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REVIEW ARTICLE

Severity and prevalence of ankyloglossia-associated breastfeeding symptoms: A systematic review and meta-analysis

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Abstract

Aim: To evaluate breastfeeding symptoms associated with ankyloglossia/tongue-tie.

Methods: Databases included PubMed, Embase, CINAHL, PsycINFO, Web of Science, and Google Scholar. Eligible studies reported baseline breastfeeding symptoms/severity from tongue-tied infants. Two reviewers independently screened studies, extracted data, and assessed quality. Low-quality studies were excluded. Main outcomes were weighted mean severity scores for dyads with ankyloglossia relative to reference values for successful breastfeeding. Meta-analyses used inverse-variance-weighted random-effects models.

Results: Of 1328 screened studies, 39 were included (5730 infants with ankyloglossia). The mean LATCH score for patients with untreated ankyloglossia, 7.1 (95% CI: 6.7–7.4), was significantly below the good-breastfeeding threshold. The mean Infant Breastfeeding Assessment Tool score, 10.0 (8.2–11.7), was not significantly below the good-breastfeeding threshold. The mean Infant-Gastroesophageal Reflux Questionnaire-Revised score, 18.2 (10.5–26.0), was consistent with gastroesophageal reflux disease. The mean Breastfeeding Self-Efficacy Scale-Short Form score, 43.7 (39.3–48.1), indicated significant risk of cessation of exclusive breastfeeding within 1–3 months. Mean nipple pain was 4.9 (4.1–5.7) on a 0–10 scale, greater than typical scores for breastfeeding mothers without nipple damage. Total prevalence of breastfeeding difficulties was 49.3% (95% CI: 47.3–51.4%). Early, undesired weaning occurred in 20.3% (18.5–22.2%) of cases before intervention.

Conclusion: Ankyloglossia is adversely associated with breastfeeding success and maternal well-being.

Abbreviations: AAP, American Academy of Paediatrics; BSES-SF, Breastfeeding Self-Efficacy Scale-Short Form; EBF, exclusive breastfeeding; GERD, gastroesophageal reflux disease; IBFAT, Infant Breastfeeding Assessment Tool; I-GERQ-R, Infant Gastroesophageal Reflux Questionnaire-Revised; LATCH, Latch, Audible swallowing, Type of nipple, Comfort, Hold; RCT, randomised controlled trial.

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KEYWORDS
ankyloglossia, breastfeeding, gastroesophageal reflux, nipple pain, tongue-tie

1 | INTRODUCTION

Ankyloglossia (tongue-tie) involves a short, tight, or thick lingual frenulum that restricts the tongue's range of motion. This congenital condition affects approximately 8% of infants,[1] and diagnoses quadrupled between 2003 and 2012 in the United States,[2] likely related to increasing concerns over possible consequences. Classical anterior ankyloglossia involves frenulum attachment near the tip of the tongue. Posterior ankyloglossia involves a less visible submucosal attachment that may impair function. Ankyloglossia can interfere with infants' ability to latch onto the breast, which can undermine breastfeeding success, affecting both the child and mother.

Although frenotomy (clipping the frenulum) is increasingly common,[2–4] controversy persists over the benefits and necessity of surgery.[5–10] Criteria for surgical management vary among paediatric practices and providers.[2,3,11,12] This variability is partly due to a lack of standard practices for grading ankyloglossia and identifying symptomatic cases.[1,13] In 2020, the American Academy of Otolaryngology–Head and Neck Surgery released a clinical consensus statement that recognised ankyloglossia as a potential contributor to infant feeding problems and recommended frenotomy when conservative management fails.[10] The panel also agreed that breastfeeding difficulties and pain may resolve without surgery.[10] Multidisciplinary consultation with lactation specialists or speech-language pathologists is recommended[10] and may lower frenotomy rates.[7,11,14,15] Current views of ankyloglossia management leave considerable room for interpretation and variation in practice, leading many mothers to feel frustrated with providers' conflicting advice and care that is not always aligned with their goals for feeding their infant.[16–19]

Research has largely focused on surgical outcomes, with limited attention to the baseline prognosis of ankyloglossia. Unsuccessful latch during breastfeeding may contribute to symptoms such as poor infant weight gain, infant gastroesophageal reflux, maternal nipple pain/damage, and early cessation of exclusive breastfeeding; our review aimed to collate ankyloglossia-associated symptoms. Fuller understanding of the prevalence and severity of adverse breastfeeding outcomes would help inform more standard guidelines for determining when early frenotomy is the best treatment plan.

This systematic review and meta-analysis focused on baseline breastfeeding outcomes of untreated ankyloglossia. The primary objective was to understand symptom severity through meta-analyses of validated outcome measures. Secondary objectives were to determine the prevalence of breastfeeding difficulties in this population and characterise the scope of symptoms.

Key notes

- The relationship between ankyloglossia and breastfeeding difficulties is highly controversial, leading to inconsistent referrals for surgical treatment.
- This systematic review and meta-analysis showed that paediatric ankyloglossia is typically associated with suboptimal breastfeeding, infant gastroesophageal reflux, low maternal breastfeeding self-efficacy, and moderately intense nipple pain; approximately half of mother-infant dyads in this population experienced breastfeeding difficulties.
- Ankyloglossia is a significant condition, and associated symptoms may deter mothers from practicing exclusive breastfeeding.

2 | MATERIALS AND METHODS

This systematic review followed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) 2020 guidelines.[20] The protocol was registered with the International Prospective Register of Systematic Reviews (PROSPERO; CRD42021290644) and is available on their website. This protocol encompassed breastfeeding and non-breastfeeding symptoms of paediatric ankyloglossia; we ultimately separated outcomes into two reviews to accommodate the large quantities of data collected.

The review used the population, exposure, and outcome (PEO) variation of the population, intervention, comparison, and outcome (PICO) framework. The population of interest was mother-infant dyads, the exposure was ankyloglossia, and outcomes were breastfeeding-related symptoms. Under this framework, the review aimed to answer the following questions: what is the severity and prevalence of breastfeeding difficulties associated with untreated ankyloglossia, and what symptoms do mother-infant dyads experience?

2.1 | Search Strategy

A clinical informationist (JN) searched five databases and Google Scholar; databases included PubMed, Embase, CINAHL, PsycInfo, and Web of Science Classic. The search strategy was developed in consultation with the research team. A second informationist peer-reviewed the strategy. According to our broader protocol, searches consisted of terms related to ankyloglossia, lingual frenulum,

tongue-tie, paediatrics, adolescents, and neonates (Table S1). Articles published in English from 1964 onward were retrieved without geographic limits. Databases were searched on October 14, 2021; Google Scholar was searched on October 15, 2021.

Search results were uploaded into EndNote (Clarivate Analytics, London, UK) and duplicates were removed. The file was then uploaded into Covidence systematic review software (Veritas Health Innovation, Melbourne, Australia), which performed follow-up duplicate removal. Remaining duplicates were flagged during screening.

2.2 | Study selection

The PEO criteria guided study selection. Studies were eligible if they involved (1) infants under a year of age with ankyloglossia, or breastfeeding mothers of infants with ankyloglossia; (2) breastfeeding outcomes before/without surgical treatment; and (3) an experimental or observational design. To limit confounding, studies were excluded if patients had major comorbidities or if adult and paediatric data were inseparable. Small case series/reports were excluded as low levels of evidence, as were studies that our quality assessment rated as low-quality. We defined small case series as descriptive accounts of individual cases; we based this criterion on study design, rather than an empirical sample size minimum, to retain small-cohort studies that could contribute higher-quality evidence to the review. Literature reviews, letters, and brief communications were also excluded. Two reviewers (HC, NR) independently screened titles and abstracts for relevance. Two reviewers (HC, GNM) reviewed selected full-text reports for eligibility. A third reviewer (CST or NR) resolved disagreements. This review was conducted in tandem with a review of non-breastfeeding ankyloglossia outcomes in children. Studies without breastfeeding outcomes that fulfilled other eligibility criteria were removed at the end of the screening.

2.3 | Data extraction

Two reviewers (HC, GNM) independently extracted data and resolved disagreements by discussion. Data included study characteristics, demographics, ankyloglossia classification, maternally reported breastfeeding symptoms, breastfeeding cessation, nonexclusive breastfeeding, and quantitative breastfeeding-related outcomes. Data were recorded in Microsoft Excel.

2.4 | Quality and risk of bias assessment

Two reviewers (HC, GNM) independently evaluated the included studies for quality and risk of bias using the National Heart, Lung, and Blood Institute (NHLBI) quality assessment tools. The tools assess 14 questions for controlled trials or observational cohort/cross-sectional studies, and 12 for pre-post studies. Studies were rated high-quality if they failed no more than two questions, medium-quality if they failed 3–5, and low-quality if they failed more than 5.[21] Disagreements were resolved by discussion.

2.5 | Data analysis

Descriptive analyses were conducted using SPSS, version 28.0 (IBM Corporation, Armonk, NY). Symptom prevalence estimates were calculated with binomial 95% CIs. On an outcome-by-outcome basis, studies that did not assess a specific symptom or did not report counts were excluded from that prevalence calculation. Studies were weighted by sample size: prevalence was pooled based on raw counts. Many studies focused exclusively on mother-infant dyads with breastfeeding difficulties (defined here as "symptomatic dyads"). To account for this selection bias while maintaining sample power, relative prevalence estimates of specific symptoms were calculated after uniformly excluding asymptomatic cases from denominators. These relative estimates characterise the distribution of problems that dyads with breastfeeding difficulty experience. Overall prevalence of difficulty breastfeeding was pooled from studies where asymptomatic dyads were eligible for inclusion. We then estimated the total prevalence of each symptom among all dyads with ankyloglossia by multiplying relative prevalence (from symptomatic dyads) by the pooled estimate of breastfeeding difficulty.

Meta-analyses were conducted using the 'meta' package for R Statistical Software (version 4.1.2).[22,23] Quantitative outcomes captured in multiple studies were eligible for meta-analysis. Weighted means and 95% CIs were determined using the inverse-variance method. Between-study variance (τ^2, I^2) was determined using the Paule–Mandel estimator. Random effects models with the Hartung–Knapp modification[24] were used to combine data because no outcomes passed heterogeneity tests. For studies that reported summary statistics as median (IQR), the Luo[25] and Shi[26] derivation methods were used to estimate the mean (SD). For studies that reported baseline outcomes by treatment arm, arms were treated as separate studies in meta-analyses. Forest plots were created in R.

3 | RESULTS

Database searches identified 2427 records. After automated duplicate removal, 1328 records were screened, and 39 studies were included in the systematic review (Figure 1).[16,17,27–63] The studies included 5730 infants with ankyloglossia. Meta-analyses used data from 16 studies.[27–42]

3.1 | Study characteristics and quality

Table 1 summarises study characteristics; Table S2 provides individual characteristics of all included studies.[16,17,27–63] The systematic review included 5 randomised controlled trials (RCTs), 19 pre-post

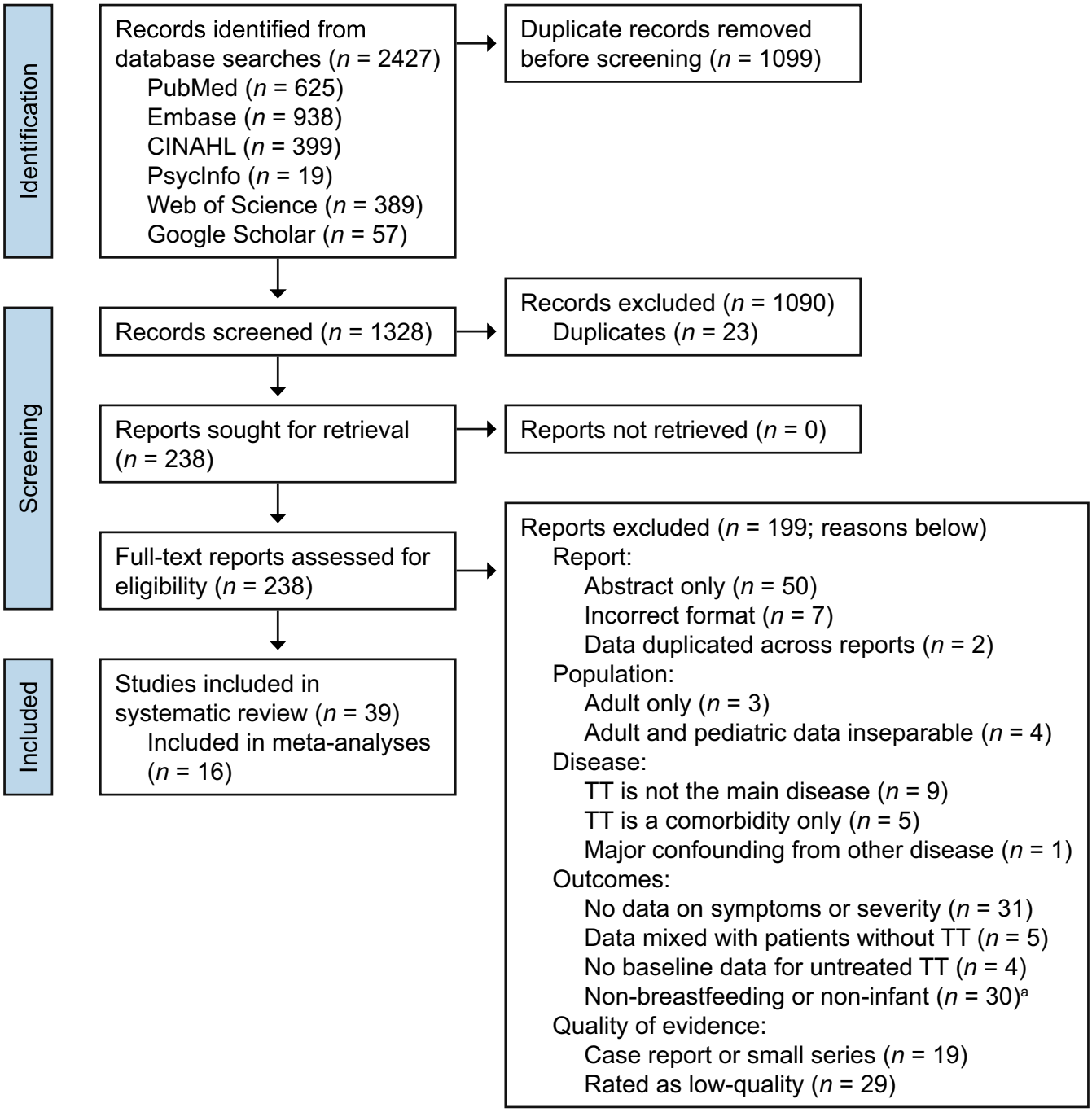

FIGURE 1 PRISMA 2020 flow diagram of study inclusion and exclusion. TT: tongue-tie (ankyloglossia). [a]This systematic review was conducted jointly with a review of non-breastfeeding outcomes; studies without breastfeeding outcomes were ultimately excluded.

intervention studies, 10 observational cohort and cross-sectional studies, 2 case–control studies, and 3 qualitative studies. Most were prospective (n = 35, 89.7%) and most addressed surgical outcomes (n = 33, 84.6%). In 20 studies (48.7%), patients were only eligible for inclusion if they were symptomatic. Publication dates ranged from 1996 to 2021 and research was conducted in 14 countries. Five studies received high-quality ratings and 34 were medium-quality. Twenty-nine studies were excluded due to low-quality ratings (Appendix S1).

Studies used a variety of methods for diagnosing and grading ankyloglossia (Table 1). Most studies (n = 25, 64.1%) used validated criteria. The most common measures were the Coryllos classification system[64] and the Hazelbaker Assessment Tool for Lingual Frenulum Function.[65] Breastfeeding was most commonly assessed by maternal report of symptoms; 19 studies used validated measures (Table 1).

3.2 | Cases and overall symptom prevalence

The male-to-female patient ratio was 1.5:1 (Table 2). For 53.6% of mothers (95% CI: 50.8–56.3%), the patient was her firstborn and

TABLE 1 Summary of study characteristics ($n = 39$ studies)

	n (%)
Design	
Randomised controlled trial	5 (12.8)
Pre-post, prospective	17 (43.6)
Pre-post, retrospective	2 (5.1)
Cohort/cross-sectional, prospective	8 (20.5)
Cohort/cross-sectional, retrospective	2 (5.1)
Case–control, prospective	2 (5.1)
Qualitative	3 (7.7)
Quality assessment[a]	
High-quality	5 (12.8)
Medium-quality	34 (87.2)
Diagnostic method[b]	
Coryllos classification system	12 (30.8)
Hazelbaker Assessment Tool for Lingual Frenulum Function	6 (15.4)
Lingual Frenulum Protocol for Infants	4 (10.3)
Kotlow classification system	2 (5.1)
Bristol Tongue Assessment Tool	1 (2.6)
Unstandardized manual/visual assessment	10 (25.6)
Unvalidated anatomical criteria	5 (12.8)
Not reported	2 (5.1)
Breastfeeding assessment method[b]	
Validated outcome measure	19 (48.7)
LATCH System	8 (20.5)
Breastfeeding Self-Efficacy Scale-Short Form	6 (15.4)
Infant Breastfeeding Assessment Tool	3 (7.7)
Bristol Breastfeeding Assessment Tool	2 (5.1)
Breastfeeding Observation Aid	1 (2.6)
Feeding/Swallowing Impact Survey	1 (2.6)
Frenotomy Decision Tool for Breastfeeding Dyads	1 (2.6)
Clinical Evaluation of Breastfeeding Efficacy Scale	1 (2.6)
UNICEF Breastfeeding Assessment and Observation Protocol	1 (2.6)
Martinelli Breastfeeding Questionnaire	1 (2.6)
Lactation consultant or clinical assessment	3 (7.7)
Maternal report of symptoms	19 (48.7)

[a]Study quality was assessed using the National Heart, Lung, and Blood Institute (NHLBI) quality assessment tools.

[b]Percentages do not sum to 100% because some studies used multiple measures.

first experience breastfeeding. The only comorbidity reported was the presence of an upper labial frenulum (lip-tie) in three studies. Another study provided separate data from the subset without lip-tie; we extracted this subset.

The overall classification ratio of anterior ankyloglossia to posterior ankyloglossia was 1.7:1 (Table 2). Among studies that used the Coryllos classification system, most cases were type 3 or 4, which represent posterior ankyloglossia. Pooling data from 2 studies that used the Kotlow system[66] showed that 25.3% of cases were mild, 51.9% were moderate, 21.0% were severe, and 1.9% were complete.

TABLE 2 Patient population

	n (%)
Sex ($n = 3523$), male	2133 (60.5)
Birth order ($n = 1236$), firstborn	662 (53.6)
Ankyloglossia type ($n = 3442$)	
Anterior	2145 (62.3)
Posterior	1294 (37.6)
Indeterminate	3 (0.1)
Coryllos classification ($n = 2088$)	
Type 1 or 2 (anterior)	944 (45.2)
Type 1	265 (12.7)
Type 2	632 (30.3)
Unspecified Type ½	47 (2.3)
Type 3 or 4 (posterior)	1144 (54.8)
Type 3	597 (28.6)
Type 4	459 (22.0)
Unspecified Type ¾	88 (4.2)
Kotlow classification system ($n = 162$)	
Grade I (mild)	41 (25.3)
Grade II (moderate)	84 (51.9)
Grade III (severe)	34 (21.0)
Grade IV (complete)	3 (1.9)

Note: Percentages are based on available data; any studies with missing values were excluded from individual analyses.

Among studies in which asymptomatic mother–infant dyads were eligible for inclusion, the total prevalence of breastfeeding difficulties was 49.3% (95% CI: 47.3–51.4%; Table 3). By study enrollment, 20.3% of mothers (95% CI: 18.5–22.2%) had weaned the patient earlier than they intended, which mothers largely attributed to ankyloglossia. Mothers breastfed nonexclusively with formula or expressed breastmilk supplements in 33.2% of cases (95% CI: 30.5–36.0%). Table S3 provides source data on breastfeeding symptoms.

3.3 | Infant symptoms

Infants experienced mechanical symptoms that reflected feeding challenges (Table 4). Relative prevalence of inability to latch properly to the mother's breast was 58.1% (95% CI: 56.1–60.0%) among dyads with breastfeeding difficulty. In context of the 49.3% overall prevalence of breastfeeding difficulty, an estimated 28.6% of all infants with ankyloglossia experience poor latch. Relative prevalence of constant feeding among symptomatic patients, including prolonged duration or high frequency of feeds, was 43.4% (95% CI: 40.3–46.5%), yielding an estimated total prevalence of 21.4%. Composite prevalence estimates indicated that 15.6% of all infants

TABLE 3 Pooled Prevalence of breastfeeding difficulty and cessation among all dyads with ankyloglossia

	Dyads affected	*n* of dyads assessed[a]	Prevalence (% [95% CI])
Breastfeeding difficulty	1126	2282	49.3 (47.3–51.4)
Early, undesired cessation (attributed to ankyloglossia)	373	1834	20.3 (18.5–22.2)
Nonexclusive breastfeeding	377	1134	33.2 (30.5–36.0)

[a]Studies that did not report on a given outcome were excluded fully from prevalence estimates. The breastfeeding difficulty analysis only included data from studies in which asymptomatic dyads were eligible.

TABLE 4 Pooled symptom prevalence estimates

	Relative prevalence among dyads with demonstrated breastfeeding difficulty			
Symptom	Dyads affected	Outcome-specific *n* of symptomatic dyads assessed[b]	Relative prevalence (% [95% CI])	Total prevalence point estimate, all dyads with ankyloglossia[a]
Infant symptoms				
Poor latch	1390	2394	58.1 (56.1–60.0)	28.6%
Infant fatigue while feeding	327	697	46.9 (43.2–50.6)	23.1%
Constant feeding	430	991	43.4 (40.3–46.5)	21.4%
Fussiness while feeding	177	431	41.1 (36.5–45.8)	20.3%
Gastroesophageal reflux	273	741	36.8 (33.4–40.4)	18.1%
Poor weight gain	309	871	35.5 (34.2–38.7)	17.5%
Failure to thrive	16	871	1.8 (1.1–2.9)	0.9%
Poor suction/clicks	262	830	31.6 (28.5–34.8)	15.6%
Milk spillage	85	384	22.1 (18.2–26.5)	10.9%
Maternal symptoms				
Nipple pain	1464	2415	60.6 (58.7–62.6)	29.9%
Breast engorgement	188	369	50.9 (45.9–56.0)	25.1%
Nipple damage	651	1580	41.2 (38.8–43.6)	20.3%
Low milk supply	49	595	8.2 (6.2–10.6)	4.0%
Mastitis	63	837	7.5 (5.9–9.5)	3.7%

[a]Composite estimates were determined by multiplying relative symptom prevalence by the estimated total prevalence of breastfeeding difficulties in dyads with ankyloglossia (49.3%, Table 3).

[b]Studies that did not evaluate a given symptom were excluded fully from the prevalence estimate on an outcome-by-outcome basis.

with ankyloglossia exhibit poor suction or clicking sounds while breastfeeding, and 10.9% exhibit frequent milk spillage from the mouth (Table 4).

Breastfeeding difficulties affected patients' quality of life (Table 4). Nearly half of infants with breastfeeding difficulty exhibited fatigue or a tendency to fall asleep while feeding (46.9% [95% CI: 43.2–50.6%]), yielding an estimated total prevalence of 23.1%. Estimated total prevalence of fussiness while breastfeeding was 20.3%. Gastroesophageal reflux affected an estimated 18.1% of all infants with ankyloglossia. More than a third of infants with breastfeeding difficulty gained weight poorly (relative prevalence 35.5% [95% CI: 34.2–38.7%]; estimated total prevalence 17.5%). A subset was diagnosed with failure to thrive (estimated total prevalence 0.9%). Jaundice, choking, hiccups, and gassiness were also noted (Table S3).

Table 5 summarises 5 meta-analyses; Table S4 provides source data. Seven studies[29,31,33,38,40–42] were combined in a LATCH System meta-analysis. An additional study was excluded because it used a modified version of the LATCH System with different domains.[37] LATCH assesses 5 domains: latch to the breast, audible swallowing, nipple type, maternal comfort, and hold/positioning (Table 5).[67] Scores range from 0 to 10; higher scores indicate greater breastfeeding success.[67] The weighted mean LATCH score for patients with untreated ankyloglossia was 7.1 (95% CI: 6.7–7.4; Figure S1). The mean was significantly below the target range of 8–10, which represents good breastfeeding.[68]

Two studies[27,31] were combined in an Infant Breastfeeding Assessment Tool (IBFAT) meta-analysis. An additional study was excluded because it lacked variance values.[61] The IBFAT assesses readiness to feed, rooting behaviours, fixing, and sucking patterns.[69]

TABLE 5 Summary of weighted mean severity scores for ankyloglossia-associated Breastfeeding Difficulties

Outcome measure and scale range	Cases (*n*)	Quality of contributing studies[a]	Heterogeneity[b]	Baseline weighted mean (95% CI)	Reference value
LATCH system (0–10)	657	High (2), medium (5)	$\tau^2 = 0.14$, $I^2 = 78.0\%$ (56.6–88.8), $p < 0.001$	7.1 (6.7–7.4)	Good breastfeeding: ≥8
Infant Breastfeeding Assessment Tool (0–12)	159	High (2)	$\tau^2 = 0.94$, $I^2 = 76.0\%$ (34.0–91.3), $p = 0.006$	10.0 (8.2–11.7)	Good breastfeeding: ≥10
Infant Gastroesophageal Reflux Questionnaire–Revised (0–42)	544	High (1), medium (2)	$\tau^2 = 9.56$, $I^2 = 98.4\%$ (97.1–99.1), $p < 0.001$	18.2 (10.5–26.0)	Infants without reflux symptoms: mean 5.5; infants with GERD (mild, moderate, severe): means 16.1, 19.0, 21.3
Breastfeeding Self-Efficacy Scale–Short Form (14–70)	793	High (3), medium (3)	$\tau^2 = 25.4$, $I^2 = 89.7\%$ (82.1–94.1), $p < 0.001$	43.7 (39.3–48.1)	High risk of exclusive breastfeeding cessation at 1–3 months: ≤58 or ≤50
Nipple pain visual analogue scale (0–10)	1233	High (4), medium (5)	$\tau^2 = 1.33$, $I^2 = 97.9\%$ (97.2–98.4), $p < 0.001$	4.9 (4.1–5.7)	Breastfeeding women with nipple damage: weighted mean 6.2 in the first week postpartum and 5.8 afterward; breastfeeding women without nipple damage: weighted mean 2.7

Abbreviation: GERD: gastroesophageal reflux disease.

[a]Assessed using the National Heart, Lung, and Blood Institute (NHLBI) quality assessment tools. Ratings describe quality, not risk of bias.

[b]I^2 values are reported with 95% CIs.

Scores range from 0 to 12; higher scores indicate greater breastfeeding success.[69] The weighted mean IBFAT score for patients with untreated ankyloglossia was 10.0 (95% CI: 8.2–11.7; Figure S2). The mean did not differ significantly from the target range of 10–12 for good breastfeeding.[69]

Three studies[34,36,39] were combined in an Infant Gastroesophageal Reflux Questionnaire-Revised (I-GERQ-R) meta-analysis (Table 5). Scores range from 0 to 42; higher scores indicate worse symptom severity.[70] The weighted mean I-GERQ-R score was 18.2 (95% CI: 10.5–26.0; Figure S3). The mean was significantly greater than the reference mean of 5.5 for infants without reflux symptoms, and within the range of mean scores for infants with mild-to-severe gastroesophageal reflux disease (GERD; Table 5).[70]

3.4 | Maternal symptoms

Breastfeeding difficulties also affected the maternal quality of life (Table 4). Most mothers with breastfeeding difficulty experienced significant nipple pain (relative prevalence among symptomatic dyads 60.6% [95% CI: 58.7–62.6%]), yielding an estimated total prevalence of 29.9% of all dyads with ankyloglossia. Nipple damage such as bleeding and cracking occurred in 41.2% of cases with breastfeeding difficulty (95% CI: 38.8–43.6%; estimated total prevalence 20.3%). Half of the mothers with breastfeeding difficulty showed breast engorgement (estimated total prevalence 25.1%). Other maternal symptoms included low milk supply and mastitis (Table 4).

The systematic review included three qualitative studies of maternal perspectives on breastfeeding infants with ankyloglossia.[16,17,47] Emergent themes included aspects of maternal well-being: physical pain;[16,17,47] desperation, guilt/shame, and disappointment;[16,17] and strained interpersonal relationships, especially partner resentment.[16] Emergent themes also included frustration with the health care system (distrust, dismissal, and a desire for greater consistency in referral, assessment, and management);[16,17] maternal resilience or persistence;[16,17,47] and relief after frenotomy.[17,47]

Six studies[28,31,34–36,39] were combined in a Breastfeeding Self-Efficacy Scale-Short Form (BSES-SF) meta-analysis (Table 5). Composite scores range from 14 to 70, where higher scores indicate greater maternal confidence in her ability to breastfeed and lower risk of early breastfeeding cessation.[71] The weighted mean BSES-SF score was 43.7 (95% CI: 39.3–48.1; Figure S4). The mean was significantly within the at-risk range based on either of two demonstrated risk thresholds; previous studies identified BSES-SF scores of 58 or 50 as significant cutoffs, where scores below these thresholds predict cessation of exclusive breastfeeding (EBF) within 1–3 months.[72,73]

Nine studies[29–32,34–37,39] were combined in a maternal nipple pain meta-analysis (Table 5) of all studies that used a 0–10 or 1–10 visual analog scale. Any 1–10 scores were converted to a 0–10 scale by linear transformation ($y = 1.11x - 1.11$). The weighted mean 0–10 pain score was 4.9 (95% CI: 4.1–5.7; Figure S5). Mothers in this cohort reported significantly greater pain than breastfeeding women without nipple damage (mean 2.7); the CI did not reach reference means for women with nipple damage (6.2 in the first week postpartum, 5.8

afterward).[74] Reference values are weighted means on a 0–10 scale from a quantitative systematic review; CIs were unavailable.[74]

4 | DISCUSSION

Whereas many studies have aimed to quantify ankyloglossia's impact on breastfeeding by measuring the benefits of frenotomy, this systematic review and meta-analysis aimed to shed greater light on the root of the controversy by characterising maternal–infant experiences at baseline. We recognise that the results of this analysis cannot confirm causality. Beyond ankyloglossia, factors including improper positioning, maternal education and age, comfort with public breastfeeding, prior breastfeeding experience, maternal anatomy and milk supply, smoking, infant health, and pacifier use can affect breastfeeding.[75–77] Disentangling these contributions can be challenging, if not impossible. We fully agree with the AAO-HNS and Academy of Breastfeeding Medicine recommendation that infants with ankyloglossia should receive multidisciplinary expert evaluation before families and providers choose to proceed with frenotomy.[10,78] We believe the associations compiled in our analysis should encourage paediatricians to refer patients more routinely for expert evaluation when ankyloglossia appears to limit tongue function.

This review showed close associations between ankyloglossia and markers of unsuccessful breastfeeding. Prevalence of breastfeeding difficulties was 49.3% (95% CI: 47.3–51.4%). In light of the estimated 8% prevalence of ankyloglossia,[1] this finding suggests that 3.9% of all mother-infant dyads experience breastfeeding difficulties associated with ankyloglossia. Our review identified a spectrum of symptoms related to low breastfeeding success that may the affect quality of life. Mean LATCH scores indicated suboptimal breastfeeding with ankyloglossia, though IBFAT scores were not significantly below the good-breastfeeding threshold. LATCH scores from 8 to 10 predict EBF at 6 weeks with high sensitivity and specificity;[68,79] all seven studies in our meta-analysis showed LATCH scores below 8. A threshold of 8 has shown significantly greater EBF rates and weight gain than the alternative cutoff of 6.[68,79,80] Results of three additional meta-analyses of infant and maternal outcomes were all consistent with breastfeeding difficulty.

Although our review did not estimate a greater prevalence of GERD in this population compared to infants generally,[81] our I-GERQ-R meta-analysis suggested that ankyloglossia is usually associated with clinically significant reflux symptoms. During validation of the I-GERQ-R, mean scores for infants diagnosed with mild, moderate, and severe GERD were 16.1, 19.0, and 21.3, whereas asymptomatic infants scored a mean of 5.5.[70] Our sample's mean was 18.2 (95% CI: 10.5–26.0). The meta-analysis yielded a value on par with reference values for clinically diagnosed GERD, suggesting that infants with ankyloglossia typically experience significant reflux symptoms. We encourage providers to consider evaluating this patient population for GERD.

From our analysis, an estimated 30% of all mothers report significant nipple pain when breastfeeding an infant with ankyloglossia. Mean pain ratings were moderately intense. Although women may experience nipple tenderness or mild discomfort when they begin breastfeeding, a successful breastfeeding dynamic should not be painful. The ankyloglossia-associated mean nipple pain rating was significantly greater than typical ratings from breastfeeding women with undamaged nipples.[74] Mothers also linked feelings of desperation, guilt/shame, disappointment, and relationship stress to ankyloglossia-associated breastfeeding challenges,[16,17,47] all of which may interact with postpartum depression.[82] We hypothesize that the physical and emotional symptoms above likely contributed to low maternal breastfeeding self-efficacy, consistent with our BSES-SF meta-analysis results. Our BSES-SF meta-analysis indicated that dyads with ankyloglossia are typically at risk of EBF cessation within 1–3 months.[72,73]

The American Academy of Paediatrics (AAP) recommends EBF for at least 6 months, followed by two or more years of continued breastfeeding while introducing complementary foods.[83] Breastfeeding has neurodevelopmental and protective benefits for the infant.[83] Breastfed infants have a significantly lower risk of otitis media, respiratory tract infections, asthma, respiratory syncytial virus bronchiolitis, necrotizing enterocolitis, atopic dermatitis, gastroenteritis, inflammatory bowel disease, celiac disease, Types 1 and 2 diabetes, obesity, leukaemia, and sudden infant death syndrome.[83] Maternal benefits include reduced postpartum blood loss and faster uterine involution, along with lower risk of breast and ovarian cancer, rheumatoid arthritis, cardiovascular disease, and Type 2 diabetes.[83] EBF for 6 months is significantly more beneficial than a 4-month duration.[83] However, rates of compliance with the AAP's recommendations are low. Only 25.6% of infants born in the United States in 2017 received EBF through 6 months.[84]

Our systematic review showed that 1 in 5 dyads discontinued breastfeeding earlier than they intended, often before pursuing surgical intervention, due to insurmountable breastfeeding difficulties. Between early cessation and nonexclusive breastfeeding, most mothers in our review were unable to follow the AAP's EBF recommendations despite a desire to do so. Notably, half of our sample lacked prior breastfeeding experience. Although many factors contribute to mothers' decision to breastfeed, ankyloglossia appears to create barriers to compliance with an optimal breastfeeding course.

This meta-analysis complements existing reviews that predominantly focus on surgical outcomes. Several limitations warrant consideration. The included studies preferentially sampled symptomatic dyads and did not provide comparative results from infants without ankyloglossia. We were able to provide comparative results against reference values for our meta-analyses, but prevalence results were descriptive. Heterogeneity also limited the precision of our weighted means. Our review identified a continuing need for high-quality research and for studies that directly evaluate the longitudinal prognosis of ankyloglossia.

5 | CONCLUSIONS

This systematic review and meta-analysis demonstrated that paediatric ankyloglossia is typically associated with suboptimal

breastfeeding, infant gastroesophageal reflux, low maternal breastfeeding self-efficacy, and moderately intense nipple pain. Half of mother-infant dyads in a population with ankyloglossia experienced breastfeeding difficulty. Ankyloglossia is a significant condition, and associated symptoms may deter mothers from practicing EBF. Implications for infant health and maternal well-being must be considered as more consistent guidelines for ankyloglossia management are established.

AUTHOR CONTRIBUTIONS

Ms Cordray screened studies, extracted data, performed quality assessment, conducted data analyses and meta-analyses, and drafted and revised the manuscript; Dr Mahendran screened studies, extracted data, performed quality assessment, and reviewed/revised the manuscript; Mr Tey conceptualised the study, supervised screening, and reviewed/revised the manuscript; Mr Nemeth developed the search strategy, conducted searches, and drafted the search strategy section of the manuscript; Dr Sutcliffe and Dr Ingram supported data acquisition and reviewed/revised the manuscript; and Dr Raol conceptualised and supervised the study, screened studies, and reviewed/revised the manuscript.

CONFLICT OF INTEREST

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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