# Manual Interventions for Musculoskeletal Factors in Infants With Suboptimal Breastfeeding: A Scoping Review

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#### **Abstract**

Exclusive breastfeeding for the first 6 months, and continuing for at least the first year of life, is strongly recommended. Suboptimal breastfeeding, which is breastfeeding that does not meet these recommendations, is a multifactorial issue. Some authorities, particularly in the nursing and lactation counseling professions, have identified musculoskeletal issues that may interfere with successful breastfeeding. The purpose of this project was to survey the literature on manual treatments to correct musculoskeletal dysfunctions in infants with suboptimal breastfeeding. Our research question was, "Have manual interventions been used to correct infants' musculoskeletal dysfunctions thought to be linked to suboptimal breastfeeding?" We searched PubMed and Index to Chiropractic Literature, from inception through July 2018, as well as relevant gray literature. We assessed quality of randomized controlled trials (RCTs) and cohort studies using modified SIGN checklists, and the overall strength of evidence using GRADE. The search yielded 461 articles, with a final inclusion of 27 articles: 7 expert commentaries, I high-quality RCT, I low-quality cohort, I pilot study, 2 cross-sectional surveys, 5 narrative reviews, and 10 case series or case reports. Combining the 10 case series and reports in our search with 18 discussed in narrative reviews included in our review yielded 201 infants who received manual therapy for nursing dysfunction. No serious adverse events were reported and improvement in nursing ability was observed using various outcome measures, usually maternal report. Based on the GRADE criteria, there is moderate positive evidence for the effect of manual therapy on suboptimal breastfeeding.

#### **Keywords**

spinal manipulation, breastfeeding, manual therapy, infants

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# Breastfeeding Is an Important Public Health Issue

Exclusive breastfeeding for the first 6 months, and continuing for at least the first year of life, is strongly recommended by authorities. These include the American Academy of Pediatrics, the American College of Obstetricians and Gynecologists, the Association of Women's Health, Obstetric and Neonatal Nurses, the US Preventive Services Task Force (USPSTF), and the World Health Organization. The term *suboptimal breastfeeding* is used to describe breastfeeding that falls short of these recommendations.

By this definition, suboptimal breastfeeding is currently highly prevalent globally, with only 40% of infants being exclusively breastfed for the first 6 months.<sup>6</sup> This applies to high-income countries like the United States, where only 25% of infants were exclusively breastfed at 6 months as of 2015.<sup>8,9</sup>

However, similar prevalence is seen not only in other high-income countries, but in middle- and low-income countries as well. <sup>10</sup> In fact, "despite its established benefits, breastfeeding is no longer a norm in many communities." <sup>11(p491)</sup>

This has health consequences for mothers and their babies as well as economic consequences for families and the nation in general. Nine serious pediatric conditions (which may extend beyond childhood) have been linked to suboptimal

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breastfeeding: acute lymphoblastic leukemia, acute otitis media, Crohn's disease, ulcerative colitis, gastrointestinal infection, lower respiratory tract infection requiring hospitalization, obesity, necrotizing enterocolitis, and sudden infant death syndrome. 1 Five serious maternal conditions are associated with suboptimal breastfeeding: breast cancer, premenopausal ovarian cancer, diabetes, hypertension, and myocardial infarction. Based only on outcomes for these conditions, a 2017 economic analysis found that suboptimal breastfeeding costs the United States \$3.0 billion in medical costs, \$1.3 billion in nonmedical costs, and \$14 billion in the cost of premature deaths (which were mostly maternal).

# **Contributing Factors to Suboptimal Breastfeeding**

Suboptimal breastfeeding is of course a multifactorial issue, best addressed by broad health behavioral theories such as the ecological model. 12 The ecological model posits that behavior is influenced at multiple levels: intrapersonal, interpersonal, community, institutional, and public policy. A model proposed by Rollins et al<sup>11</sup> is congruent with this approach, applied to the issues of breastfeeding. It suggests that interventions be applied at each level (structural, setting and individual). 11 Generally, public health agencies emphasize the broader levels of community, institutional and policy. 13 Health care providers tend to work at the intra- and interpersonal levels with patients/clients. The USPSTF, which serves as the gold standard for clinical preventive services, strongly recommends that providers counsel new mothers on breastfeeding.<sup>5</sup> All groups can contribute to solving the problem, and in fact, it is essential that all groups work together in order to provide an environment supportive of optimal breastfeeding. Medical physicians, both family practice and specialties (obstetrics and gynecology and pediatrics), nurses and International Board-Certified Lactation Consultants (IBCLCs) have established recommendations and/or guidelines to operationalize the USPSTF recommendation on breastfeeding counseling. 1,4,14

# Musculoskeletal Dysfunction and **Suboptimal Breastfeeding**

Breastfeeding recommendations often focus on interpersonal, community, institutional and policy levels of intervention. 4,6,8 However, it is important to build individual mother-infant dvad issues into the intervention model as well, as proposed by Rollins and colleagues. 11 On this level, some authorities, particularly in the nursing and lactation counseling professions, discuss musculoskeletal issues that may interfere with breastfeeding at the intra- as well as interpersonal level, such as the effects of positioning the infant at the breast. 15,16 Soft tissue dysfunctions, for example, ankyloglossia (tongue-tie)<sup>17</sup> and congenital torticollis, 18 have also been addressed in the biomedical literature. Ankyloglossia is often treated surgically and has been found to improve the infant's ability to nurse successfully. 17,19,20 The physiology and biomechanics of infants' nursing movements have been investigated and explicated.21-24

However, manual interventions that might improve infants' ability to nurse effectively are not included in current guidelines, other than an acknowledgement of a possible role for ankyloglossia, which is often surgically addressed. Since clinical practice guidelines are based on evidence, it is important to evaluate the current evidence base for manual procedures which may contribute to successful breastfeeding. We chose a scoping review as the most appropriate design for this purpose, because it is "a form of knowledge synthesis that addresses an exploratory research question aimed at mapping key concepts, types of evidence, and gaps in research related to a defined area or field by systematically searching, selecting, and synthesizing existing knowledge."25(pp1292,1294) The purpose of this scoping review is to comprehensively survey the existing literature on manual interventions for musculoskeletal dysfunctions in infants with suboptimal breastfeeding.

#### **Methods**

We followed Arksey's and O'Malley's methodology for scoping reviews,26 as follows.

## Stage 1: Identify the Research Question

Our research question was, "Have manual treatments been used to correct infants' musculoskeletal dysfunctions thought to be linked to suboptimal breastfeeding?"

#### Stage 2: Identify Relevant Articles

Because this is an emergent field, we included a variety of resources. A topic expert (SV) identified the most relevant books and organizations. A health sciences librarian (CW) conducted the literature searches.

Electronic databases: PubMed and Index to Chiropractic Literature. The search strategy was:

(("breast feeding" OR breastfeeding OR "sucking behavior" OR "sucking difficulty" OR "sucking difficulties" OR "dysfunctional suck" OR "sucking dysfunction") AND Humans[Mesh] AND English[lang] AND infant[MeSH])) AND (((((chiropract\* OR osteopath\* OR manipulati\* OR "manual therapy" OR "manual therapies" OR massage) AND Humans[Mesh] AND English[lang] AND infant[-MeSH])) OR ("Complementary Therapies" [Mesh] AND Humans[Mesh] AND English[lang] AND infant[MeSH])) AND Humans[Mesh] AND English[lang] AND infant[MeSH])

- Books on breastfeeding 15,27 were used primarily for background information and reference tracking. *Reference Tracking* from Relevant Texts<sup>15,27</sup>
- Relevant organizations and networks were used for their reference listings:

- International Chiropractors Association Pediatrics Council: http://icapediatrics.com/resources/articles/tongue-tie-and-chiropractic/
- Ankyloglossia Bodyworkers Facebook page: http:// www.ankyloglossiabodyworkers.com/professionalresources.html
- International Association of Tongue Tie Professionals: https://tonguetieprofessionals.org/resources/research/

# Stage 3: Select Sources of Information

Both peer-reviewed and non-peer-reviewed literature and other resources were eligible, including gray literature such as conference proceedings and theses. At least 2 investigators (CH, CW, or AM) screened the articles and other resources for eligibility, resolving disagreements by discussion.

Inclusion:

- English language
- Human participants
- Publications in peer-reviewed journals
- Publications in non-peer-reviewed journals
- Books and book chapters
- Webpages
- Conference proceedings

#### Exclusion:

- Case series and case reports cited in an included systematic review
- Nonrelevant (did not address manual approaches and/or musculoskeletal conditions)
- Video or PowerPoint presentations
- Websites used for advertising purposes

#### Stage 4: Charting the Data

We categorized the included articles in terms of study design. For those with a higher level of design—randomized controlled trials (RCTs) and cohort studies—we evaluated their quality using modified SIGN (Scottish Intercollegiate Guideline Network) checklists. 28,29 Studies were rated in the SIGN checklists as "high quality, low risk of bias," "acceptable quality, moderate risk of bias," "low quality, high risk of bias," or "unacceptable" quality. Tables 1 and 2 list the items in in each checklist and explain the scoring system we used to determine the quality rating. We did not assess the quality of lower level studies (narrative reviews, cross-sectional descriptive surveys, case report/series, preliminary and pilot studies, commentaries). Two investigators (CH and AM) rated the studies separately and resolved disagreements through discussion.

We made conclusions about the overall level of evidence using the GRADE (Grading of Recommendations Assessment, Development and Evaluation) system<sup>30</sup>: http://www.essentialevidenceplus.com/product/ebm\_loe.cfm?show=grade. Table 3 summarizes the GRADE system of assessing the quality of evidence.<sup>30</sup> Two investigators (CH and AM) performed the GRADE assessment independently.

### Stage 5: Collating, Summarizing, and Reporting the Results

At least 2 investigators (CH, AM, OH) extracted data from the selected articles. Disagreements were resolved by discussion.

**Table 1.** Randomized Controlled Trial Modified SIGN (Scottish Intercollegiate Guideline Network) Checklist.

tem Yes/No<sup>a</sup>

- I. The study addressed an appropriate and clearly focused question.
- 2. Group assignment was randomized.
- 3. The sample size was justified by a power calculation.
- 4. Investigators were blinded to patients' group assignment.
- 5. Patients were blinded to group assignment.
- 6. Groups were similar at the start of the trial.
- The only difference between groups was the treatment of interest.
- 8. Outcomes were measured in a standard, valid and reliable way
- 9. A power calculation was used and required sample size
- 10. An intention to treat analysis was performed. Total score<sup>b</sup>

<sup>a</sup>Rating: "Yes" = 1; "No" or unable to tell from the article = 0. <sup>b</sup>Scoring—sum of items as follows: 9-10 = high quality, low risk of bias; 6-8 = acceptable quality, moderate risk of bias; <6 = low quality, high risk of bias.

**Table 2.** Cohort Study Modified SIGN (Scottish Intercollegiate Guideline Network) Checklist.

Item Yes/No<sup>a</sup>

- I. Addresses an appropriate and clearly focused question.
- 2. Groups are similar except for factor of interest.
- 3. Number of people who declined enrollment is stated.
- Likelihood that some patients might have the outcome when enrolled are taken into account in the analysis.
- 5. Attrition in each group stated.
- Dropouts and compliant participants compared by exposure.
- 7. The outcomes are clearly defined.
- 8. Assessment of outcome is made blind to exposure status.
- 9. The method of assessment of exposure is reliable.
- 10. Evidence from other sources is used to demonstrate that the method of outcome assessment is valid and reliable.
- Main potential confounders identified and accounted for in design and analysis.
- 12. Confidence intervals are reported. Total score<sup>b</sup>

<sup>a</sup>Rating: "Yes" = 1; "No" or unable to tell from the article = 0. <sup>b</sup>Scoring—sum of items as follows: 10-12 = high quality, low risk of bias; 6-9 = 1

We summarized the results of the search in figures, tables, and text.

acceptable quality, moderate risk of bias; <6 = low quality, high risk of bias.

#### **Results**

#### Identification of Relevant Articles

Figure 1 shows the results of the search and reasons for exclusions. The search yielded 461 articles, with a final inclusion of

**Table 3.** Rating the Quality of Evidence Using the Grading of Recommendations Assessment, Development and Evaluation (GRADE) System.<sup>a</sup>

Level of Evidence	•	Explanation of Quality Rating
A	High	High level of confidence in the effects of the intervention.  Several high-quality studies with consistent outcomes
В	Moderate	Confidence in the effects of the intervention may change with future research findings  Only one high-quality study or  Several lower quality studies
С	Low	Confidence in the effects of the intervention is very likely to change with future research findings  All studies have severe limitations
D	Very low	Uncertainty about the effects of the intervention  Only expert opinion and/or  No research evidence or  Very low-quality evidence

<sup>&</sup>lt;sup>a</sup>Source: GRADE Working Group 2007 (modified by the EBM Guidelines Editorial Team) http://www.essentialevidenceplus.com/product/ebm\_loe.cfm?show=grade

27 articles for the review. Most of the exclusions made during screening (434 of 461) were made for nonrelevance to the research question. The types of studies represented in the final 27 were: 7 expert commentaries, <sup>18,31-36</sup> 1 RCT, <sup>37</sup> 1 pilot study, <sup>38</sup> 1 cohort study, <sup>39</sup> 2 cross-sectional studies (surveys), <sup>40,41</sup> 5 narrative reviews, <sup>42-46</sup> and 10 case reports/series. <sup>47-56</sup>

#### Quality and Strength of Evidence

Table 4 summarizes the level of evidence for the included studies. We identified 1 RCT; it was a high-quality, fully powered placebo-controlled study.<sup>37</sup> It had positive outcomes in terms of infants improving their ability to latch when compared with the sham (placebo) manipulation group. One low-quality cohort study<sup>39</sup> found that 25 infants with nursing dysfunction had more musculoskeletal issues, on physical examination, than 10 asymptomatic infants who successfully nursed. A proof-of-concept pilot study<sup>38</sup> with 6 infants found that chemical analysis of breast milk pre- and postosteopathic manual therapy showed improved fat content so that it closely resembled the composition of breast milk from successful mother-infant breastfeeding dyads.

Combining the 7 case reports<sup>48-51,53,55,56</sup> and 3 case series <sup>47,52,54</sup> we identified with the 13 case reports and 5 case series described in the 3 included narrative reviews<sup>42-44</sup> on manual therapy for suboptimal breastfeeding, there were a total of 201 infants who received manual therapy for nursing dysfunctions. No serious adverse events were reported and

improvement in nursing ability was observed using various outcome measures, usually maternal report.

Based on the GRADE criteria, the level of evidence is moderate, in a favorable direction, for the effect of manual therapy on suboptimal breastfeeding.

#### Clinical Studies

Randomized controlled trial. The only RCT we identified was a high-quality, low risk of bias study. It investigated the efficacy of osteopathic treatment combined with lactation consultations for infants with biomechanical sucking difficulties.<sup>37</sup> Although the authors referred to it as a single-blind study, not only were the patients' mothers successfully blinded to group status, but the lactation consultant who administered the primary outcome measure was also blinded. The primary outcome measure was the LATCH (Latch, Audible swallowing, Type of nipple, Comfort, Hold) tool, which is used by an expert observer to measure the biomechanical aspects of breastfeeding, including some factors related to the mother (see Table 4). Its interrater reliability is high, and it has been used to identify infants with sucking difficulties since 1994. A visual analog scale (VAS) for pain was used with the mothers to assess nipple pain.<sup>37</sup>

LATCH and the VAS were administered at baseline, prior to randomization and treatment, immediately after treatment, and 2 days later. The treatment consisted of a consultation by the lactation consultant and 1 osteopathic treatment (intervention group) or sham treatment (control group). At the conclusion of treatment, 60% of mothers of control group infants thought they were allocated to the intervention group, while 71% of mothers of infants in the intervention group thought they were allocated to the intervention group (P = .303).

LATCH improved significantly for infants in the intervention group, compared with the sham (control) group. Mothers' nipple pain did not improve significantly, but this may have been due to the short time frame for measurement.<sup>37</sup>

Cohort study. The only cohort study we identified was a retrospective cohort of low quality with high risk of bias. <sup>39</sup> It investigated the association of musculoskeletal dysfunction with suckling dysfunction by comparing the findings on a musculoskeletal examination of neonates with established nursing difficulties with those of neonates who successfully breastfed. Twenty-five infants aged 1 day to 3 months were referred for chiropractic care by health professionals because of suboptimal breastfeeding that had not responded to lactation consultation. Ten infants with successful breastfeeding skills presenting for "well baby" chiropractic care were given the same musculoskeletal examination. The same DC (Doctor of Chiropractic) did all the examinations and was not blinded to the infants' nursing status.

Comparing the examination findings, the cohort with nursing difficulties had many more observed musculoskeletal irregularities of movement compared with the successful nursing group. These dysfunctions included restriction and/or deviation in mandibular excursion; hypertonicity of muscles involved in

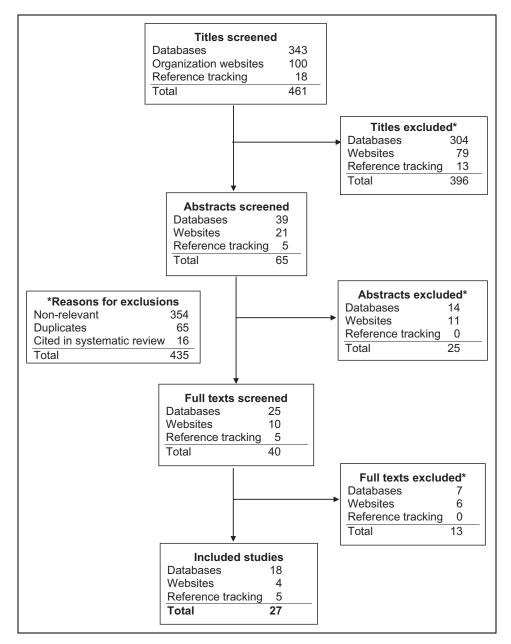


Figure 1. Results of literature search.

suckling and in turning the head; and movement restriction at the cervicocranial junction.<sup>39</sup>

Clinical pilot study. We identified 1 pilot study investigating the change in fat content in breast milk after osteopathic treatment of 6 infants.<sup>38</sup> This small pilot study might be considered a proof-of-concept study. The fat content of breast milk can be measured reliably and simply through a centrifuge method; it is called the "creamatocrit." Because the hind milk (milk produced at the end of each nursing session) is much higher in fat content, the fat content of milk measured with normally feeding infants who completely drain the breast is considered the "gold standard." This pilot study found that the fat content of breast milk was low before osteopathic manual therapy, but increased

so that it was comparable to the level considered normal after the treatment.

#### Case Series and Case Reports

We identified 10 case reports/series (7 case reports and 3 case series) which were not included in the narrative reviews identified in our search. Seven described chiropractic approaches, 2 osteopathic and 1 a lactation consultation. There were 2 case series with 2 patients in each (both twins). 47,52 One of these described hospitalized premature twins who were unable to nurse. To avoid having to surgically place gastrostomy tubes for feeding, osteopathic physicians were consulted. Manual therapy consisting of soft tissue, balanced ligamentous tension,

**Table 4.** Summary of Strength of Evidence Based on Included Clinical Studies (Including Case Series and Case Reports Reported in Included Narrative Reviews).

Study Type, First Author, Year	Topic	Sample	Outcome Measures	Results	Quality of Evidence	Adverse Events Reported
Designed studies						
RCT, Herzhaft, 2017	OMT vs sham OMT for biomechanical sucking dysfunction	97 infants, <6 wk of age	LATCH <sup>a</sup> tool	Latching improved after one OMT treatment	High	None reported
Cohort, Vallone, 2004	Evaluation and treatment of musculoskeletal dysfunction in infants demonstrating difficulty breastfeeding	35 infants	Doctor observation	Neonates with nursing dysfunction had more MSK issues than those without	Low	N/A
Pilot, Fraval, 1998	OMT for infants with sucking dysfunction	6 infants > 3wk and <6 mo of age	Creamatocrit <sup>b</sup> immediately pre-/ posttreatment	Breast milk fat content increased in all, significantly (normalized) in 4/6.	Not rated	Not stated
Large case series >	12 <sup>c</sup>			,		
Miller, 2009	SMT for infants with suboptimal breastfeeding diagnosed by physician or lactation consultant	II4 infants, mean age = 3 wk	Exclusive breastfeeding	All improved; 78% (89/ 114) exclusively breast fed after 2-5 treatments over 2 wk	Not rated	None reported
Vallone, 2004	SMT and CM for infants with MSK and nursing dysfunction	25 infants, mean age = 3 wk	Improved latch by mother report and doctor observation	80% (23/25) improved after mean of 3 treatments (range I-I2)	Not rated <sup>d</sup>	Not stated
Stewart, 2012	SMT for infants with MSK and nursing dysfunction	19 infants	Questionnaire on specific infant actions and overall stress while nursing	Improved in all aspects assessed on questionnaire by mother report	Not rated	Not stated
Summary of small of	case series (<12) <sup>c</sup>		-			
5 case series	SMT and CM for infants with MSK and nursing dysfunction	12 treated, 11 observed only	Mother report and provider observation	Favorable outcomes in treated cases	Not rated	Not stated
Summary of case re	eports <sup>c</sup>					
20 case reports	SMT and CM for infants with MSK and nursing dysfunction	20 infants	Mother report and provider observation	Favorable outcomes	Not rated	Not stated

Abbreviations: RCT, randomized controlled trial; OMT, osteopathic manipulative therapy; CM, cranial manipulation; CS, case series; CR, case report (I case); N/A, not applicable; MSK, musculoskeletal; SMT, spinal manipulative therapy.

myofascial release, inhibition pressure, and osteopathy in the cranial field was applied. The twins' nursing skills progressed until they were able to be discharged without gastrostomy tubes. The third case series described 11 infants with nursing dysfunction in whom the lactation consultant also noted musculoskeletal issues, particularly mandibular asymmetry; however, no treatment was administered. Table 5 details both the 3 case series 47,52,54 and 7 case reports. 48-51,53,55,56

#### Narrative Reviews

Two narrative reviews discussed the literature addressing chiropractic care for nursing dysfunction 42,44 and 1 narrative review, which was a thesis, covered general manual therapy and both chiropractic and osteopathic care for nursing dysfunction. In all these reviews, the emphasis was on the effect of musculos-keletal/biomechanical factors on infants' ability to suckle successfully. Two additional narrative reviews specifically addressed safety issues in manual treatment of infants.

General Manual Therapy Approach. In her 2015 thesis, Cornall summarized the basic theoretical approach of manual therapists, chiefly Beidermann, in Germany. This model is referred to as "kinematic imbalances due to suboccipital stress" (KISS) and is based on the developmental biomechanics of the

<sup>&</sup>lt;sup>a</sup>LATCH is a trained observer-reported instrument measuring latching ability: Latch, Audible swallowing, Type of nipple, Comfort, Hold.

<sup>&</sup>lt;sup>b</sup>Creamatocrit is the fat content in breast milk measured via a standardized process in a hematocrit centrifuge.

<sup>&</sup>lt;sup>c</sup>Cases are detailed in Table 5.

<sup>&</sup>lt;sup>d</sup>Vallone case series was nested within cohort study cited above but was not rated.

Table 5. Summary of Case Series and Case Reports.

Citation	Provider	Patients	Interventions	Treatment Duration	Outcomes	Adverse Events
Case series Collins, 2015	DC	2 (twins) age 7 mo	SMT and CM	I6 wk	Mother-report: reduced breastfeeding difficulties after 8 wk; decreased cranial asymmetry after 16 wk	Not stated
Lund, 2011	DO	2 (premature twins), age 15 wk	SMT and CM	Daily for 2 wk	100% of feeding by nipple after 2 weeks; discharged from hospital at that time	Not stated
Wall, 2006	LC	II neonates	Lactation consultation	No manual treatment	Lower jaw asymmetry correlated with breastfeeding difficulties and also with suspected undiagnosed torticollis	N/A
Case reports Drobbin, 2015	DC	I, age 4 wk	SMT upper cervical spine and TMJ	I treatment	Immediate increase in neck ROM and successful	Not stated
Ferranti, 2016	DC	I, age 2 wk	SMT and CM	CMT 12, CM 10 treatments	breastfeeding Breastfeeding difficulties resolved after 12 SMT; cranial deformation resolved after 10 CM	Not stated
Hubbard, 2014	DC	I, age 7 wk	SMT and CM, probiotic, bile acid and vitamin D supplementation	Weekly treatment for I mo	Complete resolution of breastfeeding difficulties	Not stated
Lavigne, 2012	DC, MD	I, age 3 wk	Examination indicated tongue-tie so patient referred for frenotomy; comanaged with SMT and CM	Not stated	Reduced breastfeeding difficulties; improved mother's nipple symptoms	Not stated
Summers, 2014	DO	I, age 15 days with Pierre Robin sequence	SMT, CM, soft tissue	3 treatments at 3-wk intervals	Sucking and breathing improved; weight gain and resolution of infant's fatigue with breastfeeding	Not stated
Tutt, 2014	DC	I, age 8 mo, with torticollis and plagiocephaly	Instrument-assisted SMT and manual CM; home stretching, light massage and passive ROM	2 treatments	Nursing improved after first treatment by mother's report	Not stated
Williams, 2014	DC	I, age 6 wk, with congenital torticollis and plagiocephaly	Instrument-assisted SMT and manual CM; SCM home stretching	6 treatments over 3 wk	Full cervical ROM and bilateral nursing after initial care, torticollis and plagiocephaly improved at 3 wk	Not stated

Abbreviations: DC, Doctor of Chiropractic; DO, Doctor Osteopathy; LC, lactation consultant; MD, Doctor of Medicine; N/A, not applicable; SMT, spinal manipulative therapy; CM, cranial manipulation; ROM, range of motion; SCM, sternocleidomastoid muscle.

infant's spine, supporting musculature and associated neurological structures. It has been observed to be associated with "unsettled babies" as well as colic and nursing dysfunction.<sup>43</sup>

Osteopathic Approach. The osteopathic literature summarized in 2015 by Cornall<sup>43</sup> was sparse and primarily descriptive and anecdotal. Three case reports and 1 clinical pilot study specifically addressing nursing difficulties were cited. "Osteopathy in the Cranial Field," also called craniosacral therapy, is the primary osteopathic approach used in all the cited studies, although its basis is theoretical and anecdotal.<sup>43</sup>

Chiropractic Approach. Two reviews, published by Alcantara et al<sup>42</sup> in 2015 and Fry<sup>44</sup> in 2014, included most of the same studies, for a combined total of 13 case reports and 5 case series. The cases included only chiropractic treatment of infants with nursing difficulties. Combined, the 5 case series had a total of 166 patients (sum of 2,<sup>59</sup> 6,<sup>60</sup> 19,<sup>61</sup> 25,<sup>39</sup> 114<sup>62</sup>). Consequently, a total of 179 infants were treated with chiropractic care. In all the case reports and most of the case series, the patients' suckling ability was observed to improve. A wide variety of commonly used manual procedures were applied, not only to the cervical spine and cranium (using cranial manipulation as developed and taught within the chiropractic profession

by DeJarnette and Upledger<sup>63</sup>) but also to the entire spine, and to the supporting soft tissue structures as well. 42,44

Biomechanical Forces Used in Manual Therapies for Infants. A review by Todd et al<sup>45</sup> summarized studies of the amount of biomechanical force used in chiropractic manual therapy of infants in terms of appropriateness and safety. European guidelines for chiropractic spinal manipulation of infants recommend low-force, low-speed manipulation that is 10% of the force (equivalent to 11.2 N of force) for infants up to 3 months of age and 30% of the force used for adults for infants and toddlers aged 3 to 23 months (equivalent to 33.6 N). Similar guidelines for different age groups were developed by Marchand et al,  $^{64}$  based on (a) a study of tensile strength and osteoligamentous failure rates in pediatric spines and (b) a report of transient bradycardia and apnea occurring in infants aged 3 months and younger, related to manual application of highvelocity, low-amplitude manipulation of 50-70 to 70 N.65 To prevent possible adverse physiological effects—even if transient—Marchand et al<sup>64</sup> recommend that the maximum application of force for spinal manipulation should be 20 N for neonates and 50 N for children 2 to 23 months of age, compared with 155 N for adults.

Todd et al<sup>45</sup> state that many manual techniques used by DCs are similar to the manual therapies used by other providers. This includes osteopathic craniosacral therapy, which has been measured at a force of 1 N and spinal manipulation/mobilization performed by physical therapists, measured at a force of 22 N in adults. Medical manual therapists have been found to apply manual therapy to children using forces of 50 to 70 N.<sup>45</sup>

Adverse Events Related to Manual Therapy for Infants and Children. Todd et al, 46 in another review, summarized the literature on adverse events in children related to use of manual therapy by any type of provider. Of the 12 articles yielded by their extensive search, an all-time total of 15 serious adverse events was reported. This included 3 deaths. In 1 of these 3 cases, a physical therapist applied electrical current therapy and spinal manipulation to a 3-month-old. In another case, a craniosacral therapist applied craniosacral therapy incorrectly. In the third case, an unspecified provider used spinal manipulation in a child with pneumonia, dislocating the first cervical vertebra. Twelve serious nonfatal injuries were reported to be caused by manual procedures delivered by 7 DCs, 2 physical therapists, 1 each by a medical, osteopathic and an unspecified practitioner. In most cases, high-velocity manipulation using extension and rotation of the spine was used. Preexisting pathology was present in most cases. The authors concluded that published cases of serious adverse events in infants and children due to manual therapies are rare.<sup>46</sup>

#### Cross-Sectional Descriptive Surveys

Prevalence of Ankyloglossia in an Infant Population With Suboptimal Breastfeeding. The mothers of 131 infants with suboptimal breastfeeding presenting to a chiropractic college teaching

clinic were surveyed. Thirty-nine percent of the infants had been previously diagnosed with ankyloglossia and of these, 77% had had a frenulotomy. However, feeding difficulties had persisted. The study concluded that the diagnostic criteria for ankyloglossia may need clarification and that more research on sustained breastfeeding following frenulotomy is needed.<sup>41</sup>

Lactation Consultants' Perceptions of Musculoskeletal Disorders Affecting Breastfeeding and Their Referrals for Manual Therapy. IBCLCs were surveyed in the United States and Canada. Of 13 017 invited, 2457 (18.9%) responded. Most (73.9%) reported referral of infants for musculoskeletal treatment. Pediatricians were the most common referral (47%), followed by craniosacral therapists (16%) and chiropractors (14%). Latch issues were the main reason for referral; tongue-tie (27%), painful latch (24%), neck problems (18%), and non-latching (9%). Congenital torticollis (25%) and neck tension (14%) were the main musculoskeletal problems that the IBCLCs identified. About half the respondents felt they were able to recognize infant musculoskeletal problems. Ninety-one percent noted that breastfeeding improved after manual treatment. 40

#### Expert Opinion/Commentaries

One expert commentary by an orthopedic medical physician who practices manual therapy<sup>36</sup> discussed the KISS theory in the context of infants' sleeping and feeding problems. He explained that the occipitocervical junction (the joint between the occipital bone and the first cervical vertebra) is more vulnerable to excessive mobility in infants and young children due to the structural angle and less developed musculature. His clinical experience has demonstrated that realignment of the cervical joints is associated in improvement in a number of symptoms common in infants and young children.<sup>36</sup>

Three expert commentaries focused on chiropractic care for infants with musculoskeletal dysfunctions contributing to nursing dysfunction. These detailed the examination of an infant for dysfunctional movement of the joints and muscles involved in latching and suckling, particularly related to the cervical spine, mandible and other cranial bones, and muscles of the neck and face. The importance of the cervicocranial junction was emphasized. They also emphasized the importance of interprofessional collaboration, particularly between the DC and the lactation consultant.

Two expert commentaries focused on osteopathic craniosacral therapy, also called "cranial osteopathy." The commentary by Westcott<sup>35</sup> recounted the history of cranial osteopathy, which began in 1900, and was based on Sutherland's observation of subtle movement of the cranial bones. Although his resultant theory on which cranial osteopathy is based has not been scientifically documented, Upledger<sup>66</sup> later argued that clinical results justify its use. Cranial manipulation for infants specifically was discussed by Upledger<sup>66</sup> as well as Sullivan,<sup>67</sup> with reference to the fact that the cranial bones are not fused in infants.<sup>35</sup> Westcott<sup>35</sup> explains that craniosacral

therapy, another term for cranial osteopathy, is practiced by doctors of osteopathy (DOs), DCs, and other health care providers. In her commentary, Gray,<sup>34</sup> a midwife who practices craniosacral therapy, advocates this therapeutic approach "should be in every midwife and lactation consultant's tool bag." She also makes the connection between various musculoskeletal dysfunctions and tongue-tie, and echoed previously mentioned points that the condition is usually surgically corrected but may be associated with musculoskeletal issues which may prevent the infant from successful nursing even after the surgical repair.<sup>34</sup>

One expert commentary specifically addressed the issue of breastfeeding in infants with congenital torticollis. <sup>18</sup> This topic has broader implications, however, because torticollis is often (50% of cases) associated with intrauterine constraint and difficult births and may also result in craniofacial, hip and spinal asymmetries. The author recommends that the physical therapy to correct these asymmetries is most effective when applied in infants under four months of age, with fastest resolution occurring when applied at less than 1 month of age. <sup>18</sup> She states that although lactation consultants may recommend various types of bodywork for infants with torticollis, research on approaches other than physical or occupational therapy, such as chiropractic, are limited, and that such research is needed. <sup>18</sup> She has, in fact, included a chapter by a DC in the latest edition of her book, *Supporting Sucking Skills in Breastfeeding Infants*. <sup>15,63</sup>

#### **Discussion**

This scoping review adds to the evidence base on one aspect of manual therapy for infants, a controversial topic that has even received attention on network television (http://www.abc.net.au/news/health/2016-06-20/newborn-chiropractic-care: where-is-the-evidence/7526116).

Previous literature reviews of the topic of manual therapy for infant musculoskeletal problems linked to suboptimal breastfeeding have been discipline specific and focused on using spinal manipulation as the primary treatment. 42,44 Relevant information about similar procedures and assessments used by other manual therapy professions remains unshared and unknown. Therefore, we searched literature that was oriented toward both chiropractic and osteopathic medicine because the two professions are most closely associated with spinal manipulation and use other manual therapies. We also searched websites of organizations that focus on infants with breastfeeding issues associated with the compensatory musculoskeletal problems resulting from congenital anomalies like ankyloglossia. Casting this wide net resulted in a surprisingly large number of articles and a higher quality of evidence than we anticipated.

#### Limitations of the Study

Because scoping reviews are often used to explore an emergent topic, the quality and quantity of the evidence tends to be lower than it is for well-researched topics, making definitive conclusions inappropriate. Often, scoping reviews do not assess the quality of the included literature, for this reason. However, we decided to offset this limitation by evaluating any randomized controlled trials, cohort studies or systematic reviews identified. Another limitation is that we may have missed some studies, particularly because our search covered several different disciplines. We attempted to mitigate this through using reference tracking in key articles. Another inherent limitation in the broad approach is that there was diversity in the procedures and terminology used, making comparisons difficult or impossible.

#### Key Findings

Relationship of Musculoskeletal Dysfunction to Suboptimal Breastfeeding. Although the procedures for conducting musculoskeletal examinations were often unspecified in the articles, several general sites of musculoskeletal dysfunction were pervasive throughout: the cervicocranial junction, cranial bones, mandible, upper cervical spine and neck muscles, especially the sternocleidomastoid. However, the effect of any of these dysfunctions on breastfeeding success remains theoretical, based on the biomechanics of suckling. Correlation between resolution of musculoskeletal dysfunction and improved nursing was seldom attempted, with the exception of the single cohort study included. <sup>39</sup> Developing general protocols for these examinations, and assessment of intra- and interrater reliability would facilitate future controlled studies.

Measures of Successful and Unsuccessful Breastfeeding. Most of the case reports and series relied on mothers' report and/or doctors' observation. A few studies attempted to develop standardized mother-reported questionnaires, 61,62 but these have yet to be fully validated. In one of these, 62 exclusive breastfeeding was one of the recorded outcomes. Although this is, in fact, the ultimate goal of the musculoskeletal interventions investigated, breastfeeding is a complex activity that includes both maternal and infant factors. Thus, a measurement that isolates the musculoskeletal factor is necessary in order to assess the effect of these interventions. The single RCT in our review used what is considered to be a reliable and valid outcome measure, the LATCH tool. 37 However, even LATCH has been found by others to lack sensitivity to change, <sup>68</sup> and it also measures maternal factors as well as infant factors, which decreases its utility in measuring only infant factors. One 1998 pilot study identified the "creamatocrit" as a way to accurately measure fat content of breast milk, which can be used as a surrogate measure of successful latching/suckling.<sup>38</sup> However, although a more user-friendly instrument than the original centrifuge method has been developed, <sup>69</sup> it does not appear that this method has been used in other studies of suboptimal breastfeeding. Further testing of this method may be warranted.

Blinded Assessment. Only one of all the articles, the RCT, blinded the person conducting the measure to the infants' treatment status.<sup>37</sup> Without this crucial feature, it will be very

difficult, if not impossible, to rule out placebo effects related to the doctor's attention, the hands-on treatment, and possible rater bias.

Common Terminology Among Disciplines. As the importance of interprofessional practice and collaboration continues to grow, discipline-specific terminology must give way to common terminology understood by all related disciplines. Collaborative research among various types of manual therapists as well as with any providers who work with pregnant women and infants could greatly enhance research productivity rather than remaining in their respective research silos.

Safety Issues Related to Manual Interventions. A 2015 review concluded that adverse events caused by manual interventions with children are rare. A 2018 systematic review of manual interventions for infants in particular found that the risk of nonserious adverse events was low. It is nevertheless important to maximize the safety of infants receiving manual care. In most of the articles in this review, authors did not state whether or not there were any adverse effects. It would be helpful if future studies and case reports would explicitly state whether there were any adverse effects, even minor ones. Currently, a surveillance system is being developed by chiropractors to gather data on adverse events among pediatric chiropractic patients.

The 2016 review of biomechanical forces used in manual interventions with infants and children made recommendations based on international consensus among DCs, and included some information about forces used by other manual therapists. 45 A more recent study went further into this topic by measuring biomechanical forces with the aid of sophisticated force-sensing technology and manikins, delivered by a highly experienced DC who specializes in treating infants.<sup>73</sup> The authors found that the DC was in fact capable of modifying biomechanical forces to accommodate an individual patient's needs, and that the forces were similar to those recommended by the international consensus of DCs. 45,64 Currently, most chiropractic educational institutions in North America have instituted the use of "force-sensing table technology"—the equipment used in the 2017 study cited above—to train students in spinal manipulation. 74,75 Additionally, the chiropractic profession has developed a set of "best practices" recommendations for the chiropractic treatment of children, which recommends additional examination, treatment, and referral practices to safeguard pediatric patients.<sup>76</sup>

#### **Conclusion**

Because breastfeeding is a significant public health issue, exploring ways to increase its prevalence and success is important. This scoping review found that there is a moderate level of favorable evidence supporting the use of manual interventions for infants with musculoskeletal dysfunctions and suboptimal breastfeeding. Coupled with the rarity of adverse events noted

for manual treatment of infants and children, additional controlled studies of this topic are warranted.

#### **Author Contributions**

CH contributed toward design, data collection and analysis, writing, and review. AM was involved with data analysis, writing, and review. CW performed the literature search and as involved with writing and review. OH contributed toward data analysis, writing, and review. SV contributed toward design, writing, and review.

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#### **Ethical Approval**

Ethical approval was not required for this scoping review.

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