examined adverse events (No. (%))	No. of studies that reported adverse event / no adverse event	Incidence of adverse event (No. (%))	Type of adverse event in each study arm	No. of participants in studies reporting presence or absence of adverse events	Certainty of evidence
Comparison 1*. Adding manual therapy to a control intervention versus control alone					
14 (42) [8, 16, 34–36, 69–77]	2 / 12	Philippi 2006 [8] IG: 4 (25) CG: 6 (37.5)	IG: osteopathic treatment, positioning and exercises: excessive crying CG: sham treatment, positioning and exercises: excessive crying	880 ^a	Very low
		Yue 2018 [75] IG: NNR (NR) CG 0 (0)	IG: improved Tuina with stretching: some parents were distressed by the stretching and some participants withdrew from the trial CG: NA		
Comparison 2*. Adding exercises to a control intervention versus control alone					
2 (25) [78, 79]	0 / 2	0 (0)	NA	189	Very low
Comparison 3*. Adding electrophysical agents to a control intervention versus control alone					
4 (29) [18, 80–82]	2 / 2	Li 2013 [80] IG: 1 (1.6) CG: 0 (0)	IG: wax therapy, Tuina with stretching: coryzal symptoms CG: NA	300	Very low
		Wang 2016b [82] IG: NNR (NR) CG: NNR (NR)	IG: wax therapy, biofeedback, Tuina with stretching: excess crying CG: Tuina with stretching: excess crying		
Comparison 4*. Manual therapy versus control manual therapy					
1 (9) [83]	1 / 0	Cui 2019 [83] IG: 0 (0) CG: 2 (6)	IG: Tuina without stretching CG: textbook Tuina with stretching: dropped out due to distress	68	Low
Comparison 6*. Manual therapy versus control manual therapy with exercises					
1 (50) [84]	0 / 1	0 (0)	NA	48	Very low
Comparison 9*. Exercises versus control exercises with manual therapy					
1 (50) [7]	0 / 1	0 (0)	NA	37	Very low
Comparison 10*. Manual therapy with exercises versus control manual therapy with exercises					
1 (100) [19]	1 / 0	Öhman 2010 [19] IG: 1 (10) CG 0 (0)	IG: passive stretching provided by a manual therapist and caregiver exercises: partial rupture of the SCM muscle CG: NA	20	Very low
Comparison 11*. Manual therapy with exercises versus control manual therapy with control exercises					
1 (50) [85]	0 / 1	0 (0)	NA	204	Very low
Comparison 15*. Electrophysical agents with manual therapy and exercises versus control manual therapy					
2 (67) [86, 87]	1 / 1	Chen 2020b [86] IG: 1 (4) CG: 7 (28)	IG: passive stretching, strengthening exercises, ultrasound, magnetic therapy, home positioning and handling: events such as oedema and intramuscular bleeding CG: Tuina with stretching: events such as oedema and intramuscular bleeding	155	Low

Table 1 (continued)

Studies that examined adverse events (No. (%))	No. of studies that reported adverse event / no adverse event	Incidence of adverse event (No. (%))	Type of adverse event in each study arm	No. of participants in studies reporting presence or absence of adverse events	Certainty of evidence
Comparison 16*. Electrophysical agents with manual therapy versus control electrophysical agents, control manual therapy and exercises					
1 (100) [55]	1 / 0	Lin 2023[87] IG: 0 (0) CG: NNR	IG: modified Tuina, exercises, Biofeedback and ultrasound Or 'NA' CG: Tuina with stretching	15	Very low
Comparison 17*. Acupuncture or electroacupuncture with manual therapy versus manual therapy alone					
1 (14) [88]	0 / 1	Kim 2009[55] IG: 3 (43) CG: 8 (100)	IG: microcurrent and stretching; crying CG: physiotherapy with stretching; crying	64	Very low

* Comparisons 5, 7–8, 12–14 and 18–24 reported neither the presence nor absence of adverse events

AE adverse events, CG control group, IG intervention group, NA not applicable, NNR number not reported, NR not reported, SCM sternocleidomastoid

^a Number of participants greater than 880 because an unreported number with side effects were excluded from the analysis

of these studies contributed data to 15 meta-analyses. While there was low certainty evidence that adding manual therapy to a control intervention may result in a greater improvement in the range of passive cervical rotation at 4 to 7 weeks (OR 9.79, 95% CI 4.26 to 22.50; 4 studies, 231 participants; $I^2=57\%$), there was also low certainty evidence of no additional benefit after 8 weeks; removing a quasi-RCT indicated no overall difference in the results (Fig. 3, A; Fig. 3, H in Additional file 8). Results for range of passive cervical lateroflexion and active cervical rotation showed low certainty evidence of similar benefits at 4 to 7 weeks for adding manual therapy; however, removing a quasi-RCT showed low certainty evidence of benefits to passive cervical lateroflexion at 8 to 13 weeks for adding manual therapy (OR 3.13, 95% CI 1.56 to 6.31; 4 studies, 139 participants; $I^2=0\%$). The evidence was downgraded for passive cervical rotation, passive cervical lateroflexion and active cervical rotation to low certainty because of a combination of high risk of bias and study heterogeneity. (Fig. 3, B; Fig. 3, C; Table 2, Effects of interventions in Additional file 7; Fig. 3, I in Additional file 8).

There was very low certainty of evidence that additional manual therapy may achieve greater gains in symmetric head posture at 4 to 7 weeks (OR 4.55, 95% CI 2.57 to 8.05; 4 studies, 231 participants; $I^2=22\%$), but the analysis found very low certainty of evidence of no further benefit after 8 weeks (Fig. 3, D). This review found no clear difference to lateral head-righting with additional manual therapy at any time point but this was based on very low certainty evidence with a high overall RoB (Fig. 3, E); the evidence was downgraded for both symmetric head posture and lateral head-righting due to a combination of high risk of bias, small sample

sizes and study heterogeneity. There was low certainty evidence that adding manual therapy may reduce the thickness of SCM tumour at one month (MD -1.76 mm, 95% CI -2.61 to -0.90 ; 3 studies, 166 participants; $I^2=45\%$) and at three months (MD -2.69 mm, 95% CI -4.45 to -0.93 ; 4 studies, 251 participants; $I^2=88\%$). The evidence was downgraded to low certainty due to a combination of high risk of bias and study heterogeneity, and removing a quasi-RCT indicated no important difference in the results. (Fig. 3, F; Table 2, Effects of interventions in Additional file 7; Fig. 3, J in Additional file 8). The review found low certainty evidence of greater development of symmetrical movement after 4 to 6 weeks, which was the only available time point (SMD -0.70 , 95% CI -0.95 to -0.45 ; 3 studies, 265 participants; $I^2=0\%$) (Fig. 3, G). This is a medium to large effect size according to Cohen; the evidence was downgraded to low certainty due to a combination of high risk of bias and small sample sizes.

Adding electrophysical agents to a control intervention

Fourteen studies examined adding a range of electrophysical agents to a control intervention, with low certainty evidence showing that electrophysical agents may result in a reduction in SCM tumour thickness after 8 weeks (MD -2.03 mm, 95% CI -2.67 to -1.39 ; 2 studies, 130 participants; $I^2=73\%$) (Fig. 4). The evidence for adding electrophysical agents was downgraded to low certainty because of a high risk of bias and imprecision.

Substituting manual therapy with a control manual therapy

Eleven studies examined the efficacy of manual therapy compared with a different manual therapy on SCM

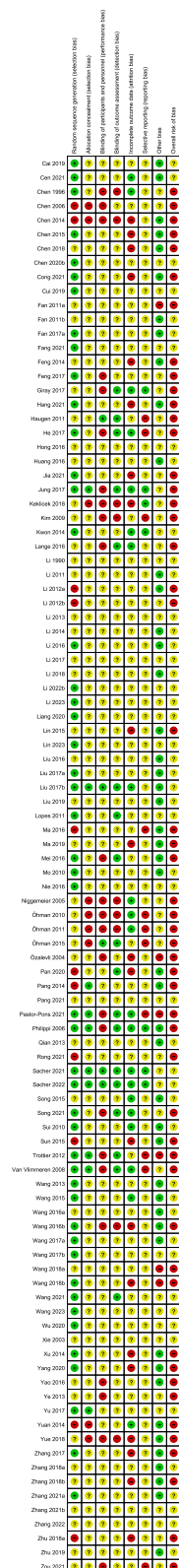


Fig. 2 Risk of bias summary: review authors' judgements of risk of bias for each included study

tumour thickness. Of these, two studies examined an optimised Tuina protocol versus a traditional Tuina protocol and contributed data to a meta-analysis. This review found very low certainty evidence that optimised Tuina protocol achieved a greater reduction in the thickness of the SCM tumour at 3 to 4 months (MD -1.20 mm, 95% CI -1.80 to -0.59 ; 2 studies, 132 participants; $I^2=52\%$) (Fig. 5). The evidence for substituting manual therapy was downgraded to very low certainty because of high risk of bias in three domains and imprecision.

Adverse events

Of the 100 studies, 29 (29%) reported on the presence or absence of adverse events (see Table 1), while 71 (71%) did not provide any data on adverse events. Of the 29 studies reporting on adverse events, 21 studies (72%) reported no adverse event, and eight studies (28%) reported a range of events. This review found low to very low certainty evidence for adverse events for conservative treatments for CMT across six of the 24 comparisons; in a further five comparisons, no adverse events occurred in intervention or control arms, and of the remaining 13 comparisons, no study reported on the presence or absence of adverse events. Of the adverse events, excessive crying or parental distress was reported in five studies which examined manual therapy alone, or manual therapy with exercises, or with an electrophysical agent, or with both exercises and an electrophysical agent. More serious events such as oedema, intramuscular bleeding and a snapped SCM muscle were reported in two studies investigating manual therapy with exercises, manual therapy with an electrophysical agent and exercises, or manual therapy alone.

Discussion

This is the first systematic review to examine the effectiveness and safety of non-surgical, non-pharmacological treatments for CMT across the disciplines of Physiotherapy, Traditional Chinese Medicine, Osteopathy and Chiropractic, and without restrictions on language or publication status. Interventions considered included manual therapy, exercises, electrophysical agents, acupuncture, electroacupuncture, taping or an orthopaedic support. Low to very low certainty evidence was found that adding manual therapy to an active control may result in greater improvement in passive cervical rotation and lateroflexion, active cervical rotation, symmetric head posture and developing symmetrical movement over the short-term of 4 to 7 weeks and reduce the thickness of SCM tumour at 1 and 3 months. This review found low certainty evidence that adding electrophysical agents to an active control may achieve greater reduction in the thickness of the SCM tumour after 8 weeks, and

Table 2 Summary of evidence from meta-analyses, examining manual therapy or electrotherapy compared to a control intervention

Comparison 1*: Adding manual therapy to a control intervention versus control alone						
Outcome	No. of contributing studies (No. of studies contributing to meta-analyses)	Assessment time point	No. of participants in meta-analyses	Effect size and 95% CI (OR unless otherwise indicated)	Direction of effect	Overall RoB
Range of neck motion: passive cervical rotation ^b	8 (7)	4 to 7 weeks 8 to 13 weeks 18 to 26 weeks	231 259 149	9.79 [4.26, 22.50] 1.91 [0.57, 6.39] 4.23 [0.30, 60.14]	Favours intervention in shorter term	High
Range of neck motion: passive cervical lateroflexion ^c	8 (5)	4 to 7 weeks 8 to 13 weeks 18 to 26 weeks	79 259 149	2.66 [1.17, 6.04] 1.96 [0.88, 4.38] 7.97 [0.24, 266.09]	Favours intervention in shorter term ^c	Low
Range of neck motion: active cervical rotation	5 (4)	4 to 7 weeks 8 to 13 weeks	32 90	3.94 [1.08, 14.35] 2.24 [0.52, 9.70]	Favours intervention in shorter term	High
Symmetric head posture	7 (6)	4 to 7 weeks 8 to 13 weeks 18 weeks	231 139 29	4.55 [2.57, 8.05] 2.28 [0.87, 5.98] 1.79 [0.47, 6.77]	Favours intervention in shorter term	High
Lateral head-righting	5 (4)	4 to 7 weeks 8 to 13 weeks 18 weeks	79 139 29	1.88 [0.83, 4.24] 1.37 [0.69, 2.71] 0.78 [0.21, 2.90]	No clear evidence of a difference	High
Thickness of SCM tumour ^d	10 (9)	After 1 month After 3 months Unspecified	166 252 168	MD -1.76 [-2.61, -0.90] MD -2.69 [-4.45, -0.93] MD -1.53 [-2.09, -0.97]	Favours intervention at all timepoints	High
Development of symmetrical movement	3 (3)	After 4 to 6 weeks	265	SMD -0.70 [-0.95, -0.45]	Favours intervention	High
Comparison 3*: Adding electrophysical agents to a control intervention versus control alone						
Outcome	No. of contributing studies (No. of studies contributing to meta-analyses)	Assessment time point	No. of participants in meta-analyses	Effect size and 95% CI (OR unless otherwise indicated)	Direction of effect	Overall RoB
Thickness of SCM tumour	3 (2)	After 8 weeks	130	MD -2.03 [-2.67, -1.39]	Favours intervention	High
Comparison 4*: Manual therapy versus control manual therapy						
Outcome	No. of contributing studies (No. of studies contributing to meta-analyses)	Assessment time point	No. of participants in meta-analyses	Effect size and 95% CI (OR unless otherwise indicated)	Direction of effect	Overall RoB
Thickness of SCM tumour	6 (2)	After 3 to 4 months	132	MD -1.20 [-1.80, -0.59]	Favours intervention	High

* Comparison 2 did not attract a meta-analysis

CI confidence interval, MD mean difference, No number, OR odds ratio, RoB Risk of bias, SCM sternocleidomastoid, SMD standardised mean difference

^a Low to very low certainty evidence was due to a combination of high risk of bias, small sample sizes and study heterogeneity. See Additional File 7 (Effects of interventions) for an explanation of each assessment

^b After removing the quasi-RCT, the analysis showed no important difference in the result at 8 to 13 weeks (OR 3.00, 95% CI 0.87 to 10.33; $n = 139$) or at 18 to 26 weeks (OR 1.12, 95% CI 0.30 to 4.15; $n = 29$)

^c After removing the quasi-RCT, the analysis favoured the intervention at 8 to 13 weeks (OR 3.13, 95% CI 1.56 to 6.31; $n = 139$), but showed no important difference in the result at 18 to 26 weeks (OR 1.28, 95% CI 0.35 to 4.68; $n = 29$)

^d After removing the quasi-RCT, the analysis showed no important difference in the result at the unspecified time period (MD -1.53, 95% CI -2.31 to -0.75; $n = 68$)

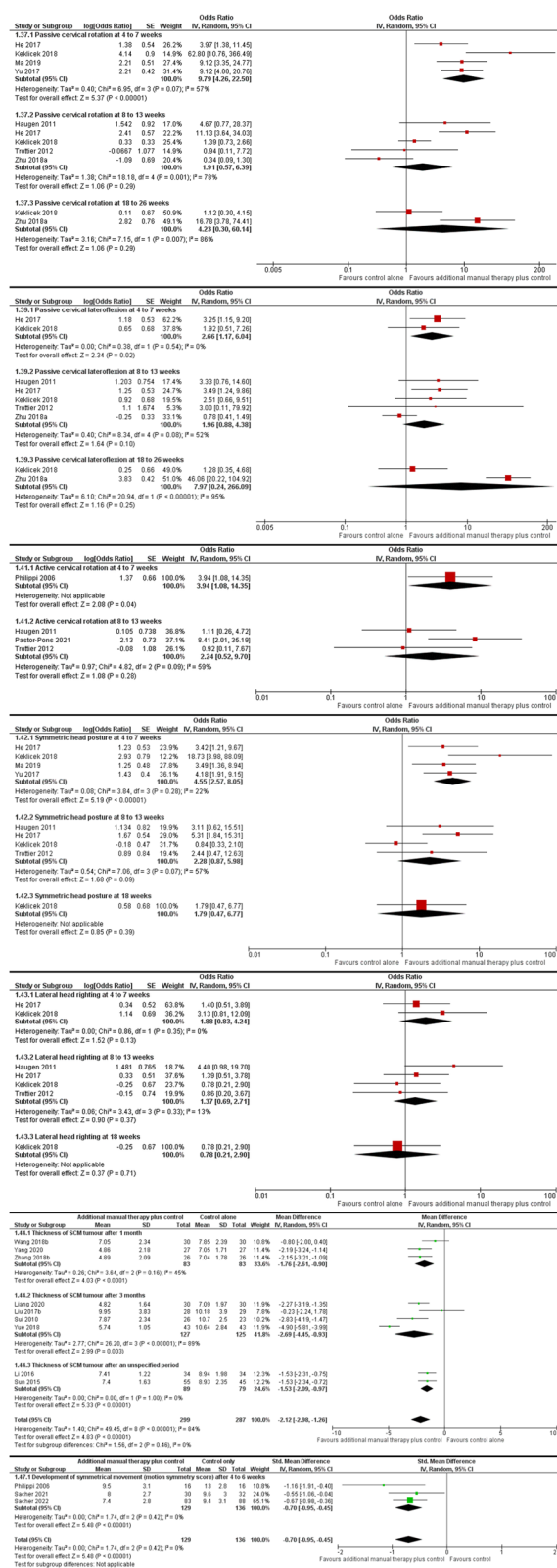


Fig. 3 Meta-analyses of studies that add manual therapy to a control intervention. **A** passive cervical rotation. **B** passive cervical lateroflexion. **C** active cervical rotation. **D** symmetric head posture. **E** lateral head-righting. **F** thickness of SCM tumour. **G** development of symmetrical movement

very low certainty evidence that optimised Tuina may achieve greater reduction in SCM tumour thickness than traditional Tuina at 3 to 4 months. High levels of heterogeneity were evident across all meta-analyses, which may be explained by differences in type and severity of CMT in component studies, differences in the content and delivery of interventions and controls, and differences in how outcomes were measured. Adverse events were not common, however they were poorly reported and the certainty of evidence was low to very low.

Based on 15 meta-analyses and nine single RCTs, this review found adding manual therapy (passive stretching, soft tissue mobilisation (including Tuina), osteopathy (including cranial osteopathy and myofascial release), mobilisation and infant-adapted, high-velocity, low-amplitude manipulation) to an active control may improve cervical range of movement and symmetry (except lateral head-righting) [8, 16, 34, 69–74, 89], tumour thickness [75, 76, 90–97] and the development of symmetrical movement [8, 35, 36] over a short-term period. However, due to small sample sizes and high RoB in many studies, the evidence was of low to very low certainty. Current clinical guidelines recommend passive and active stretching as first choice interventions for CMT, rating the quality of evidence as moderate [17]. The guidelines suggest that soft tissue mobilisation and massage may augment first choice interventions, but found no evidence for or against craniosacral therapy (cranial osteopathy), and recommend against cervical manipulation due to perceived risks. A number of factors may explain the differences in findings and conclusions between this systematic review and the guidelines.

The current review provides an update of the evidence that includes 100 RCTs based on a search up until August 2023, including 33 RCTs that examined adding manual therapy to an active control. This compares to the clinical guidelines, which are informed by observational studies and nine RCTs, of which four RCTs examined manual therapy. [16, 19, 69, 71] This review examined RCTs from a range of professions and in any language, while the guidelines included studies from a single discipline identified through English-language databases; moreover, the findings are based on adding manual therapy to an active control, versus considering manual therapy alone. Also,

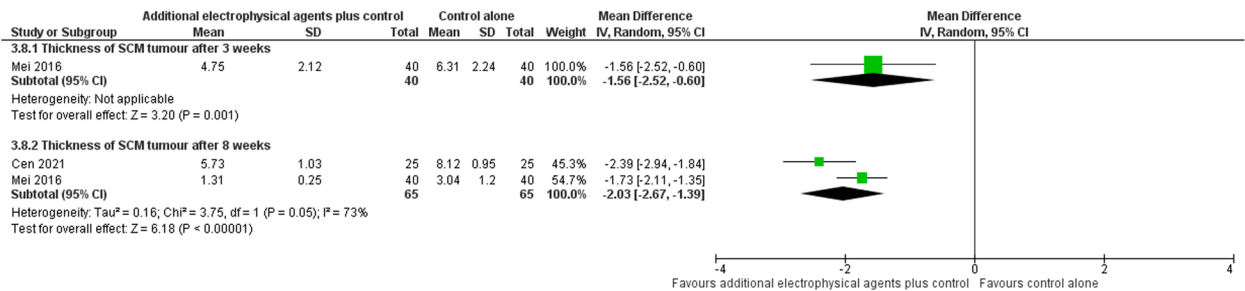


Fig. 4 Meta-analyses of studies that add electrophysical agents to a control intervention. Thickness of SCM tumour at 8 weeks

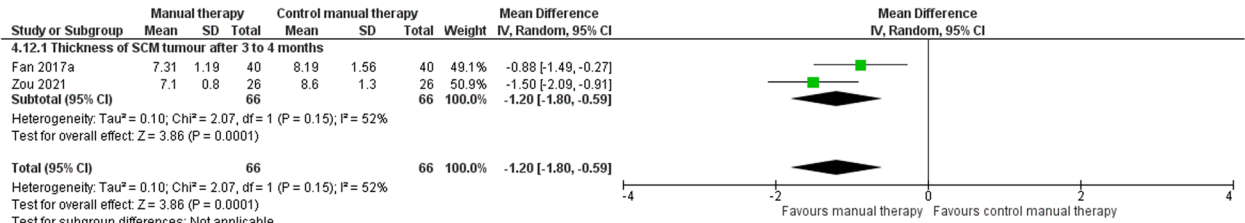


Fig. 5 Meta-analyses of studies that compare manual therapy with a control manual therapy. Thickness of SCM tumour after 3 to 4 months

there are differences between this review and the guidelines in their respective RoB and study quality judgements [29, 59, 61]. As a result of these dissimilarities, this review draws different conclusions, underlines the need to improve the quality of clinical trials and trial reporting, and highlights the impact of including a broad range of evidence-based manual therapies without language restriction when examining the evidence.

This study found that adding electrophysical agents to an active control may improve passive cervical range of movement [18, 80, 98] and symmetric head posture [98] and reduce the thickness of the SCM tumour; [81, 99, 100] however, this was low-certainty evidence based on one meta-analysis and six additional single RCTs with small sample sizes and high RoB. National guidelines suggest electrophysical agents may be used as a supplementary intervention to improve range or alignment, but do not refer to their use for reducing SCM tumour thickness, so the low certainty evidence for this outcome based upon a two-study meta-analysis and two single RCTs is a novel finding. [17, 81, 99] This review also investigated a range of Tuina protocols with or without passive stretching for a range of outcomes. Two RCTs provided low certainty evidence that some protocols of Tuina with stretching may achieve better range or alignment outcomes than other protocols, [37, 101] but a two-study meta-analysis and four single RCTs showed an inconsistent effect for Tuina with or without stretching on reducing the size of the SCM tumour; for this the evidence of effect is very uncertain. [37, 83, 102–105] Two systematic

reviews reported a small meta-analysis comparing Tuina versus stretching plus exercises[27, 32], but this review did not include this analysis because one of the studies amalgamated data across outcomes and provided no unamalgamated data, [106] whilst the other study included Tuina in both study arms, which is inconsistent with this meta-analysis’s criteria [107].

This review found low to very low certainty of evidence for an adverse event, but noted poor reporting of adverse events overall. Amongst 100 RCTs involving 8125 participants, 29 adverse events were reported in 29 trials involving 1905 participants (with not all trials reporting adverse events in both treatment and control groups), and 71 trials did not state whether events occurred. Manual stretching was the most common form of intervention for CMT, however it has been suggested to snap the SCM muscle in about 9% of infants with an SCM tumour, causing bruising and atrophy in about 25% of those cases. [17, 108] In comparison, this review only noted one instance of snapping and 11 cases of oedema and bruising across 64 RCTs that used manual therapy for infants with an SCM tumour, and it is possible that the incidence of snapping, bleeding and oedema is underreported. Moreover, the use of high-velocity, low-amplitude cervical manipulation as a treatment for CMT is unclear, with the clinical guidelines recommending against their use due to insufficient evidence of benefits, [17] a report of a death, with an association to a “thrust” technique questionable, [17, 23, 109, 110], Jung[personal communication]

and accounts of autonomic reactions following low-force manipulative treatment. [23, 111] This review, based on 3 RCTs, found very low certainty evidence for adverse events with high-velocity low amplitude manipulation. [16, 35–6] The studies incorporated infant-adapted techniques, of which one study reported zero adverse events, [16] and the remaining two studies noted autonomic reactions, but found no evidence that the reactions were associated with manipulation. [35, 36] Consequently, this review is unable to recommend against infant-adapted upper cervical manipulation in a carefully selected population with postural torticollis. More rigorous reporting of adverse events is needed, without which clinicians and patients cannot make informed decisions concerning the risks of any proposed treatment.

Although it is possible that nonsurgical and nonpharmacological treatment may be effective for CMT, poor trial reporting, small sample sizes, inadequate recording of adverse events and a high RoB in many trials reduced the certainty of evidence and did not allow any firm conclusions to be made regarding the benefits or safety of interventions. Further, minimal clinically important differences in CMT symptoms have not been established; this is an important gap in the literature, because without these we cannot understand the importance of the change for patients or clinicians. Whilst the evidence is insufficient to confidently guide the use of these therapies, these findings may assist clinicians in educating and advising patients so they can make informed decisions with respect to treatment for CMT. Future trials are urgently needed that build on existing research which in turn create larger, robust meta-analyses and provide higher certainty evidence. To minimise the issue of studies being rated as ‘unclear’ risk of bias, and to facilitate pooling studies for meta-analysis and subgroup analyses, there is an important need for trial authors to follow the reporting recommendations set out in the CONSORT statement. [112] In addition to providing information about random sequence generation and sufficient reporting on outcomes, trials need to clearly state the CMT type and intervention, and report unamalgamated outcome data measured with validated tools. Moreover, minimal clinically important differences need to be established and adverse events more rigorously reported, because without these, neither clinicians nor patients can make meaningful or informed decisions concerning the value of any proposed treatment.

This review has a number of strengths. The review protocol was published in the Cochrane database, 31 electronic databases and trial registers were searched across a range of disciplines without language restriction, and

Cochrane methodology was used to determine RoB and grade the certainty of evidence. The review team located 100 RCTs, formed 18 original meta-analyses and collected comprehensive information regarding benefits and risks. This review also has some important limitations. Trials rarely conformed to the reporting recommendations set out in CONSORT guidelines, [112] and there were considerable difficulties obtaining further information from study authors. Ninety-six studies (96%) were limited to infants aged 0 to 12 months or did not report the participants’ age, possibly limiting the relevance of the review to babies aged under 12 months. Further, poor consistency across controls and interventions, substantial differences in the ways that trials measured outcomes and negligible replication of study designs limited the pooling of studies for meta-analysis and prevented construction of a network meta-analysis. Quasi-RCTs were included in this review due to the significant limitations with reporting of adverse events in trials of CMT. While quasi-RCTs provided important data and were found not to introduce bias, reporting of adverse events was still restricted and limited the conclusions that could be drawn about the safety of interventions in this review.

Conclusions

This systematic review with meta-analysis found that adding manual therapy or electrophysical agents to an active control and the treatment of Tuina may be effective for CMT, but owing to study heterogeneity and poor trial reporting, the certainty of the evidence was low to very low. Adverse events were not common but poorly reported, and the evidence was of low to very low certainty. These findings are important in informing clinical guidelines and treatment for CMT and highlight the urgent need for better quality reporting of trials and outcomes including adverse events, replicating existing study designs to enable robust meta-analyses, and implementing large, definitive randomised trials to address the limitations of current studies.

Abbreviations

CI	Confidence interval
CMT	Congenital muscular torticollis
CONSORT	Consolidated Standards of Reporting Trials
GRADE	Grading of recommendations assessment, development and evaluation
MD	Mean difference
OR	Odds ratio
PRISMA	Preferred Reporting Items for Systematic Reviews and Meta-analyses
RoB	Risk of bias
RCT	Randomised controlled trial
RR	Risk ratio
SCM	Sternocleidomastoid
SMD	Standardised mean difference

Supplementary Information

The online version contains supplementary material available at <https://doi.org/10.1186/s12891-025-08407-3>.

Additional file 1: Appendix 1 Search strategy

Additional file 2: Appendix 2 Appendix 2A. Inclusion and exclusion criteria. Appendix 2B. Data extraction form

Additional file 3: Supplementary Table 3. Evidence for each outcome by comparison

Additional file 4: Supplementary Table 4. Primary study characteristics

Additional file 5: Supplementary Table 5. Characteristics of studies. Supplementary Table 5A: Characteristics of included studies. Supplementary Table 5B: Characteristics of excluded studies. Supplementary Table 5C: Characteristics of studies awaiting classification. Supplementary Table 5D: Characteristics of ongoing studies

Additional file 6: Supplementary Table 6. Interventions

Additional file 7: Effects of interventions

Additional file 8: Fig. 3. Meta-analyses of studies excluding quasi-RCTs that add manual therapy to a control intervention. H. passive cervical rotation (excluding quasi-RCT). I. passive cervical lateroflexion (excluding quasi-RCT). J. thickness of SCM tumour (excluding quasi-RCT).

Additional file 9: Appendix 3: Differences between protocol and review. Supplementary Table 7A: Unused methods archived for future updates of this review. Supplementary Table 7B: Expansion on methods given in protocol

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Authors' contributions

JA conceptualised and designed the study, collected and investigated data, assessed the quality of evidence and outcome validation, synthesised the evidence, drafted the initial manuscript and critically reviewed and revised the manuscript. MJ conceptualised and designed the study, conducted statistical analysis, supervised the project, drafted the initial manuscript and critically reviewed and revised the manuscript. NC collected and investigated data, assessed the quality of evidence and outcome validation, drafted the initial manuscript and critically reviewed and revised the manuscript. YC, ML and EC collected and investigated data, assessed the quality of evidence, and drafted the initial manuscript. HL collected and investigated data, assessed the quality of evidence, drafted the initial manuscript and critically reviewed and revised the manuscript. TC assisted with conceptualising the study, assessing outcome validation, drafted the initial manuscript and critically reviewed and revised the manuscript. CL contributed to study methodology and analysis, drafted the initial manuscript and critically reviewed and revised the manuscript. DU conceptualised and designed the study, supervised the project, drafted the initial manuscript and critically reviewed and revised the manuscript. All authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

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Data availability

All data analysed during this study are included in this published article and its supplementary information files.

Declarations

Ethics approval and consent to participate

Not applicable.

Consent for publication

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Competing interests

The authors declare no competing interests.

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