Informing the Physical Therapy Management of Congenital Muscular Torticollis Clinical Practice Guideline: A Systematic Review

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Purpose: To systematically review current evidence on the physical therapy assessment, intervention, and prognosis of congenital muscular torticollis (CMT) to inform the update to the 2018 CMT Clinical Practice Guideline (CPG).
Methods: Six databases were searched for studies that informed assessment, intervention, and prognosis for physical therapy management of infants with CMT.
Results: Fifteen studies were included. Four studies investigated the psychometric properties of new and established assessments. Six studies informed the feasibility and efficacy of first-choice and supplemental interventions including traditional Chinese medicine and neural and visceral manipulation. One qualitative study found that parents of infants with mild and severe CMT had different concerns. Five studies informed prognosis, including factors associated with treatment duration, clinical outcomes, and use of supplemental interventions.
Conclusion: Newer evidence reaffirms 5 of 17 recommendations of the 2018 CMT CPG and could increase the recommendation strength to strong for neck passive range of motion. (Pediatr Phys Ther 2023;00:1–11)
Key words: assessment tools, congenital muscular torticollis, infant, intervention, physical therapy, prognosis, psychometric properties, systematic review

INTRODUCTION

Congenital muscular torticollis (CMT) is a postural, musculoskeletal deformity evident at or shortly after birth. CMT is characterized by the shortening or stiffness of the sternocleidomastoid muscle (SCM) resulting in ipsilateral lateral flexion of the head with contralateral rotation. The incidence of CMT ranges from 3.9%[1,2] to 16%[3] of newborns. Evidence supports that early referral and initiation of physical therapy (PT) leads to improved outcomes,[4,5] shorter durations of care,[4-6] and reduced need for surgical intervention.[4,7-9] If an infant diagnosed with CMT begins PT before 1 month of age, the prognosis for full cervical range of motion (ROM) is 98% with 1.5 ± 0.3 months of PT.[5] Beginning between 1 and 3 months of age, the prognosis for full cervical ROM declines to 89% with 5.9 ± 0.6 months of PT.[5] Beginning between 3 and 6 months of age, the prognosis for full cervical ROM declines to 63% with 7.2 ± 0.6 months of PT, and beginning between 6 and 12 months of age, the prognosis for full cervical ROM declines to 19% with 9.8 ± 0.6 months of PT.[5] Yet, a survey of pediatric physical therapists in the United States found that infants with CMT are most commonly referred to PT at 3 to 6 months of age.[10] Therefore, it is imperative that infants with CMT are identified early and receive appropriate PT intervention to achieve optimal outcomes.

The Physical Therapy Management of Congenital Muscular Torticollis Evidence-Based Clinical Practice Guideline (CPG) was originally published in 2013 (2013 CMT CPG)[11] and revised in 2018 (2018 CMT CPG)[12] by the American Physical Therapy Association Academy of Pediatric Physical Therapy. The 2018 CMT CPG reflected new research regarding referral, screening, examination and evaluation, diagnosis, prognosis, intervention, consultation, discharge, and follow-up of infants with CMT.[12] Recommendations are organized into 17 action statements based on critical appraisal of the literature. Additionally, the Academy of Pediatric Physical Therapy

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provides resources to support implementation of the 2018 CMT CPG into clinical practice, including educational handouts for parents and caregivers, for healthcare providers, and for clinicians and educators (https://pediatricapta.org/clinical-practice-guidelines/Congenital-Muscular-Torticollis.cfm).

Implementation of the CMT CPG promotes alignment of clinical practice with research evidence leading to improved consistency of care and improved outcomes for infants with CMT. After publication of the 2013 CMT CPG,[11] a survey of pediatric physical therapists found a 93% increase in implementation of the recommended evidence-based practices.[13] Positive perceptions of the 2013 CMT CPG[11] by pediatric physical therapists included the use of flowcharts, validation of examination and intervention approaches, and easier access to synthesized literature leading to improved consistency of care and a standard of best practice.[14] When the 2013 CMT CPG[11] was implemented in a hospital-based outpatient setting, the percentage of infants who achieved full CMT resolution within a 6-month episode of care increased from 42% to 61%.[15]

International practice and the American Physical Therapy Association indicate that CPGs should be updated every 5 years to reflect new research and ensure optimal clinical care. The purpose of this systematic review (SR) is to evaluate current evidence on assessment tools, interventions, and prognosis for CMT to inform the update of the 2018 CMT CPG.[12]

METHODS

Protocol and Registration

This SR followed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA).[16] A written protocol was established prior to the conduct of the review and was registered with PROSPERO, an international database of prospectively registered SRs on health-related outcomes (registration number: CRD42022288129).[17]

Search Strategy

A comprehensive search was completed from January 2017 to November 2021 and rerun in June 2022 by a research librarian of 5 databases: PubMed, Embase, Cochrane, CINAHL, and Web of Science. In addition, the first 200 results from Google Scholar were retrieved in November 2021. Search terms included infant and concepts related to CMT. Searches were not restricted by language. Supplemental Digital Content 1 (available at: http://links.lww.com/PPT/A428) includes the full search strategy by database. References of included studies were examined and experts in the field were contacted to identify additional relevant studies.

Selection Criteria

Studies were included based on the following criteria: (1) participants were children aged birth to 2 years diagnosed with CMT, and if other diagnoses or ages were included, the results of infants with CMT were statistically analyzed separately; (2) studies on the psychometric properties of assessment tools commonly used in the management of CMT; (3) intervention studies on PT management of CMT that statistically analyzed motor outcomes with or without a control group of another motor intervention; (4) qualitative studies on the experiences of parents of infants diagnosed with CMT that relate to PT; (5) surveys of physical therapists managing infants with CMT; and (6) prognostic studies that predicted response to PT intervention. Studies were excluded based on the following criteria: (1) only included infants with plagiocephaly; (2) conference abstracts, dissertations, and literature reviews; (3) previously included in the 2018 CMT CPG[12]; (4) assessment of non-PT interventions, such as acupuncture, botulinum toxin injections, or surgery; and (5) published in a language other than English when an adequate English translation could not be obtained.

Study Selection

Studies were screened using a web-based screening and data extraction tool, Covidence (www.covidence.org), based on title and abstract, using the inclusion and exclusion criteria. If necessary, a full-text review of studies was completed. Two authors reviewed the studies independently and a third author resolved disagreements. Studies that were read in full-text but excluded are listed in Supplemental Digital Content 2 (available at: http://links.lww.com/PPT/A429).

Level of Evidence

Studies were assigned a level of design rigor using criteria from the Oxford Center for Evidence Based Medicine levels of evidence.[18] Levels of evidence range from level I, as the highest, to level V, as the lowest.

Study Appraisal

Two authors independently appraised the studies using a risk of bias tool appropriate to the study type, data were compared for agreement, and a third author resolved disagreements. Risk of bias was assessed using the revised Cochrane Risk-Of-Bias tool (ROB-2)[19] for randomized clinical trials (RCTs), the A MeaSurement Tool to Assess systematic Reviews (AMSTAR-2)[20] and the Risk Of Bias In Systematic reviews (ROBIS)[21] for SRs, the COnsensus-based Standards for the selection of health status Measurement INstruments (COSMIN)[22] for psychometric properties of assessment tools, the quality assessment tool for before-after studies with no control group for pre-/post-studies, Critical Appraisal Skills Programme (CASP) for qualitative studies, and the QUality In Prognosis Studies (QUIPS) for prognostic studies. Sources of funding were assessed to determine conflicts of interest.

Data Extraction

Data extracted from each study were determined by mutual consensus. Data were extracted independently by 2 of 3 authors, compared for agreement, and discrepancies were resolved through discussion.

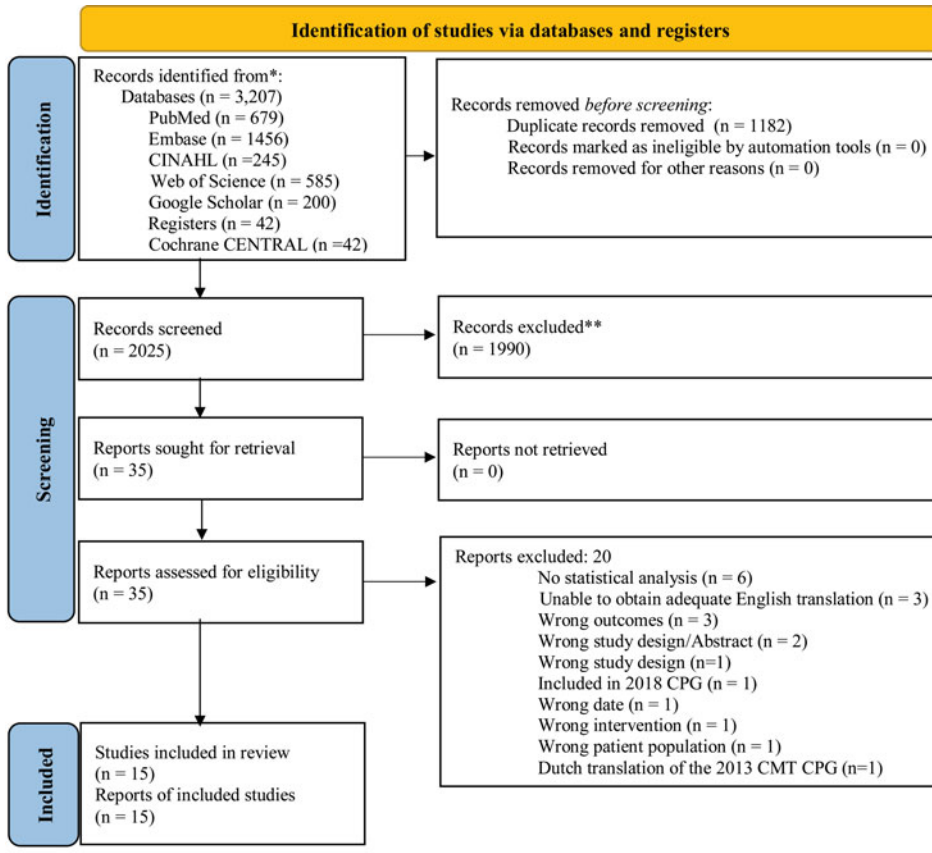

Fig. PRISMA flow diagram.

RESULTS

Search strategy and selection details are in the Figure. Fifteen studies were included in the SR. Four studies informed psychometric properties of assessment tools commonly used in the management of CMT,[23-26] 6 studies informed PT intervention for infants with CMT,[27-32] and 5 studies informed prognosis or predicted response to PT intervention.[33-37] Deviations were made from the original protocol related to the types of quality assessment tools used due to the types of studies included in this review. Funding sources of each article were extracted and no conflicts of interest were identified. Study characteristics are in Table 1, including their level of evidence, study design, and overall risk of bias. Refer to Supplemental Digital Content 3 (available at: http://links.lww.com/PPT/A430), Supplemental Digital Content 4 (available at: http://links.lww.com/PPT/A431), and Supplemental Digital Content 5 (available at: http://links.lww.com/PPT/A432) for risk of bias results.

Psychometric Properties of Assessment Tools

Four studies informed the psychometric properties of assessment tools: 1 SR[25] and 1 study each on the Functional Symmetry Observation Scale,[23] the Therapy Behavior Scale,[24] and the visual estimation of cervical active rotation and head tilt.[26] Table 2 includes data on the participants, assessment tools, psychometric properties, and clinical implications of these studies. Each is briefly reviewed next.

Psychometric Property Systematic Review. This SR on the psychometric properties of tools to assess the cervical spine function of children with CMT[25] had a low risk of bias and reviewed 5 studies on the psychometric properties of 6 assessment tools: the goniometer,[38] electronic pendular goniometer,[25] protractor,[38] Muscle Function Scale (MFS),[39] still photography,[40] and the ROM limitation scale.[41] Studies included 242 infants with CMT younger than 18 months. All studies assessed reliability, 1 study assessed content validity, and 1 study assessed concurrent validity. Still photography for assessing habitual head tilt in supine positions had sufficient reliability (interclass correlation coefficient [ICC] $\geq$ 0.7) and the MFS for assessing side-flexor muscle function in lateral head righting had sufficient content validity and reliability ($\kappa \geq 0.7$); the quality of both studies was excellent.[25] The goniometer, electronic pendular goniometer, and protractor had sufficient intra- and/or interrater reliability (ICC $\geq$ 0.7) for passive cervical spine rotation and/or lateral flexion, but the quality of these studies was rated fair to poor.[25] The ROM limitation scale had sufficient intra- and interrater reliability for passive and active cervical rotation, but insufficient intra- and interrater reliability for passive cervical lateral flexion; however, this screening tool

TABLE 1
Study Characteristics

Authors	Level of Evidence	Study Design	Study Category	Overall Risk of Bias
Rahlin et al[23]	IV	Cohort	Assessment	Poor
Rahlin et al[24]	IV	Cohort	Assessment	Excellent
Seager et al[25]	I	Systematic review	Assessment	Low risk
Seager et al[26]	IV	Cohort	Assessment	Good
Chen et al[27]	I	Systematic review + meta-analysis	Intervention	Low risk
Cui et al[28]	II	RCT	Intervention	High risk
Fenton et al[29]	IV	Retrospective	Intervention	Poor
Oledzka et al[30]	VI	Qualitative	Intervention	N/A
Song et al[31]	II	RCT	Intervention	Some concerns
Zollars et al[32]	IV	Cohort	Intervention	Poor
Kim et al[35]	IV	Retrospective	Prognosis	High risk
Knudsen et al[36]	IV	Retrospective	Prognosis	Moderate risk
Greve et al[34]	IV	Retrospective	Prognosis	High risk
Greve et al[33]	IV	Retrospective	Prognosis	Moderate risk
Song et al[37]	IV	Cohort	Prognosis	High risk

Abbreviations: N/A, not available: RCT, randomized controlled trial.

has only 3 categories of ROM (full, moderate limitation, and severe limitation) and the quality of this study was fair.[25]

Functional Symmetry Observation Scale. The content validity of the Functional Symmetry Observation Scale Version 2 (FSOS-2) for the assessment of spontaneous movement and posture in infants with CMT was investigated.[23] Thirteen physical therapists completed 2 rounds of surveys using Likert scale questions. Content validity for the FSOS-2 was established[23] and overall study quality was rated excellent.

Therapy Behavior Scale. The intra- and interrater reliability of the Therapy Behavior Scale Version 2.2 (TBS-2) for the assessment of infant and toddler behavior during a therapy session was investigated.[24] Two physical therapists either live scored or scored the videos of 10 infants aged 3 to 6 months with CMT during 2 intervention sessions, conducted 1 month apart. The TBS-2 had sufficient intra- and interrater reliability (ICC $\geq$ 0.7)[24]; however, study quality was rated poor due to small sample sizes of raters and infants.

Visual Estimation. The intra- and interrater reliability of the visual estimation of head tilt and active cervical spine rotation in an upright position was investigated.[26] Twenty-six physical therapists scored videos of 31 infants aged 4 to 24 months with CMT during 2 sessions conducted at least 1 week apart.[26] Visual estimation had sufficient intrarater reliability (ICC $\geq$ 0.7) for active cervical spine rotation and head tilt.[26] It also had sufficient interrater reliability for active cervical spine rotation, but insufficient reliability for head tilt[26]; study quality was rated good. No significant correlation was found between physical therapists' experience and intrarater reliability.[26]

Intervention Studies

Six studies informed PT intervention for infants with CMT: 1 SR,[27] 2 RCTs,[28,31] 1 cohort study,[32] 1 retrospective study,[29] and 1 qualitative study on the experiences of parents of infants with CMT.[30] Table 3 includes data on the participants, interventions, results, and clinical implications of these studies.

Traditional Chinese Medicine. A SR with meta-analysis and an RCT informed the efficacy of traditional Chinese medicine (TCM) massage.[27,28] The SR included 6 RCTs and 1 quasi-RCT[27] and had a low risk of bias. Pooled analysis of 2 RCTs showed that TCM massage had similar effects to stretching based on effective rate, the percentage of infants with CMT that improved (risk ratio [RR]: 1.00; 95% confidence interval [CI]: 0.94-1.06, $P = .99$); however, both RCTs in the meta-analysis had a high risk of bias.[27]

The RCT compared the outcomes of 68 infants younger than 1 year with CMT randomized between 2 types of TCM massage, textbook tuina or modified tuina, provided for approximately 20 minutes a day, 6 days a week for 60 days by trained manipulators.[28] After 30 days of intervention, infants in the modified tuina group had a greater effective rate (94.1%) compared with infants in the textbook group (70.6%, $P < .05$).[28] After 30 and 60 days, infants in both groups decreased their SCM mass diameter, but the infants in the modified tuina group had greater improvement ($P < .05$).[28] This study had a high risk of bias due to insufficient information on deviations from intended intervention and increased risk of bias in the outcome measure of effective rate.

Passive Stretching. An RCT compared the outcomes of 61 infants with CMT younger than 3 months randomized into 1 of 3 groups: passive stretching, handling for active and active-assisted movements, or thermotherapy provided for 30 minutes, 3 times a week, by physical therapists, until the head tilt was 5° or less.[31] After intervention, passive cervical rotation ROM was significantly improved in the passive stretching group compared with the other 2 groups, but there was no difference in SCM thickness on the affected side or the ratio of SCM thickness on the affected side compared with the nonaffected side (A/N ratio).[31] The study's risk of bias was rated some concerns due to insufficient information on deviations from the intended intervention.

Neural and Visceral Manipulation. A cohort study compared the outcome of 10 infants younger than 12 months with

TABLE 2

Psychometric Properties of Assessment Tools Commonly Used in the Management of Congenital Muscular Torticollis

Study	Assessment	Participants	Outcome	Intrarater Reliability	Interrater Reliability	Validity
Rahlin et al[24]	Therapy Behavior Scale Ver 2.2	10 infants with CMT, 0-6 mo	Behavior	ICC 0.95 (0.81-0.99) and ICC 0.92 (0.73-0.98)	ICC 0.88 (0.71-0.95)	NA
Rahlin et al[23]	Functional Symmetry Observation Scale Ver 2	13 pediatric physical therapists	Spontaneous posture and movement	NA	NA	Content: Consensus among experts on all items ranged from 85% to 100%.
Seager et al[25] from Chen et al[b]	Electronic pendular goniometer	12 infants with CMT, 0-12 mo	PROM CS ROT	ICC 0.71-0.79[a] Quality fair[a]	ICC 0.87-0.91[a] Quality fair[a]	NA
Seager et al[25] from Klackenberg et al[38]	Standard goniometer	23 infants with CMT, 1-5 mo	PROM CS ROT	ICC 0.98-0.99[a] Quality poor[a]	NA	Concurrent: Standard goniometer or protractor vs still photography ICC 0.74-0.90[c]
Seager et al[25] from Klackenberg et al[38]	Large protractor	23 infants with CMT, 1-5 mo	PROM CS LF	ICC 0.97-0.98[a] Quality poor[a]	NA	
Seager et al[25] from Ohman et al[39]	Muscle Function Scale, Ver 1 described in words, Ver II described in degrees	68 infants with CMT, 0-2 y	Strength CS LF, Ver 1	κ 0.96-0.99[a] ICC 0.94-0.98[a] Quality excellent[a]	κ 0.96[a] ICC 0.92-0.96[a] Quality excellent[a]	Content: Rated positively by panel of experts
			Strength CS LF, Ver II	κ 0.96-0.99[a] ICC 0.93-0.97[a] Quality excellent[a]	κ 0.96[a] ICC 0.90-0.95[a] Quality excellent[a]	
Seager et al[25] from Rahlin and Sarmiento[40]	Still photography	30 infants with CMT, 4.5-16 mo	Head tilt	ICC 0.79-0.84[a] Quality excellent[a]	ICC 0.72-.99[a] Quality excellent[a]	NA
Seager et al[25] from Murgia et al[41]	ROM limitation scale	109 infants with CMT, 0-18 mo	AROM CS ROT PROM CS ROT PROM CS LF	κ 0.72[a] κ 0.73[a] κ 0.41[a], quality fair[a]	κ 0.80[a] κ 0.83[a] κ 0.49[a], quality fair[a]	NA
Seager et al[26]	Visual estimation	31 infants with CMT, 4-24 mo	Head tilt AROM CS ROT	ICC 0.84 ICC 0.85	ICC 0.58, SEM 4.3° ICC 0.79, SEM 5.9°	NA

Abbreviations: AROM, active range of motion; CMT, congenital muscular torticollis; CS, cervical spine; ICC, interclass correlation coefficient; LF, lateral flexion; NA, not applicable; NR, not reported; PROM, passive range of motion; ROT, rotation; SEM, standard error measurement; Ver, version.

[a]Data from Seager et al[26] systematic review.

[b]Data collected from personal communication between Seager et al[25] and the author of the original study.

[c]Reported as reliability in Klackenberg et al[38] and Seager et al[25] articles. Upon further review of data determined to be concurrent validity.

TABLE 3

Physical Therapy Intervention for Infants With Congenital Muscular Torticollis

Study	Participants	Experimental vs Comparison Group	Intervention Parameters	Outcomes	Between-Group Differences	Within-Group Differences	Clinical Implications
Chen et al[27]	95 infants with CMT, 1-12 mo	EG: TCM massage + stretching (n = 48) CG: Stretching (n = 47)	Mo et al, 2010:[46] *F*: 5x/wk *I*: NR *T*: 35-45 min *D*: 16 wk Pang et al, 2014:[47] *F*: 1x/d *I*: NR *T*: 45 min *D*: 8 wk	Effective rate	NS (RR = 1.0, 95% CI = 0.94-1.06, *P* = .99)	NR	For infants with CMT, TCM was no more effective than stretching for overall improvement
Cui et al[28]	68 infants with CMT, <1 y, SCM mass with head tilt	EG: Textbook tuina (n = 34) CG: Modified tuina (n = 34)	*F*: 6 d/wk *I*: NR *T*: ~20 min in EG; NR in CG *D*: 60 d	SCM lump diameter Effective rate	↓ 30 d & 60 d (*P* < .05) ↑ 30 d (*P* < .05)	↓ 30 d & 60 d in EG and CG (*P* < .05) NR	For infants with CMT, modified vs textbook tuina, was more effective for overall improvement and to decrease SCM lump diameter
Fenton et al[29]	10 infants with CMT, ≤9 mo, DP, mand asym	EG: Standard care PT (n = 10)	*F*: 2-4x/mo *I*: NR *T*: NR *D*: 4-9 mo	Ramal height ratio	NA	↑ End of PT (*P* = .005)	For infants with CMT, 4 mo of standard care PT resulted in more symmetrical ramal height ratio
Song et al[31]	61 infants with CMT, <3 mo, >15° head tilt	EG1: Dev exer; passive stretching (n = 21) EG2: Dev exer; active or active-assist exercises (n = 19) EG3: US of affected SCM (n = 20)	*F*: 3x/wk *I*: NR for EG 1&2, EG 3 MHz, 1-5 W/cm^2 *T*: 30 min *D*: <5° head tilt	PROM CS ROT SCM muscle thickness SCM A/N ratio	↑ Between group 1 and other groups (*P* < .05) NS (*P* > .05) NS (*P* > .05)	NR NR NR	For infants with CMT, passive stretching was more effective than active or active assist exercises or thermotherapy for improvement in cervical ROT PROM
Zollars et al[32]	10 infants with CMT, 0-12 mo	EG: NM/VM (n = 10)	*F*: 1x/2 wk *I*: NR *T*: 30-50 min *D*: 8 sessions	AROM CS ROT PROM CS ROT PROM CS LF AIMS BSID-III, SE	NA NA NA NA NA	↑ (*P* = .005) ↑ (*P* = .002) ↑ (*P* = .001) NR NR	For infants with CMT, 8 sessions of NM/VM resulted in improved CS ROT and LF after intervention, which persisted 4 mo post-intervention. At 4 mo post-intervention, gross motor skills and socioemotional development increased from baseline

Abbreviations: ↑, statistically significant increase; ↓, statistically significant decrease; AIMS, Alberta Infant Motor Scale; asym, asymmetry; A/N, affected side compared to the nonaffected side; BSID-III, Bayley Scales of Infant Development-Third Edition; CG, control group; CI, confidence interval; CMT, congenital muscular torticollis; CS, cervical spine; d, day; D, duration; Dev, developmental; DP, deformational plagiocephaly; EG, experimental group; exer, exercise; F, frequency; I, intensity; ipsi, ipsilateral; LF, lateral flexion; mand, mandibular; NA, not applicable; NM/VM, neural manipulation and visceral manipulation; NR, not reported; NS, not significant; PROM, passive range of motion; PT, physical therapy; ROT, rotation; RR, risk ratio; SCM, sternocleidomastoid; SE, social emotional; T, time; TCM, traditional Chinese medicine; US, ultrasound.

TABLE 4

Prognostic Factors for Infants With Congenital Muscular Torticollis

Study	Participants	Outcome Measure	Results (Significant Prognostic Factors)	Results (Nonsignificant Prognostic Factors)	Clinical Implications
Greve et al[34]	445 infants with CMT, 0-16 mo; G1 = 67%, G2 = 25%, G3 = 8%	PT completion with discharge criteria met (G1) vs partial PT with loss to follow-up (G2) vs PT eval and HP with loss to follow-up (G3)	Comparing G1 and G2: ↓ Age at initial examination ↓ PROM CS LF differences ↓ PROM CS ROT differences ↓ MFS score difference Comparing G1 and G3: ↓ AROM CS ROT differences	Gender, race, insurance type, birth order	Infants with CMT who complete PT with discharge criteria met, compared with infants with some PT who were lost to follow-up, had lower age at initial evaluation and less differences between sides in CS PROM and the MFS.
Greve et al[33]	907 infants with CMT, 0-16 mo; G1 = 9%, G2 = 91%	Use of supplemental interventions (G1) vs no use of supplemental interventions (G2)	Comparing G1 to G2: ↑ Age at initial examination ↑ PROM CS LF differences ↑ PROM CS ROT differences ↑ MFS score difference ↑ Number of treatment visits ↑ Treatment duration Differences in CMT classification grade, CMT presentation, plagiocephaly type, and recommended referrals	Gender, ethnicity, race, insurance type, age CMT first noticed, cranial shape, AROM CS ROT, achieving goals by end of intervention	Infants with CMT who received supplemental interventions, compared with no supplemental interventions, were older at initial evaluation, had greater differences between sides in CS PROM and the MFS, had a greater number of PT visits with longer treatment durations, however, they had similar CMT resolution.
Kim et al[35]	1719 infants with CMT, age of diagnosis median: 4 mo, mean (SD): 16.5 mo (20.1); G1 = 53% G2 = 47%	Developmental delay at 42-71 mo of infants with CMT who did not receive PT (G1) and infants who received PT (G2)		Comparing G1 to G2 for combination of all development areas: Adjusted RR = 0.97; 95% CI, 0.93-0.99 at $P = .10$. No differences were found for each individual subtest: communication, gross motor, fine motor, problem solving, personal/social.	The risk of neurodevelopmental delay is not different in children with CMT who did not receive PT, compared with those that did.
Knudsen et al[36]	46 infants with CMT, 0-6 mo, separated by CMT severity grades 1, 2, and 3.	↑ Total units billed ↑ Episode duration ↑ Total visits Unresolved CMT	↑ CMT severity grade (between 1, 2, and 3) ↑ PROM CS ROT restrictions ↑ CMT severity grade (between 1, 2, and 3) ↑ CMT severity grade (between 1 and 3) ↑ Insured by public vs private insurance	CMT severity grade (between 1 and 2) Age at referral, age at initial eval, gender, direction of CMT, plagiocephaly, facial asymmetry, SCM mass, CMT severity	For infants aged birth to 6 mo with CMT, as CMT severity grade increases physical therapy utilization increases.
Song et al[37]	63 infants with CMT, 0-3 mo	↑ Treatment duration	↓ Age ↑ SCM thickness ↑ head tilt angle (PROM CS LF)	Sex Direction of CMT Delivery type Hip dislocation	The duration of treatment for infants with CMT <3 mo of age is correlated with age, thickness of SCM on the affected side, and head tilt angle.

Abbreviations: AROM, active range of motion; CI, confidence interval; CMT, congenital muscular torticollis; CS, cervical spine; G1, group 1; G2, group 2; G3, group 3; HP, home program; LF, lateral flexion; MFS, Muscle Function Scale; NA, not applicable; PROM, passive range of motion; PT, physical therapy; ROM, range of motion; ROT, rotation; RR, risk ratio; SCM, sternocleidomastoid.

CMT before, immediately following, and 4 months after 8 sessions of neural and visceral manipulation, a type of manual therapy that mobilizes specific tissues in the body, including nerves, fascia, joints, bones, organs, and vasculature.[32] Active and passive cervical rotation and passive lateral flexion ROM improved from pre- to post-intervention and at follow-up ($P < .005$) and infant development improved from pre-intervention to follow-up ($P < .04$)[32]; however, the study's risk of bias was poor since there was no control group to differentiate the effect of PT from the effect of time.

Physical Therapy. A retrospective study compared ramal height asymmetry between the affected and nonaffected mandible of ten 3 to 9-month-old infants with CMT before and after 4 to 9 months of PT.[29] Mandibular symmetry improved from pre- to post-intervention[29]; however, the study's risk of bias was high since there was no control group to differentiate the effect of PT from the effect of time.

Experiences of Parents of Infants With CMT. A qualitative study described and compared the experiences of parents of infants younger than 7 months with mild or severe CMT regarding the medical diagnosis and PT management of CMT.[30] The following themes were identified among all parents: unfamiliarity with CMT diagnosis, varying approaches of pediatricians, worrying about diagnoses of CMT and plagiocephaly, needing physical therapist's support and reassurance, managing the home program, appreciating family member's support, dealing with more than CMT, and experiencing additional benefits.[30] The following themes were unique to the parents of infants with severe CMT: reflecting on pregnancy to look for a cause and experiencing anxiety after finding an SCM mass.[30]

Prognostic Studies

Five studies informed prognosis associated with treatment duration, clinical outcomes, and use of supplemental interventions.[33-37] Table 4 includes data on participants, outcome measures, prognostic factors, and clinical implications of these studies.

A cohort study investigated factors influencing treatment duration in 63 infants under 3 months of age diagnosed with CMT.[37] Treatment duration positively correlated with age, thickness of the SCM on the affected side, and head tilt angle.[37] Age and head tilt explained 21% of the variance in treatment duration[37]; however, the study had a high risk of bias due to lack of acknowledgment of confounding variables that could impact the study results.

A retrospective study investigated PT episode of care in 445 infants aged 0 to 16 months diagnosed with CMT across 3 groups.[34] Group 1 (G1) were infants who completed PT with discharge criteria met, group 2 (G2) were infants who partially completed PT and were lost to follow-up, and group 3 (G3) were infants who underwent PT evaluations with home programs and were lost to follow-up.[34] When comparing G1 to G2, G1 was younger at initial examination and had less difference between sides in cervical passive range of motion (PROM) and MFS scores.[34] When comparing G1 and G3, G3 demonstrated less difference between sides in active cervical spine rotation.[34] No significant differences were found in gender, race, insurance type, or birth order.[34] This study had a high risk of bias due to lack of description of procedures for assessment of outcome measures.

A retrospective study investigated the frequency of supplemental interventions use in 907 infants aged 0 to 16 months diagnosed with CMT across 2 groups.[33] Group 1 (G1) received first-choice interventions and supplemental interventions and group 2 (G2) received first-choice interventions only.[33] Compared with G2, G1 was older at initial examination, had greater PROM cervical spine limitations, greater differences in MFS scores, increased number of PT visits, and longer treatment duration.[33] No significant differences were found in gender, ethnicity, race, insurance type, age when CMT first noticed, cranial shape, active cervical spine rotation ROM, and achieving goals by end of intervention.[33] This study had a moderate risk of bias due to lack of consideration for confounding variables when analyzing results.

A retrospective study investigated how PT utilization varies with the assigned CMT severity grade in 46 infants aged 0 to 6 months diagnosed with CMT severity grades 1 to 3.[36] Units billed, episode-of-care duration, and total visits each increased across CMT severity grades 1 to 3, with statistically significant differences between grades 1 and 2 for units billed and total visits and statistically significant differences between grades 1 and 3 for all 3 variables.[36] There were significant correlations between cervical rotation ROM restrictions at initial evaluation and total units billed but not between age and total units billed.[36] When comparing infants with residual head tilts at discharge with infants without head tilts, more infants insured by Medicaid, versus private insurance, had unresolved head tilts (34.8% vs 8.7%).[36] The study had a moderate risk of bias due to the lack of consideration for confounding variables when analyzing results.

A retrospective study compared the difference in neurodevelopmental outcomes between 1719 children with CMT who did and did not receive PT.[35] Although the authors state infants who did not receive PT intervention had a higher risk of neurodevelopmental delay, the meta-analysis was not statistically significant and had an adjusted RR of 0.97 (95% CI, 0.93-0.99, $P = .10$).[35] The study had a high risk of bias due to biased interpretation of the results.

DISCUSSION

The findings from this SR inform 5 action statements of the 2018 CMT CPG.[12] To inform clinical practice, this discussion is organized by each action statement and states whether newer evidence justifies revision of the recommendation or changes to its strength.

Action Statement 8: Examine Body Structures

The 2018 CMT CPG recommends measuring 7 body structures as important components of evaluation for CMT.[12] Newer evidence reaffirms this statement and adds information on 4 of 7 body structures.

The recommendation strength for the assessment of infant posture remains moderate. A newer appraisal supports

that habitual head tilt can be reliably measured with still photography[25]; however, adding pictures to the medical record may not be feasible in some clinical settings. The FSOS-2 has evidence of content validity[23]; further research on other psychometric properties is needed before recommending it for clinical use.

The recommendation strength for measuring cervical PROM remains moderate. Newer evidence supports that goniometry, electronic pendular goniometry, arthrodial protractor, and still photography can be used to reliably measure passive cervical lateral flexion and/or rotation,[25] but this is based on fair- to low-quality studies and further high-quality research is needed. Although the ROM limitation scale[25] and the visual estimation scale[26] were found to be reliable tools for screening passive and active cervical rotation ROM, respectively, they may not be appropriate to use during a PT evaluation of an infant with CMT since they do not have the precision necessary to document differences in passive cervical rotation ROM between sides to assign a CMT severity grade[12] and to document change with intervention.

The recommendation strength for bilateral active cervical ROM remains moderate. Newer evidence is strong for the use of the MFS to measure cervical lateral flexion strength,[25] but the evidence continues to be moderate for active cervical rotation.

The recommendation strength for pain remains weak. The 2018 CMT CPG recommends using the Face, Legs, Activity, Cry, and Consolability (FLACC) Scale to document the infant's pain or discomfort based on expert opinion; the FLACC has 5 items rated 0 to 2.[12] Newer evidence supports that the TBS-2 may have sufficient reliability to measure behavior of infants with CMT during intervention[24]; however, this scale has 11 items rated 1 to 4, which may not be feasible in some clinical settings.

Action Statement 9: Classify the Level of Severity

Newer evidence reaffirms the importance of classifying and documenting the level of CMT severity because units billed, episode-of-care duration, and total visits each increased across CMT severity grades 1 to 3.[36] The recommendation strength remains moderate since less is known about severity grades 4 through 8 to guide practice for infants older than 6 months with CMT.

Action Statement 12: Determine Prognosis

The 2018 CMT CPG states that the prognoses for symptom resolution, episode-of-care, and/or the need to refer for more invasive interventions are related to age of initiation of treatment, severity classification, intervention intensity, presence of comorbidities, rate of change, and home program adherence.[12] Newer evidence reaffirms this statement and adds information on age at initiation of treatment,[34,37] severity classification,[36] and effect of social determinants of health.[36] The recommendation strength remains moderate.

Evidence on age of initiation of treatment and treatment duration continues to be inconsistent, with one study finding a positive association[37] and another finding a negative association.[34] Strong evidence supports that younger age at initiation of treatment results in shorter treatment durations[4-6]; however, severity may be a confounding factor since infants with more severe symptoms may be referred for assessment earlier than infants with less severe symptoms.[42]

Newer evidence on severity classification supports that treatment duration is positively correlated both with cervical rotation PROM restrictions[34] and lateral flexion PROM restrictions,[34,37] and that the first 3 grades of the 2018 CMT Severity Grading Scale[12] are related to units billed, episode-of-care duration, and total number of visits.[36] This may support physical therapists in determining intervention duration since the average intervention duration was approximately 3 months (98 ± 13 days), 5 months (152 ± 29 days) and 6 months (180 ± 29 days) for grades 1 through 3, respectively.[36] However, the average number of visits was just over once a month in each severity group[36]; therefore, a different intensity may affect the intervention duration.

Newer evidence supports that more infants with CMT who were insured by Medicaid, versus private insurance, had unresolved habitual head tilt at discharge.[36] This is consistent with findings that infants with public insurance were significantly older at the time of craniosynostosis surgery compared with infants with private insurance.[43] These suboptimal outcomes may be due to socioeconomic barriers. Therefore, it is important for physical therapists to create a therapeutic alliance that fosters open communication with families they work with, to customize care according to each family's circumstances, and to assist families with accessing and navigating barriers to care.

Action Statement 13: Provide the Following 5 Components as the First-Choice Intervention

The 2018 CMT CPG states that physical therapists should provide and document 5 components as the first-choice intervention for infants with CMT: neck PROM, neck and trunk active ROM (AROM), development of symmetrical movement, environmental adaptations, and parent/caregiver education.[12] Newer evidence affirms this statement and adds information on 2 components.[29-31]

The recommendation to use neck PROM could increase from moderate to strong. Newer evidence supports that passive stretching may be more effective than thermotherapy or AROM for improving cervical rotation PROM,[31] and that PT intervention may minimize or prevent secondary complications of CMT, such as asymmetrical mandibular height.[29]

The recommendation to provide parent/caregiver education continues to have moderate strength. Evidence from the experiences of parents of infants with different severities of CMT highlights the importance of building a strong parent-physical therapist relationship over the episode-of-care, educating the family on CMT, tailoring the home program intensity to meet infant needs and family capacities, and providing strong support and reassurance to parents, especially to those with infants with severe CMT.[30] Larger studies on parental experiences are needed to strengthen the recommendation rating.

Action Statement 14: Provide Supplemental Interventions, After Appraising Appropriateness for the Infant, to Augment the First-Choice Intervention

The 2018 CMT CPG states that physical therapists may provide and document supplemental interventions after evaluating their appropriateness for treating CMT or postural asymmetries, as adjuncts to the first-choice intervention when the first-choice intervention has not adequately improved ROM or postural alignment, when access to services is limited, when the infant is unable to tolerate the intensity of the first-choice intervention, and if the physical therapist has the appropriate training to administer the intervention.[12] This statement is reaffirmed and the recommendation strength remains weak.

Newer evidence supports that manual techniques, such as TCM massage[27,28] and neural and visceral manipulation,[32] may be feasible, but further high-quality research is needed to establish efficacy. This is consistent with previous research that demonstrated the feasibility of myokinetic stretching,[44] and evidence that a home program and soft tissue mobilization, compared with a home program alone, resulted in improved outcomes after 6 weeks of intervention, but no difference after 12 weeks of intervention or at follow-up.[45] An anecdotal finding of a survey of pediatric physical therapists supports that manual techniques were added to the first-choice intervention early in the episode-of-care, in contrast to other supplemental interventions that were used later when the first-choice intervention did not result in the expected outcome.[33] The infants who received supplemental interventions were older with more severe CMT at initiation of intervention; however, their treatment duration was substantially longer.[33] Further high-quality prospective studies that control for CMT severity are needed to determine the efficacy of manual therapy early in the episode-of-care, its added benefits when combined with the first-choice intervention, and its contributions to treatment duration.

Implications for Research

Physical therapy management of CMT would benefit from further research in the following 5 areas. First, studies on whether instructing expectant parents and parents of newborns to prevent asymmetries and CMT results in decreased PT utilization either through earlier referral to PT or reduction in the number of infants who need PT intervention for CMT. Second, studies are needed to determine a valid and reliable method of measuring cervical lateral flexion PROM and how differences in lateral flexion between sides relate to the CMT Severity Classification Scale. Third, prognostic studies on relation of age, CMT severity grades, and other factors on full symptom resolution and episode-of-care are needed to support physical therapists in determining accurate treatment durations. Fourth, studies are needed on the efficacy of supplemental interventions, specifically the addition of manual techniques, compared with the first-choice intervention, and the efficacy of different types of service delivery models, such as telehealth and group programs. Last, studies are needed on infants with relapsing CMT who develop asymmetries, and to determine the risk factors and percentage of infants who will develop relapsing CMT.

Limitations

Limitations of this review are the small number of high-quality studies and heterogeneity of interventions and outcome measures, resulting in an inability to complete a meta-analysis.

CONCLUSIONS

For infants and toddlers aged 0 to 2 years with CMT, this review provides new evidence on the psychometric properties of new and established assessment tools, the feasibility and efficacy of first-choice and supplemental interventions, the experiences of parents of infants with CMT, and prognostic factors associated with treatment duration, clinical outcomes, and the use of supplemental interventions. Although newer evidence on infants with CMT does not warrant a revision of the 2018 CMT CPG[12] recommendations, it does reaffirm 5 of 17 action statements and could increase the recommendation strength to strong for neck PROM as part of the first-choice intervention.

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