Frenotomy and Frenectomy with Breastfeeding Support

Draft Evidence Report

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Health Technology Assessment Program (HTA)

Washington State Health Care Authority

PO Box 42712
Olympia, WA 98504-2712
(360) 725-5126
www.hca.wa.gov/hta
shtap@hca.wa.gov

Prepared by:
RTI International–University of North Carolina Evidence-based Practice Center
Research Triangle Park, NC 27709 www.rti.org

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The information in this report is intended to help the State of Washington's independent Health Technology Clinical Committee make well-informed coverage determinations. This report is not intended to be a substitute for the application of clinical judgment. Anyone who makes decisions concerning the provision of clinical care should consider this report in the same way as any medical reference and in conjunction with all other pertinent information (i.e., in the context of available resources and circumstances presented by individual patients).

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The following individuals contributed to this report:
Lead Investigator: Jennifer Cook Middleton, PhD
Co-Investigator: Meera Viswanathan, PhD
Project Coordinator/Research Specialist: Jessica Vaughan, MPH
Research Analyst: Nila Sathe, MA, MLIS
Clinical Consultant: Wade Harrison, MD, MPH
Information Specialist: Mark Howell, MLS
Scientific Reviewer: Leila C. Kahwati, MD MPH
Editing and Document Preparation: Mary Gendron, BA; Alexander Cone, BA

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List of Abbreviations

AMD	absolute mean difference
ARD	absolute risk difference
COE	certainty of evidence
CI	confidence interval
CPG	Clinical Practice Guideline
CQ	cost question
ENT	ear, nose, and throat
EQ	efficacy question
ES	executive summary
FDA	Food and Drug Administration
GRADE	Grading of Recommendations, Assessment, Development and Evaluations
HTA	health technology assessment
IBFAT	Infant Breastfeeding Assessment Tool
IQR	interquartile range
ITT	intention to treat
LATCH	Latch, Audible swallowing, Type of nipple, Comfort, Hold
NA	not applicable
NICU	neonatal intensive care unit
NR	not reported
NRSI	nonrandomized study of intervention
PR	prevalence ratio
QALY	quality-adjusted life year
RCT	randomized controlled trial
ROB	risk of bias
RR	risk ratio
SD	standard deviation
SQ	safety question
SR	systematic review
U.K.	United Kingdom
U.S.	United States

Executive Summary

Structured Abstract

Purpose: To conduct a health technology assessment (HTA) on the efficacy, safety, and cost of frenotomy and frenectomy for breastfeeding support in infants up to 1 year of age with tongue-tie and/or lip-tie.

Data Sources: PubMed (including MEDLINE) and Cochrane from database inception through August 30, 2024; clinical trial registry; government, payor, and clinical specialty organization websites; hand searches of bibliographies, relevant Clinical Practice Guidelines (CPG), and systematic reviews to identify relevant studies.

Study Selection: We sought English-language primary research studies that were conducted in very highly developed countries, that enrolled breastfeeding infants up to 1 year of age with tongue-tie and/or lip-tie, and that compared frenotomy or frenectomy using all methods (e.g., scissor, laser) with all comparators (e.g., sham procedure, wait list control, nonsurgical interventions, complementary and alternative medicine, and observation only). We did not include studies that enrolled infants undergoing tongue-tie and/or lip-tie release procedures for reasons outside of infant breastfeeding (e.g., speech issues, sleep apnea), infants with major comorbidities or other abnormalities, or infants born at less than 37 weeks gestation. Randomized controlled trials (RCTs), nonrandomized controlled trials, cohort studies, crossover studies, and case-control studies that reported efficacy outcomes and safety outcomes (e.g., adverse events, revision surgery) were eligible. In addition, case series were also eligible for safety outcomes. We also sought cost-effectiveness or cost-utility studies.

Data Extraction and Synthesis: One research team member extracted data and assessed the quality of included studies, and a second checked for accuracy. We rated the certainty of the body of evidence for each comparison and outcome using the Grading of Recommendations, Assessment, Development, and Evaluation (GRADE) approach. We narratively synthesized findings, efficacy outcomes were organized by outcomes specific to the mother or the dyad (mother and child) followed by infant outcomes; we grouped safety outcomes by study design, frenotomy method, and oral tie type.

Results: We included 60 studies: 7 RCTs, 6 cohort with comparisons, 47 single-arm/uncontrolled studies, and 0 cost studies. Comparison groups included breastfeeding support, immediate or delayed frenotomy, sham, and no frenotomy. The level of certainty across all graded outcomes was low or very low. Among infants with tongue-tie, frenotomy was associated with improvements in breastfeeding self-efficacy (low certainty of evidence). No differences were found between frenotomy and control for exclusive breastfeeding at 2 months or less in cohort studies or any breastfeeding at greater than two months in RCTs (low certainty of evidence). For maternal breastfeeding and dyad outcomes of breastfeeding pain, breastfeeding effectiveness, any breastfeeding at 2 months or less, exclusive breastfeeding at more than 2 months, improvement in breastfeeding, breastfeeding problems, and cessation of breastfeeding,

certainty of evidence was very low, and we could not determine the direction of effect. Similarly, for the infant breastfeeding outcomes of infant weight gain, infant breastfeeding assessment scores, and infant gastroesophageal symptoms, certainty of evidence was very low, and we could not determine the direction of effect.

We identified no evidence that examined the effectiveness or comparative effectiveness of frenotomy for tongue-tie with concomitant lip-tie or lip-tie alone.

The level of severity for safety outcomes ranged from studies reporting no harms or adverse events, minor harms including bleeding or crying, or serious harms including accidental cut to the tongue and salivary duct damage. Most comparative studies only reported overall study complication rates or reported that no harms occurred. Only 1 study provided comparative data on harms of frenotomy for tongue-tie, and no differences were reported between frenotomy and control on rates of complications. Other safety data were obtained from single-arm studies.

We identified no evidence that examined the cost-effectiveness or cost-utility of frenotomy for tongue-tie with or without lip-tie or lip-tie alone.

Limitations: Most efficacy studies were small; the median sample size was 58. Some studies were unable to maintain blinding of mothers. Both planned and unplanned crossover occurred, limiting measurement of long-term outcomes. Additionally, it was difficult to determine the level of exposure to other interventions that could impact outcomes (e.g., interaction with lactation consultants or other breastfeeding assistance). Most safety studies were single-arm or comparative studies that only provided overall harms data for the entire study sample. Safety studies also lacked detailed measurement information and consistency in how harms and complications were classified. We did not evaluate unpublished data, and we did not consider efficacy outcomes from uncontrolled studies.

Conclusions: We identified methodologically limited evidence for evaluating the efficacy and safety of frenotomy for breastfeeding support in infants up to 1 year of age with tongue-tie and/or lip-tie and no evidence reporting on cost-effectiveness.

ES 1. Background

ES 1.1 Condition Description

Ankyloglossia, colloquially known as “tongue-tie,” is a condition that limits the movement of the tongue[1] and can cause difficulty with breastfeeding in the newborn.[2] “Lip-tie” refers to a similar condition in which the maxillary labial frenum connecting the upper lip to the maxillary alveolar ridge restricts movement and similarly causes difficulty with breastfeeding.[3]

Estimates of ankyloglossia in infants vary from less than 1% to about 11%, with prevalence more common among males than females.[4-6] Reasons for the wide variance in prevalence arise from unclear diagnostic methods, which may include visual inspection of the oral anatomy,

assessment of functional impairment and decreased mobility, and the effect on mothers during breastfeeding (such as nipple pain[6]).

ES 1.2 Disease Burden

Diagnosis of ankyloglossia in infants and rates of frenotomy have increased sharply over the past 2 decades. Diagnoses of ankyloglossia in the United States increased from 3,377 in 2004 to 13,200 in 2019, and lingual frenotomy increased from 1,483 in 2004 to 6,213 in 2019.[7] Reasons suggested for the increase include efforts to support breastfeeding, increased awareness of ankyloglossia and its role in breastfeeding, and the increased role of pediatricians as proceduralists.

Outcomes potentially associated with untreated ankyloglossia include breastfeeding difficulties that may result in restricted weight gain in the infant,[8-11] speech difficulties and problems with dentition,[12,13] maternal pain, reduced milk supply, or incomplete breast emptying that may result in infections.[14,15]

ES 1.3 Technology Description

Frenectomy, frenotomy (also called frenulotomy), and frenuloplasty are sometimes used interchangeably but refer to different procedures. Lingual frenotomy—conducted with a laser, electrocautery, scalpel, or surgical scissors[16]—refers to surgical procedures to release the tongue-tie; lingual frenectomy refers to the removal of the lingual frenulum; and frenuloplasty (also called z-plasty) refers to plastic surgery of the tongue, often used during repeat procedures or clinical scenarios requiring a complex approach.[16,17] Labial frenectomies (and related procedures) refer to procedures to release the frenum that connects the lips to the gums. Going forward, we use the term "frenotomy" to refer to all such procedures.

ES 1.4 Regulatory Status

The U.S. Food and Drug Administration (FDA) does not regulate the performance of these procedures. However, tools such as lasers and electrocauterizers used in the procedure may have premarket authorizations that mention frenotomies.[18]

ES 1.5 Policy Context

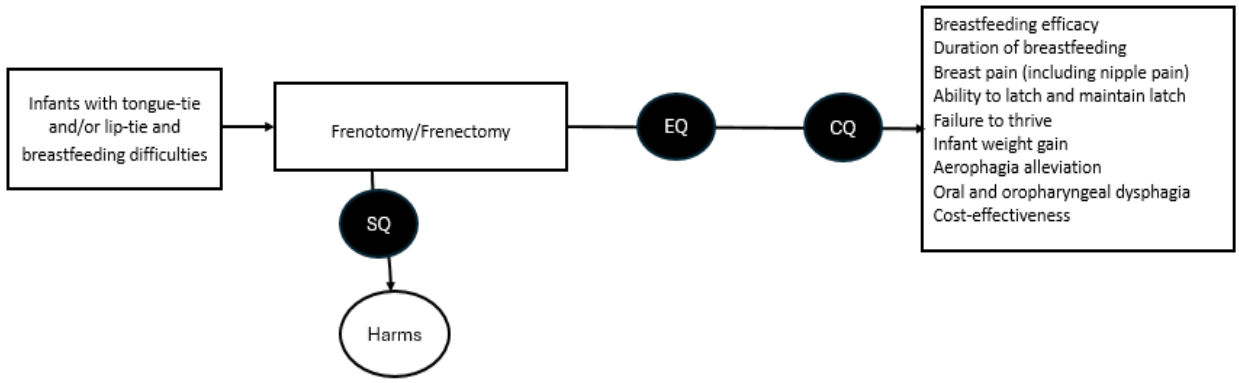
The State of Washington Health Care Authority selected frenotomy and frenectomy with breastfeeding support for a health technology assessment (HTA) because of high concerns for efficacy and medium concerns for safety and cost.

ES 2. Methods

This section describes the methods we used to conduct this HTA.

ES 2.1 Research Questions and Analytic Framework

Figure ES-1. Analytic framework for HTA on frenotomy and frenectomy with breastfeeding support



Abbreviations: CQ = cost question; EQ = efficacy question; SQ = safety question.

Efficacy Question (EQ). What is the effectiveness and comparative effectiveness of frenotomy or frenectomy for tongue-tie and/or lip-tie on breastfeeding outcomes?

Safety Question (SQ). What are the harms of frenotomy or frenectomy for tongue-tie and/or lip-tie as a support for breastfeeding?

Cost Question (CQ). What is the cost-effectiveness of frenotomy or frenectomy for tongue-tie and/or lip-tie for breastfeeding support?

ES 2.2 Data Sources and Search

We searched MEDLINE (via PubMed) and the Cochrane Library for relevant articles and clinical trials. Search dates ranged from database inception to August 30, 2024. In addition, we reviewed reference lists of relevant studies, systematic reviews, and practice guidelines to identify any relevant research studies not identified through the electronic search. The detailed search strategy is in ***Appendix A.***

ES 2.3 Study Selection

Two review team members independently screened all titles/abstracts and full-text articles based on the following study selection criteria. Complete details are in ***Table 1*** of the Full Technical Report.

- Population: Breastfeeding infants up to 1 year of age with tongue-tie and/or lip-tie.
- Intervention(s): Frenotomy, frenectomy, frenulotomy, frenuloplasty, or z-plasty to improve breastfeeding using all methods.

- Comparator(s): For the EQ, comparators included other surgical approaches, nonsurgical interventions, complementary and alternative medicine, and observation only. For the SQ, no comparators were necessary. For the CQ, any comparator was eligible.

- Outcomes: Efficacy outcomes, including breastfeeding outcomes, such as breastfeeding issues and breast pain or discomfort, and feeding issues. Infant outcomes included weight gain, aerophagia, swallowing function, and failure to thrive. Safety outcomes included any harms, such as surgical site complications, readherence of tongue- or lip-tie, need for further surgery/revision, and hospital/emergency department visits. For the CQ, eligible outcomes were cost-effectiveness or cost-utility.

- Timing: Efficacy outcomes were limited from after intervention/comparator through 12 months of age. There were no timing limits for the SQ or the CQ.

- Setting: Inpatient or outpatient pediatric care, operating room, newborn nursery or neonatal intensive care unit, ear, nose, and throat clinic, primary care outpatient, dental office, or breastfeeding medicine clinics. The country where the study took place had to be categorized as "very high" on the 2023/2024 United Nations (UN) Human Development Index.[19] for the EQ and the SQ. Only studies using cost inputs from the United States were eligible for the CQ.

- Study design(s): Randomized controlled trials (RCTs), nonrandomized controlled trials, cohort studies with comparisons, crossover studies, and case-control studies were considered for the EQ and the SQ. Case series were also considered for the SQ. Cost-effectiveness or cost-utility studies were considered for the CQ.

- Other: We included publications in English. Only full text articles reporting original research were considered.

ES 2.4 What Is Excluded from This HTA

This review did not include studies published in languages other than English or conducted in countries that are not rated as "very high" on the 2023/2024 UN Human Development Index.[19] This review did not include studies on tongue-tie release procedures done for reasons outside of infant breastfeeding, including speech issues or sleep apnea issues, which may appear in older children and adults. This review also does not include studies conducted among infants with major comorbidities or other abnormalities, in particular craniofacial abnormalities, or infants born at less than 37 weeks gestation. This review does not include studies with no comparison group (including pre-post studies) for the EQ, although these study types were considered for the SQ and the CQ.

ES 2.5 Data Abstraction and Risk of Bias Assessment

One team member abstracted relevant data from eligible studies and conducted risk of bias (ROB) assessment, which were both checked for accuracy by a second team member. We used the Cochrane Risk of Bias (RoB 2) tool[20] to assess the ROB for included RCTs. We used the ROBINS-I tool[21] to assess ROB for nonrandomized comparative studies (i.e., cohort studies with comparisons). To assess the quality of case reports and single-arm studies evaluated for the SQ, we used a modified version of the tool developed by Murad et al.[22] To assess bias in cost-effectiveness and cost-utility studies, we planned to use a grading system developed by Chiou et al.[23]

ES 2.6 Data Synthesis and Quality of Evidence Assessment

We qualitatively synthesized study characteristics and results in tabular and narrative formats. We graded the certainty of evidence for each comparison using the Grading of Recommendations Assessment, Development, and Evaluation (GRADE) approach,[24] which requires ratings for ROB, consistency, directness, and precision. We based ROB ratings on ROB 2 and ROBINS-I ratings, as described above. Additionally, when we noted errors in reporting we attempted to contact study authors and downgraded the body of evidence for those outcomes when corrected data was not obtained.

To assess the consistency domain, we evaluated both the consistency in the direction and magnitude of effect for a statistically significant difference between intervention and comparator groups, or consistently supported no statistically significant difference.

To assess directness, we considered whether outcome measures were validated. Unvalidated measures were downgraded.

To assess precision, we used a minimally contextualized approach, where the target of certainty was the null effect.[25] Outcomes with confidence intervals that excluded the null were not downgraded for precision unless the effect sizes were implausibly large (e.g., relative risk reduction or relative risk increase exceeding 30%). In such instances, we evaluated whether the ratio of the confidence interval exceeded 3 for relative risks and 2.5 for odds ratios for categorical outcomes, or if the sample size was smaller than 30% to 50% of the optimal information size for continuous outcomes. In such instances, we downgraded outcomes for precision by 2 levels. When outcomes had confidence intervals that included the null, we considered whether the confidence intervals included both appreciable benefit or appreciable harm (25% relative increase or reduction for categorical outcomes, 1 standard deviation for continuous outcomes). If this was the case, we downgraded for precision by at least 2 levels. When confidence intervals were not available or calculable, we relied on reported information such as *p* values and sample sizes.

Two team members independently graded each body of evidence, and we resolved discrepancies through discussion. With GRADE, the certainty of evidence can be graded as "very low," "low," "moderate," or "high." ***Table 2*** in the Full Technical Report defines these levels.

ES 3. Results

ES 3.1 Literature Yield

Of 1,131 unique citations, we included 60 studies reported in 59 articles, representing 7 RCTs,[8,26-31] 6 cohort studies with comparison groups in 5 articles,[32-36] and 47 single-arm studies[11,37-82] for the EQ and the SQ. No studies were identified for the CQ.

Comparison groups included breastfeeding support,[8,28,30,31,33-35] immediate or delayed frenotomy,[8,28-30] sham,[26-28] and no frenotomy.[32-36] Six of the 7 RCTs offered frenotomy to the control group, either immediately (2 studies[26,28]) or within 2 weeks (4 studies[8,27,29,30]). The 2 RCTs that planned immediate crossovers had 100% crossover rates. For those offering delayed crossover, 67% to 96% of the control arm chose to have frenotomies. For these RCTs, we did not analyze outcomes reported after the planned crossover.

One trial (FROSTTIE) did not plan crossovers; the comparator was breastfeeding support only.[31] By the end of the trial, 73% of the control arm had received a frenotomy. We included all relevant outcomes and rated them as high risk of bias.

Of the comparative studies, we assessed 2 as low ROB,[27,28] 0 to have moderate ROB, and 10 to have high ROB.[8,29-36] One RCT[26] had low ROB for the outcome of breastfeeding pain but high ROB for the outcome of breastfeeding improvement.

Specific concerns for high ROB RCTs included flaws in randomization, departure from intended interventions, bias in measurement, and attrition. Specific concerns for high ROB nonrandomized studies included lack of controls for confounding and attrition.

For single-arm studies, although all studies appeared to have included representative populations and adequately measured exposures, only 1 study ruled out alternative explanations for the outcome.[70] The majority of studies (n=38) did not ascertain the outcome adequately or follow up patients long enough for harms to occur. As a result, these studies do not support a robust estimate of potential harms associated with frenotomy.

ES 3.2 Efficacy

Thirteen studies (12 publications)[8,26-36] reported on efficacy outcomes. Of these, all 13 studies reported maternal breastfeeding outcomes (pain, effectiveness, self-efficacy, initiation, exclusivity, and change)[8,26-36] and 4 reported on breastfeeding-related infant outcomes (weight gain, infant breastfeeding behavior, and gastroesophageal symptoms).[27 566,29-31] Serious or very serious ROB, inconsistency, imprecision, and infrequently, indirectness of the outcome measure resulted in low or very low certainty of evidence across all outcomes (***Table ES-1***).

Table ES-1.Summary of findings and certainty of evidence ratings for efficacy outcomes

№ of Studies (№ of participants)	Risk of Bias	Inconsistency	Indirectness	Imprecision	Summary of Findings	CERTAINTY/ Direction of Effect
Breastfeeding pain						
6 RCTs[26-31] (452)	Very serious[a]	Serious[b]	Not serious	Serious[c]	Inconsistent changes in pain scores. Variations in outcome measures and comparators, and timing precluded additional syntheses	⊕◯◯◯ VERY LOW
Breastfeeding effectiveness						
1 RCT[29] (105)	Serious[d]	NA—single study	Not serious	Very serious[e]	No significant difference between frenotomy vs delayed frenotomy (LATCH score of 1 [interquartile ranges from 0–2] in both arms; *P*=0.52 at 5 days)	⊕◯◯◯ VERY LOW
Breastfeeding self-efficacy scale						
3 RCTs[29-31] (312)	Very serious[f]	Not serious	Not serious	Not serious	Significantly larger changes for frenotomy vs. control (delayed or no frenotomy) in 2 of 3 studies; Study 1 (delayed frenotomy)[29]: median change [IQR]: 9 (1.8 to 12.3) vs. 1 (−4 to 7.5); *p*=0.002 at 5 days Study 2 (delayed frenotomy with breastfeeding support)[30]: mean change 13.4 vs. −1.0; 95% CI, 9.2 to 19.7; *P*<0.001 at 10 days Study 3 (No frenotomy with breastfeeding support)[31]: median difference at 3 months, −0.3; 95% CI, −5.2 to 5.8, after significant unplanned crossover	⊕⊕◯◯ LOW for benefit
Any breastfeeding at 2 months or less						
1 RCT[29] (105)	Very serious[g]	NA—single study	Not serious	Very serious[h]	No significant differences at 5 days for frenotomy vs. delayed frenotomy (48/53 [91%] vs. 44/52 [85%], OR, 0.57; 95% CI, 0.17 to 1.88)	⊕◯◯◯ VERY LOW
1 cohort with comparison[33] (159)	Very serious[i]	NA-single study	Not serious	Serious[j]	No difference at 1 month follow-up for frenotomy vs. no frenotomy (114/120 [95%] vs.33/39 [85%]; calculated PR, 1.12; 95% CI, 0.98 to 1.29)	⊕◯◯◯ VERY LOW
Any breastfeeding at more than 2 months						
1 RCT[31] (163)	Very serious[k]	NA—single study	Not serious	Not serious	No statistically significant differences for frenotomy vs. no frenotomy ITT (outcomes with significant unplanned crossover) 3 months: 67/80 (88%) vs. 75/89 (86%); aRR, 1.02; 95% CI, 0.90 to 1.16; *p*=0.73	⊕⊕◯◯ LOW No difference

№ of Studies (№ of participants)	Risk of Bias	Inconsistency	Indirectness	Imprecision	Summary of Findings	CERTAINTY/ Direction of Effect
					6 months: 55/66 (83%) vs. 60/71 (85%), aRR, 0.98; 95% CI, 0.84 to 1.14 Per protocol (at 3 months) n=65/75 (90%) vs. 16/24 (27%); aRR, 1.27; 95% CI, 0.99 to 1.64; *p*=0.06	
4 cohorts with comparison[32,33,36] (1 publication reports 2 studies[32]) (471)	Very serious[l]	Not serious	Not serious	Serious[m]	Similar prevalence between study arms (frenotomy vs. no frenotomy) Study 1[33]: At 3 months, 112/120 [93%] vs. 31/30 [79%]; calculated PR, 1.17; 95% CI, 0.99 to 1.39) At 6 months, 110/120 [92%] vs. 31/39 [79%]; calculated PR, 1.15; 95% CI, 0.97 to 1.36 Study 2[36]: At mean 6 to 7 months, 68/82 (83%) vs. 6/9 (67%); calculated RR, 1.24; 95% CI, 0.78 to 1.99 Study 3(Dixon et al 2018, study 1)[32]: At median 87 days, 127/164 (77%) vs. 18/22 (82%); calculated PR, 0.94; 95% CI, 0.76 to 1.17 Study 4(Dixon et al 2018, study 2)[32]: At median 118 days, 24/34 (71%) vs. 1/1 (100%), calculated Peto OR, 0.247; 95% CI, 0.003 to 18.89	⊕◯◯◯ VERY LOW
Exclusive breastfeeding at 2 months or less						
2 RCTs[29,31] (268)	Very serious[n]	NA—single study	Not serious	Very serious[i]	No statistically significant differences for frenotomy vs. no frenotomy Study 1 at 5 days follow-up: 35/53 [66%] vs. 38/52 [73%]; OR, 1.40; 95% CI, 0.60 to 3.22)[29] Study 2 at 1 to 2 weeks follow-up (35/80 [45%] vs. 43/89 [49%]; aRR, 0.92; 95% CI, 0.59 to 1.45)[31]	⊕◯◯◯ VERY LOW
1 cohort with comparison[33] (159)	Very Serious[o]	NA-single study	Not serious	Not serious	No difference at 1 month follow-up for frenotomy vs. control (88/120 [73%] vs. 30/39 [77%]; PR, 0.95; 95% CI, 0.78 to 1.17)[33]	⊕⊕◯◯ LOW No difference

№ of Studies (№ of participants)	Risk of Bias	Inconsistency	Indirectness	Imprecision	Summary of Findings	CERTAINTY/ Direction of Effect
Exclusive breastfeeding at more than 2 months follow-up						
1 RCT[31] (163)	Very Serious[n]	NA-single study	Not serious	Very serious[i]	No difference at 3 months for frenotomy vs. no frenotomy (38/80 [54%] vs. 39/89 [53%]; aRR,1.03; 95% CI, 0.65 to 1.62); outcome had significant unplanned crossover[31]	⊕◯◯◯ VERY LOW
3 cohorts with comparison[32,33] (1 publication reports 2 studies[32]) (380)	Very Serious[o]	Not serious	Not serious	Very serious[i]	No difference for frenotomy vs. control Study 1 (Dixon et al. 2018 study 1[32]): 89/164 [54%] vs.10/22 [46%]; calculated RR, 1.19; 95% CI, 0.74 to 1.93 at median 87 days followup Study 2 (Dixon et al. 2018, study 2[32]): 19/34 [56%] vs. 0/1 [0%]; calculated Peto OR, 8.91; 95% CI, 0.17 to 455.73) at median 118 days Study 3[33]: at 3 months 81/120 [68%] vs. 28/39 [72%]; PR, 0.94, 95% CI, 0.75 to 1.19) and at 6 months (79/120 [66%] vs. 29/39 [7%]; PR, 0.92; 95% CI, 0.72 to 1.16)	⊕◯◯◯ VERY LOW
Changes in breastfeeding						
2 RCTs[8,26] (114)	Serious[p]	Not serious	Serious[q]	Serious[r]	Significant improvement in frenotomy arm vs. control in both RCTs: Study 1[26]: 78% (21/26) vs. 47% (14/30); *p*<0.02; calculated RR; 1.73; 95% CI: 1.13 to 2.65 Study 2[8]: 96% (27/28) vs. 3% (1/29); *p*<0.001; calculated RR: 28.0; 95% CI, 4.07 to 192.12	⊕◯◯◯ VERY LOW
1 cohort study[34] (33)	Very serious[s]	NA—single study	Serious[q]	Serious[r]	Fewer problems in frenotomy arm vs. control participants (13% [n=3/23] vs. 60% [n=6/10]; calculated RR: 0.22; 95% CI, 0.07 to 0.70)[34]	⊕◯◯◯ VERY LOW
1 cohort study[36] (91)	Very serious[s]	NA-single study	Not serious	Very serious[t]	Fewer individuals in frenotomy arm vs. control stopped breastfeeding due to tongue-tie related difficulty or pain (17%, [14/82] vs. 33% [3/9]; calculated RR: 0.51; 95% CI, 0.18 to 1.45)[36]	⊕◯◯◯ VERY LOW
Infant weight gain						
1 RCT[31] (169)	Very serious[k]	NA-single study	Not serious	Serious[u]	No significant difference at 3 months, z-score for weight for age, −1.0 (SD 1.6) vs. −1.1 (SD 1.3); adjusted mean difference in z-score: 0.10 (95% CI, −0.83 to 1.03; *p*=0.83)	⊕◯◯◯ VERY LOW

№ of Studies (№ of participants)	Risk of Bias	Inconsistency	Indirectness	Imprecision	Summary of Findings	CERTAINTY/ Direction of Effect
Infant Breastfeeding Assessment Tool (IBFAT)						
2 RCTs[27,29] (165)	Serious[v]	Serious[w]	Not serious	Serious[x]	Study 1[27]: Significant improvement in score immediately after procedure (calculated mean difference: 3.53; 95% CI, 1.22 to 5.84) Study 2[29]: No change in median score 5 days after procedure: 0 (95% CI, −1.8 to 1) vs. 0 (95% CI, 0 to 1); *p*=0.36	⨁◯◯◯ VERY LOW
Gastroesophageal Symptom Questionnaire for Infants (GSQ-I)						
1 RCT[30] (48)	Very serious[y]	NA—single study	Not serious	Serious[z]	No significant differences (after Bonferroni corrections for multiple comparison) for calculated mean differences and CIs for 10 of 12 domain measures in the GSQ-I;[**]	⨁◯◯◯ VERY LOW

[a] Very serious concerns for bias because of failure to account for baseline differences;[29] randomization issues;[30] high rate of crossover;[29,31] outcome measurement.[29-31] In addition to these potential biases in study conduct, the evidence base may have had errors or inconsistencies in study reporting (see full report).
[b] Serious concerns for consistency because of differences in point estimates.
[c] Confidence spanning the null and inclusive of appreciable benefits and harms for some outcomes. For 1 study,[29] it was unclear whether sample size calculations when reported assumed normal distribution, non-normal distribution reported.
[d] Serious concerns for bias because of failure to account for baseline differences.
[e] Unclear whether sample size calculations assumed normal distribution, non-normal distribution reported. IQR of difference between arms NR, nonsignificant *P* value.
[f] Very serious concerns for bias because of failure to account for baseline differences;[29] randomization issues;[30] high rate of unplanned[31] or early crossover;[29] potential bias in outcome measurement associated with delayed crossover or desired frenotomy.[29-31]
[g] Very serious concerns for bias because of differences at baseline and early crossover.
[h] Confidence spanning the null and inclusive of appreciable benefits.
[i] Very serious concerns for bias in cohort study due to confounding, potential attrition bias, and potential bias in outcome measurement.
[j] Confidence spanning the null and inclusive of appreciable benefits and harms.
[k] Very serious concerns for bias because of high crossover rate and lack of blinding.
[l] Very serious concerns for bias in cohort studies due to lack of controls for confounding, potential attrition bias, and potential bias in outcome measurement.
[m] Confidence spanning the null and inclusive of appreciable benefits and/or harms.
[n] Very serious concerns for bias in RCTs because of failure to account for baseline differences, randomization issues, rate of crossover, and outcome measurement.
[o] Very serious concerns for bias in outcome measurement and because of confounding, outcome reporting, and attrition in cohort study.
[p] High ROB in RCTs due to issues with randomization and outcome assessment.[8,26]
[q] Serious indirectness due to unvalidated measures.
[r] Serious imprecision due to ratio of confidence intervals (≥3 for RR), suggesting that OIS was not met.
[s] Very serious ROB due to confounding and potential bias in outcome measurement.
[t] Very serious imprecision because confidence intervals do not exclude the null and include appreciable benefit and appreciable harm.
[u] Confidence intervals span the null and exceed +1 standard deviation, thus including both no difference and appreciable benefit.
[v] One study[27] was rated at low ROB for conduct of the study but reported standard deviations as standard errors. One study[29] was rated with high ROB because of failure to account for baseline differences.
[w] Inconsistent evidence of benefit arising from multiple possible sources including ROB, timing of outcome measurement, and errors in reporting.
[x] One study reported a large effect size. The sample size of the study (N=58) did not reach 30% to 50% of the optimal information size of 336, based on a baseline value of 8.5 in the control arm and 9.3 in the intervention arm, 1 standard deviation of 3.7, alpha=0.05, and power of 0.80.[27] The second study reported nonoverlapping confidence intervals for median scores suggesting lack of precision.[29]
[y] Very serious concerns for bias because of method of randomization and measurement of outcomes.

[z] Confidence intervals for most domain measures do not exclude the null.
[*] Reported confidence intervals appeared incorrect (negative signs were missing), so we calculated them for each domain.
[**] Mean differences (CIs and *p* values) for (1) vomiting (times): −7.9 (−15.68 to −0.12; 0.057); (2) vomiting (severity): −1.9 (−2.99 to −0.81; 0.001); (3) irritability/fussiness (times): −5.5 (−13.77 to 2.77, 0.19); (4) irritability/fussiness (severity): −1.5 (−2.73 to −0.27; 0.02); (5) refusal to feed (times): −2.3 (−4.62 to 2.48; 0.05); (6) refusal to feed (severity): −1.3 (−2.39 to −0.21; 0.02); (7) choking/gagging (times): −9.8 (−15.93 to −3.67; 0.002); (8) choking/gagging (severity): −0.8 (−1.72 to 0.12; 0.09); (9) arching back (times): −9.7 (−17.03 to −2.37; 0.01); arching back (severity): −1.4 (−2.37 to −0.43; 0.005); (10) episodes of hiccups (times): −4.2 (−8.76 to 0.36; 0.07); and (11) episodes of hiccups (severity): −1.1 (−1.88 to −0.32; 0.006).

Abbreviations: aRR = adjusted risk ratio; BSES = Breastfeeding Self-Efficacy Scale; GSQ-I = Gastroesophageal Symptom Questionnaire for Infants; ITT = intention to treat; IQR = interquartile range; LATCH = Latch, Audible swallowing, Type of nipple, Comfort, Hold; N/n = number; OIS = optimal information size; OR = odds ratio; PR = prevalence ratio; RCT = randomized controlled trial; RR = risk ratio; SD = standard deviation.

ES 3.3 Safety

Fifty-eight studies (57 publications) reported on harms of frenotomy for tongue-tie and/or lip-tie for breastfeeding support. The section is organized by study design, frenotomy method, and oral tie type. Seven of the included studies were RCTs, four were nonrandomized studies of intervention, and 47 were single-arm studies. Because harms may vary by frenotomy method used, we organized the findings by method: scissors, lasers, or unspecified. ***Table ES-2*** presents the number of studies and designs addressing each procedure and indication. Of the included studies, the majority reported harms that were not considered severe or that related to the procedure itself such as minor bleeding, crying, or pain. However, more serious complications were sometimes reported, including damage to other structures in the mouth (e.g., salivary ducts), weight loss and increased feeding difficulties following the procedure, and hospital readmission. Adverse event rates varied significantly across studies owing to inconsistencies in how they were assessed and reported, which prohibited comparisons by frenotomy method.

Table ES-2. Harms of frenotomy for tongue-tie by procedure type

Method of Frenotomy Procedure	Indication	Comparative Studies Reporting Specific Complications by Study Arm	Comparative Studies Reporting "No Complications"	Comparative Studies Reporting Overall Complications, Not by Study Arm	Single-Arm Studies Reporting "No Complications"	Single-Arm Studies Reporting Overall Complications
Scissors	Tongue-tie only	NA	3[8,27,35]	4[26,32,36]	14[38,40,45,48,50,62,64,66,71,72,76,78,80,81]	14[11,37,39,41,43,53,58,59,63,65,70,75,77,79]
	Tongue-tie and/or lip-tie	NA	NA	NA	2[69,79]	3[42,67,82]
	Unspecified tie type	NA	NA	NA	1[73]	NA
Laser	Tongue-tie only	NA	1[30]	NA	NA	2[47,74]
	Tongue-tie and/or other ties (specified)	NA	NA	NA	2[51,55]	3[56,68,74]
Unspecified method	Tongue-tie only	1[31]	NA	3[28,29,31]	2[44,49]	1[46]
	Tongue-tie and/or unspecified ties	NA	NA	NA	1[57]	1[54]
	Unclear/ unspecified tie type	NA	NA	NA	NA	1[60]

Abbreviations: NA = not applicable.

A single high ROB RCT compared rates of complications between frenotomy with unspecified methods for tongue-tie and control arms (1/80 [1.25%] vs. 2/89 [2.2%]).[31] Potential for bias, few events, and small sample sizes resulted in very low certainty of evidence.

Table ES-3. Harms of frenotomy with unspecified methods for tongue-tie: Summary of findings and certainty of evidence ratings

№ of Studies	Risk of Bias	Inconsistency	Indirectness	Imprecision	Summary of Findings	CERTAINTY
Incidence of Complications						
1 RCT[31] (169)	Very serious[a]	NA-single study	Not serious	Extremely serious[b]	No significant difference (1/80 [1.25%] vs. 2/89 [2.2%]; OR: 0.55; 95% CI: 0.05 to 6.19)[31]	⊕◯◯◯ VERY LOW

[a] Serious concerns for bias because of high crossover rate and lack of blinding.
[b] Extremely wide confidence intervals including appreciable benefit and appreciable harm and suggest very different inferences.

Abbreviations: CI=Confidence interval; OR = odds ratio; RCT = randomized controlled trial.

ES 3.4 Cost Effectiveness

No studies that met criteria for the cost-effectiveness question were identified.

ES 3.5 Clinical Practice Guidelines

Clinical practice guidelines related to frenotomy and frenectomy from professional organizations are listed in ***Table ES-4***. Statements related to frenotomy were available from the American Academy of Otolaryngology—Head and Neck Surgery, the American Academy of Pediatric Dentistry, American Academy of Pediatrics, The Academy of Breastfeeding Medicine, International Board of Lactation Consultant Examiners, the Canadian Paediatric Society, and the Canadian Agency for Drugs and Technologies in Health. None explicitly relied on systematic reviews. We attempted to appraise each publication using the Appraisal of Guidelines for Research & Evaluation II (AGREE II) instrument, but as this instrument focuses on evaluating the process through which guidelines are developed, it had limited applicability for the included documents. The tool also does not assess how well the evidence included in each guideline is evaluated, interpreted, or whether the conclusions were consistent with the evidence. Due to these concerns a quality rating of NA is reported for all included statements.

Table ES-4.Clinical practice statements relating to frenotomy and frenectomy with breastfeeding support

Title/Organization Guideline Quality[a]	Year Published	Excerpts of Findings	Rating/Quality of Evidence Narrative Assessment Used
American Academy of Otolaryngology—Head and Neck Surgery Foundation[16] Quality rating: NA	2020	**For frenotomy:** A survey of expert pediatric otolaryngologists agreed that frenotomy in infants with ankyloglossia can lead to an improvement in breastfeeding, not all infants with ankyloglossia need a frenotomy, and there are more common conditions which may impede breastfeeding. The Academy recommends further study to refine evidence.	Based on 2 systematic reviews. Quality of evidence assessment not performed.
American Academy of Pediatric Dentistry[83] Quality rating: NA	2022	**For surgical interventions on the frenulum:** Recognizes that difficulties with breastfeeding may have another cause and not all infants with ankyloglossia require surgical intervention. Recommends a team-based approach to treatment planning. The Academy supports further research in the causative association between ankyloglossia and difficulties in breastfeeding.	Based on a nonsystematic review of the literature. Quality of evidence assessment not performed.
American Academy of Pediatrics[84] Quality rating: NA	2024	**For frenotomy:** It is unclear if release of a tight lingual frenulum in neonates improves breastfeeding. Because symptoms of ankyloglossia overlap those of other breastfeeding difficulties, a team partnership is necessary. Frenotomy may decrease maternal nipple pain. Further research is necessary.	Based on a nonsystematic review of the literature. Quality of evidence assessment not performed.

Title/Organization Guideline Quality[a]	Year Published	Excerpts of Findings	Rating/Quality of Evidence Narrative Assessment Used
The Academy of Breastfeeding Medicine[85] Quality rating: NA	2021	**For surgical tongue-tie release:** If there is the presence of a restrictive sublingual frenulum, frenotomy can be an effective way to increase maternal comfort and milk transfer and may prevent premature breastfeeding cessation. The Academy urges for more research on clear definitions of "tongue-tie," optimal surgical methods, and long-term outcomes.	Based on a nonsystematic review of the literature. Quality of evidence assessment not performed.
International Board of Lactation Consultant Examiners[86] Quality rating: NA	2017	Members of the International Board of Lactation Consultant Examiners should not diagnose tongue-tie but may refer parents to a clinician who can diagnose.	Overview of International Board of Lactation Consultant Examiners scope of practice, clinical competencies, code of conduct, and advisory opinions. Quality of evidence assessment not performed.

ES 4. Discussion

ES 4.1 Summary of the Evidence

As depicted in ***Figure ES-2***, the level of certainty across all outcomes was low or very low.

Figure ES-2. Evidence map—Frenotomy and frenectomy with breastfeeding support

Outcome category	Specific outcome	Favors frenotomy	No difference	Favors control	Cannot determine a direction of effect
Maternal outcomes	Breastfeeding pain				6 RCTs, N=452
	Breastfeeding effectiveness				1 RCT, N=105
	Breastfeeding self-efficacy	3 RCTs, N=312			
	Any breastfeeding at ≤2 months				1 RCTs, N=105 1 NRSI, N=159
	Any breastfeeding at >2 months		1 RCT, N=163		4 NRSIs, N=471
	Exclusive breastfeeding at ≤2 months		1 NRSI, N=159		2 RCTs, N=265
	Exclusive breastfeeding at >2 months				1 RCT, N=163 3 NRSIs, N=380
	Improvement in breastfeeding				2 RCTs, N=114
	Breastfeeding problems				1 NRSI, N=33
	Cessation of breastfeeding				1 NRSI, N=91

Outcome category	Specific outcome	Favors frenotomy	No difference	Favors control	Cannot determine a direction of effect
Infant outcomes	Infant weight gain				1 RCT, N=163
	Infant breastfeeding assessment				2 RCTs, N=165
	Gastroesophageal symptoms				1 RCT, N=48
Safety	Complications				1 RCT, N=163

[a] We did not grade the safety evidence from 47 single-arm studies, 6 RCTs, and 4 NRSIs.

Abbreviations: N = number; NRSI = nonrandomized study of interventions; RCT = randomized controlled trial.

Table legend

	High certainty of evidence
	Moderate certainty of evidence
	Low certainty of evidence
	Very low certainty of evidence

The inconclusive evidence for the effects of frenotomy on breastfeeding outcomes arises from study design limitations. Ethical and pragmatic considerations around the widespread belief among participants and some clinicians that frenotomy will be helpful ("lack of equipoise")[31] meant that most RCTs offered frenotomy to the control arm, either immediately or after a delay. The only RCT without planned frenotomy in the control arm reported very high rates of unplanned crossover;[31] even trials with planned delayed crossover (at 5 days) reported early crossover.[29] The expectation of frenotomy in the control groups may have influenced patient-reported outcomes. Sham RCTs[26-28] attempted to address this issue by blinding participants in both arms during the procedure but could only provide blinded outcomes immediately after the procedure. Authors of 1 study noted that they "do not foresee a way to prevent mothers from looking in their infants' mouths."[27] Additionally, blinding was not always successful during the sham procedure. All these considerations limited the utility of trial evidence. Nonrandomized retrospective cohort studies did not have these specific design issues but the potential for confounding also seriously limited their utility. Beyond these design considerations, there is also an unclear understanding of the typical time to achieve "good" breastfeeding, and thus the appropriate time to consider surgical or other intervention for breastfeeding problems may vary. Other reasons for lack of certainty are related to small, underpowered studies and the use of unvalidated measures. No evidence was identified that examined the effectiveness or comparative effectiveness of frenotomy for tongue-tie with concomitant lip-tie or lip-tie alone.

Various harms were reported for frenotomy using scissors, lasers, and unspecified methods across oral tie types. The level of severity for reported harms ranged from studies reporting no harms or adverse events, minor harms including bleeding or crying, and more serious harms including accidental cut to the tongue and salivary duct damage. Only 1 study provided comparative data on harms of frenotomy for tongue-tie and reported no differences in the incidence of complications between frenotomy and breastfeeding support (very low certainty of evidence). Other included comparative safety studies reported no harms from frenotomy or only overall study complication rates (i.e., across all study groups). All other reported safety data was

from single-arm studies, which were not designed to provide causal inference or provide clear associations between frenotomy and harms.

We did not identify evidence that examined the cost-effectiveness or cost-utility of frenotomy for tongue-tie with or without lip-tie or lip-tie alone.

ES 4.2 Limitations of the Evidence Base

Most efficacy studies were small; the median sample size was 58. Some studies were unable to maintain blinding of mothers. Both planned and unplanned crossover occurred, limiting measurement of long-term outcomes. Additionally, it was difficult to determine the level of exposure to other interventions that could impact outcomes (i.e., interaction with lactation consultants or other breastfeeding assistance).

The majority of safety studies were single-arm studies or comparative studies that only provided overall harms data for study samples. Safety studies also lacked detailed measurement information and lacked consistency in how harms and complications were classified.

ES 4.3 Payer Coverage

Payer coverage for frenotomy and frenectomy with breastfeeding support is presented in ***Tables ES-5***. Additional details are available in the Full Technical report.

Table ES-5.Overview of payer coverage policies for frenotomy and frenectomy with breastfeeding support

Condition	Washington Apple Health (Medicaid)[87]	Cigna	Kaiser Permanente	Premera Blue Cross[88]	Regence BlueShield[89]	UnitedHealth[90]
Labial frenotomy/ frenulotomy	—	—	—	—	—	—
Lingual frenotomy /frenulotomy	—	—	—	—	—	—
Labial frenoplasty/ frenuloplasty	✓	—	—	—	✓	✓
Lingual frenoplasty/ frenuloplasty	✓	—	—	—	✓	—
Labial frenectomy/ frenulectomy	✓	—	—	✓	✓	✓
Lingual frenectomy/ frenulectomy	✓	—	—	✓	✓	—

Notes: ✓ = covered; X = not covered; — = no policy identified.

ES 4.4 Limitations of this HTA

We limited the scope to English-language publications conducted only in countries rated as "very high" on the 2023/2024 UN Human Development Index. Also, the literature search was limited to 2 databases and studies published from database inception through August 2024. We did not seek unpublished data and did not use data presented only in conference abstracts. We did not consider efficacy outcomes from uncontrolled studies and did not use GRADE to evaluate the body of evidence consisting of uncontrolled studies for safety outcomes.

ES 4.5 Ongoing Research and Future Research Needs

We identified 1 relevant clinical trial registered in clinicaltrials.gov.[91] The crossover RCT consisted of Group 1 receiving sham frenotomy followed by a lingual frenotomy, and Group 2 receiving a lingual frenotomy followed by a sham frenotomy. Newborns who continued having feeding difficulties received a third intervention. May 2018 was listed as the trial completion date, but no published results are currently available. The trial record was last updated June 12, 2019.

Future efficacy, safety, and cost-effectiveness studies should seek to address the limitations of the current evidence base. Due to the ethical concerns of randomizing mother and infant dyads to intervention versus control, the difficulty of blinding, and lack of equipoise because most mothers likely believe that frenotomy will be helpful as evidenced by high crossover rates, future studies could focus on comparing frenotomy methods (i.e., scissors vs. lasers), timing (e.g., frenotomy performed soon after birth vs. later), including assessing for improvement in breastfeeding over time absent frenotomy, and whether the benefits of frenotomy vary based on contextual factors such as the availability of intensive and comprehensive breastfeeding support. Future studies should attempt to report on longer-term outcomes and should consider using large health care system data in an effort to include larger sample sizes. For efficacy, future studies should look to explore the impact of frenotomy on the outcomes of nipple excoriations, nipple infections (mastitis), aerophagia, swallowing function, failure to thrive, milk transfer, low milk supply, and other feeding issues that appear to be absent in the literature based on the current review's inclusion criteria. For harms specifically, future studies should use more intentional approaches for collecting and reporting harms data, including using predetermined and well-defined measures and collecting data from various sources (e.g., parents, provider, medical records).

ES 5. Conclusion

We identified methodologically limited evidence for evaluating the efficacy and safety of frenotomy for breastfeeding support in infants up to 1 year of age with tongue-tie and/or lip-tie and no evidence reporting on cost-effectiveness.

Full Technical Report

1. Background

This health technology assessment (HTA) reviews the efficacy, safety, and cost-effectiveness of frenotomy and frenectomy with breastfeeding support to assist the State of Washington's Health Technology Clinical Committee in determining coverage for frenotomy and frenectomy to treat tongue-tie and/or lip-tie in infants.

1.1 Condition Description

Ankyloglossia, colloquially known as "tongue-tie," is a condition that limits the movement of the tongue[1] and can cause difficulty with breastfeeding in the newborn.[2] "Lip-tie" refers to a similar condition in which the maxillary labial frenum connecting the upper lip to the maxillary alveolar ridge restricts movement and similarly causes difficulty with breastfeeding.[3]

Estimates of ankyloglossia in infants vary from less than 1% to about 11%, with prevalence more common among males than females.[4-6] Reasons for the wide variance in prevalence arise from unclear diagnostic methods, which may include visual inspection of the oral anatomy, assessment of functional impairment and decreased mobility, and the effect on mothers during breastfeeding (such as nipple pain).[6] Ankyloglossia may be most commonly anterior, that is, where the frenum attaches near the tip of the tongue and is visible, or less commonly, posterior, where the frenulum is attached further back on the tongue and may be harder to see.[58] Of note, there is no consensus regarding the definition of "posterior ankyloglossia," including whether this represents a distinct clinical entity.[16,84] Categories of severity have been proposed that rely on free tongue length[92] and additional anatomical features (thickness, notching),[93] but the relationship between these categories and breastfeeding difficulty have not been established.[94] As a result, additional functional assessments of breastfeeding such as the Latch, Audible swallowing, Type of nipple, Comfort, Hold (LATCH) index, Infant Breastfeeding Assessment Tool (IBFAT), or Frenotomy Decision Rule for Breastfeeding Infants may be needed.[94] The absence of validated diagnostic criteria creates uncertainty around the threshold for management.[6]

1.2 Disease Burden

Diagnosis of ankyloglossia in infants and rates of frenotomy have increased sharply over the past 2 decades. Diagnoses of ankyloglossia in the United States increased from 3,377 in 2004 to 13,200 in 2019, and lingual frenotomy increased from 1,483 in 2004 to 6,213 in 2019.[7] Reasons suggested for the increase include efforts to supporting breastfeeding, increased awareness of ankyloglossia and its role in breastfeeding, and the increased role of pediatricians as proceduralists.[7,95]

Risk factors associated with ankyloglossia and receiving a frenotomy include male sex, private insurance, and higher income. The extent to which the increase in procedures represents increased incidence of the condition, improved diagnoses, or overdiagnosis is unclear.[84]

Outcomes potentially associated with untreated ankyloglossia include breastfeeding difficulties that may result in restricted weight gain in the infant,[8-11] speech difficulties and problems with dentition,[12,13] maternal pain, reduced milk supply, or incomplete breast emptying that may result in infections.[14,15]

1.3 Technology Description

Frenectomy, frenotomy (also called frenulotomy), and frenuloplasty are sometimes used interchangeably but refer to different procedures. Lingual frenotomy—conducted with a laser, electrocautery, scalpel, or surgical scissors[16]—refers to surgical procedures to release the tongue-tie; lingual frenectomy refers to the removal of the lingual frenulum; and frenuloplasty (also called z-plasty) refers to plastic surgery of the tongue, often used during repeat procedures or clinical scenarios requiring a complex approach.[16,17] Labial frenectomies (and related procedures) for lip-tie refer to procedures to release the frenum that connects the lips to the gums. Going forward we use the term "frenotomy" to refer to all such procedures.

Release procedures have long been offered by different specialties with different interests and expertise.[96] In modern practice, the procedure is offered by pediatricians, otolaryngologists, oral and maxillofacial surgeons, dentists, and lactation consultants.[97] Clinical training and specialty, access to equipment,[7] and consultation with other providers[98] may influence the type of procedure. Each procedure has a different billing code, required documentation, and reimbursement rate.

Although rates of surgical complications after the procedure are difficult to ascertain, reported complications include need for a repeat procedure, bleeding that may lead to hypovolemic shock, swelling, scarring, pain, airway obstruction that may lead to sleep apnea, infection and Ludwig's angina, damage to the tongue or salivary ducts, or oral aversion that interferes with feeding.[97,99-102] Some types of procedures may be more closely associated with specific complications than others (e.g., bleeding with scalpel or scissors when compared with laser, and oral aversion with laser when compared with scalpel or scissors).[97]

The range of potential clinical providers and attendant variations in clinical practice, thresholds for action, and financial incentives add further uncertainty in management of tongue-tie and lip-tie. Recent media coverage has also drawn attention to these uncertainties.[103]

1.4 Regulatory Status

The U.S. Food and Drug Administration (FDA) does not regulate the performance of these release procedures. However, tools such as lasers and electrocauterizers used in the procedure may have FDA clearance or premarket approvals that mention frenotomies.[18]

1.5 Policy Context

The State of Washington Health Care Authority selected frenotomy and frenectomy with breastfeeding support for an HTA because of high concerns for efficacy and medium concerns for safety and cost.

2. Methods

This section describes the methods we used to conduct this HTA.

2.1 Research Questions and Analytic Framework

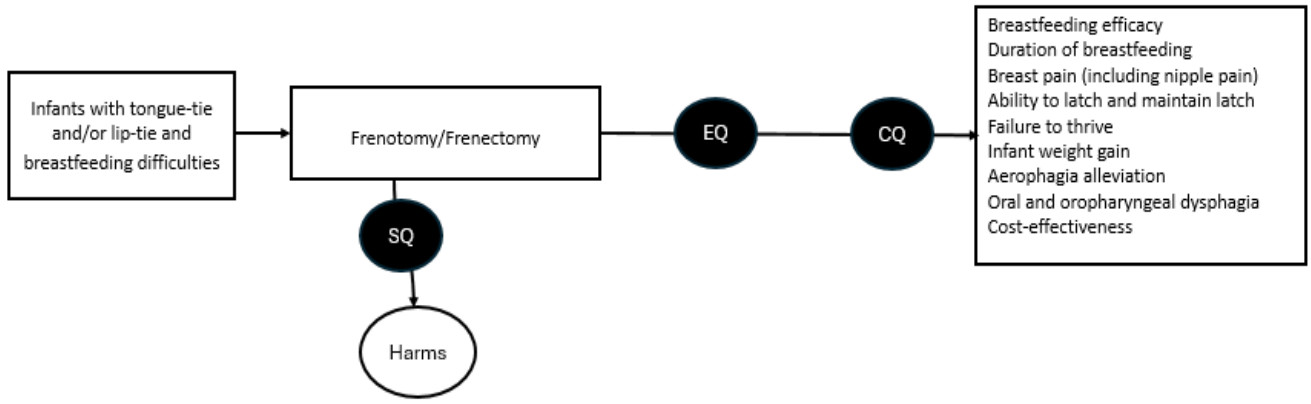
We developed the following research questions and analytic framework (***Figure 1***) to guide the systematic evidence review of primary research studies:

Efficacy Question (EQ). What is the effectiveness and comparative effectiveness of frenotomy or frenectomy for tongue-tie and/or lip-tie on breastfeeding outcomes?

Safety Question (SQ). What are the harms of frenotomy or frenectomy for tongue-tie and/or lip-tie as a support for breastfeeding?

Cost Question (CQ). What is the cost-effectiveness of frenotomy or frenectomy for tongue-tie and/or lip-tie for breastfeeding support?

Figure 1. Analytic framework for HTA on frenotomy and frenectomy with breastfeeding support



Abbreviations: CQ = cost question; EQ = efficacy question; SQ = safety question

The State of Washington HTA Program posted a draft of these research questions with study selection criteria for public comment from September 27, 2024, to October 10, 2024. The final key questions and response to public comments on the draft key questions are available at the Program's website. Two independent, external peer reviewers will review a draft version of this evidence report, and it will also be posted for public comment from February 28, 2025, until March 31, 2025. Feedback from peer reviewers and from public comments will be incorporated

into the Final Evidence Report; responses to public and peer review comments will be summarized in a separate document and will also be available at the Program's website.

2.2 Data Sources and Searches

We searched MEDLINE (via PubMed) and the Cochrane Library for relevant articles and clinical trials. Search dates ranged from database inception to August 30, 2024. In addition, we reviewed reference lists of relevant studies, systematic reviews, and practice guidelines to identify any relevant research studies not identified through the electronic search. The detailed search strategy is in ***Appendix A.***

In brief, we used medical subject headings (MeSH terms) and text words associated with "mouth abnormalities", "tongue diseases/congenital", "tongue/abnormalities", "lingual frenum", "lip diseases/congenital", "lip/abnormalities", "labial frenum", "ankyloglossia", and "oral surgical procedures".

2.3 Study Selection

Table 1. Population, intervention, comparator, outcome, timing, setting, and other study selection criteria for HTA on frenotomy and frenectomy with breastfeeding support***1*** summarizes the study selection criteria related to the population, intervention, comparator, outcomes, time period, study designs, and setting that defined the scope of this HTA; these are further described following the table. We screened titles/abstracts and full-text articles based on these study selection criteria. Two review team members independently screened all titles/abstracts and full-text articles; discrepancies in the study selection at the full-text level were resolved by consensus among the team or by a senior investigator. Reasons for exclusion are listed in ***Appendix B***.

Table 1. Population, intervention, comparator, outcome, timing, setting, and other study selection criteria for HTA on frenotomy and frenectomy with breastfeeding support

Domain	Included	Excluded
Population	Breastfeeding infants up to 1 year of age with tongue-tie and/or lip-tie	Infants with physical/anatomic comorbidities, such as hypotonia Infants with Pierre Robin syndrome or sequence, Down syndrome, or craniofacial or airway abnormalities (i.e., cleft palate) Infants born at less than <37 weeks of gestation
Intervention	Frenotomy, frenectomy, frenulotomy, frenulopasty, or z-plasty to improve breastfeeding using all methods (i.e., scissors, lasers)	Frenotomy, frenectomy, frenulotomy, or Z-plasty done for indications other than breastfeeding support
Comparator	• EQ: All comparators including other surgical approaches, sham surgery, nonsurgical interventions (i.e., lactation intervention, speech therapy, physical/occupational therapy, oral motor therapy, and stretching exercises/therapy), complementary and alternative medicine therapies (e.g., craniosacral therapy), observation only • SQ: No comparator necessary • CQ: Any comparator	• EQ: No comparator group • SQ: NA • CQ: No comparator group

Domain	Included	Excluded
Outcomes	• EQ: Breastfeeding, including latch, nipple pain, nipple excoriations, nipple infections (mastitis), weight gain, aerophagia, swallowing function, failure to thrive, milk transfer, low milk supply, breastfeeding cessation/duration of breastfeeding, and other feeding issues • SQ: Any harms, including excessive bleeding, airway obstruction, pain, transient poor feeding secondary to discomfort, dysphagia, complications related to dysphagia such as aspiration pneumonia, surgical site infection, nerve damage, salivary gland damage, ranulae, scarring, soft tissue damage, oral aversion, readherence of tongue- or lip-tie, need for further surgery/revision, ED visits, hospitalizations, extension of current hospitalization • CQ: cost-effectiveness or cost-utility	Outcomes not listed as eligible Cost-effectiveness based on cost inputs from countries other than the U.S.
Timing	• EQ: Outcomes measured after intervention/comparator through 12 months of age • SQ: No time limitation • CQ: No time limitations	Outcomes measured after 12 months of age
Setting	Inpatient or outpatient pediatric care, operating room, newborn nursery or NICU, ENT clinic, primary care, outpatient, dental office, or breastfeeding medicine clinics in countries categorized as "very high" on the 2023/2024 United Nations Human Development Index[19]	Studies conducted in countries not categorized as "very high" on the 2023/2024 United Nations Human Development Index[19]
Study Design and Risk of Bias Rating	• EQ: RCTs, nonrandomized controlled trials, prospective and retrospective cohort studies, cross over studies, and case-control studies • SQ: Same as for EQ plus case series • CQ: Cost-effectiveness or cost-utility studies	• EQ: Case reports, case series, qualitative studies • SQ: Qualitative studies, and all study designs not already specified • CQ: Studies that use non-U.S.-based cost inputs • EQ, SQ, and CQ: Relevant SRs will be excluded but will be hand searched to identify potentially eligible primary studies
Language	English	Non-English
Publication Type	Original research	Editorial, commentaries, narrative reviews, or letters

Abbreviations: CQ =cost question; ED = emergency department; ENT = ear, nose and throat; EQ = efficacy question; NA = not applicable; NICU = neonatal intensive care unit; RCT = randomized controlled trial; SQ = safety question; SR = systematic review; U.S. = United States.

2.3.1 Population

Studies were selected if they enrolled full-term newborns with diagnosed tongue-tie and/or lip-tie who were not diagnosed with another condition such as Pierre Robin syndrome, Down syndrome, or craniofacial abnormalities that could interfere with breastfeeding. Newborns were required to be breastfed, although studies that also included non-breastfed infants were considered if data relevant to breastfeeding success could be abstracted.

2.3.2 Intervention and Comparator

We considered frenotomy, frenectomy, frenulotomy, frenuloplasty, or Z-plasty as eligible interventions. All methods of intervention (e.g., scissors, laser, scalpel, electrocautery) were included. For the EQ, all comparators, including sham procedure, wait list control, nonsurgical interventions, complementary and alternative medicine, and observation only were included. For the SQ and the CQ, no comparators were necessary.

2.3.3 Outcomes

For the EQ, breastfeeding outcomes were eligible for selection, including latch, nipple pain, nipple excoriations, nipple infections (mastitis), weight gain, aerophagia, swallowing function, failure to thrive, milk transfer, low milk supply, breastfeeding cessation/duration of breastfeeding, and other feeding issues. Outcomes eligible for selection could be relevant to the lactating person or the infant. For the SQ, any harms reported after the procedure, including excessive bleeding, airway obstruction, pain, transient poor feeding secondary to discomfort, dysphagia, complications related to dysphagia such as aspiration pneumonia, surgical site infection, nerve damage, salivary gland damage, ranulae, scarring, soft tissue damage, oral aversion, readherence of tongue- or lip-tie, need for further surgery/revision, emergency department (ED) visits, hospitalizations, or extension of current hospitalization, were eligible. For the CQ, studies with cost-effectiveness or cost-utility outcomes were eligible for inclusion.

2.3.4 Settings

Eligible settings were the same for all research questions. Inpatient or outpatient pediatric care facilities, operating room, newborn nursery or neonatal intensive care unit (NICU), ear, nose and throat (ENT) clinic, primary care outpatient, dental office, or breastfeeding medicine clinics were eligible settings. Only studies conducted in countries considered "very high" on the 2023/2024 United Nations (UN) Human Development Index[19] were eligible for the EQ and the SQ. Only studies using cost inputs from the United States were eligible for the CQ.

2.3.5 Study Design

Studies to address the EQ and SQ were required to be randomized controlled trials (RCTs), nonrandomized controlled trials, prospective and retrospective cohort studies, crossover studies, or case-control studies. We also included case series for the SQ. For the CQ, eligible study designs were cost-effectiveness or cost-utility studies.

2.3.6 Time Period

For the EQ, outcomes measured before infants reached 12 months of age were eligible. There were no time limits on SQ or CQ outcomes. There were no restrictions on publication dates in the search methods.

2.4 What Is Excluded from This HTA

This review did not include studies published in languages other than English or conducted in countries that are not rated as "very high" on the 2023/2024 UN Human Development Index.[19] This review did not include studies on tongue-tie release procedures done for reasons outside of infant breastfeeding, including speech issues or sleep apnea issues, which may appear in older

children and adults. This review also does not include studies conducted among infants with major comorbidities or other abnormalities, in particular craniofacial abnormalities such as Pierre Robin syndrome, Down syndrome, or cleft lip/palate, or infants born at less than 37 weeks' gestation. This review does not include studies with no comparison group (including pre-post studies) for the EQ, although these study types were considered for the SQ and the CQ.

2.5 Data Abstraction and Risk of Bias Assessment

One team member abstracted relevant data from each eligible study using a structured form and a second team member checked these data for accuracy.

We used the Cochrane Risk of Bias (RoB 2) tool[20] to assess the risk of bias (ROB) of study conduct for included RCTs. Domains assessed with this tool include bias arising from the randomization process, bias due to deviations from intended interventions, bias due to missing outcome data, bias in measurement of the outcome, and bias in selection of the reported result. ROB was assessed as "high," "moderate," or "low" at the study level, unless different outcomes within a single study required outcome-level ROB ratings.

We used the ROBINS-I tool[21] to assess the ROB of study conduct for nonrandomized comparative studies (i.e., cohort studies with comparisons). Domains assessed with this tool include bias due to confounding, selection of participants in the study, selection, classification, deviations from intended interventions, missing data, measurements of outcomes, and selection of reported result. ROB was assessed as "low," "moderate," "serious," or "critical."

To assess the quality of case reports and single-arm studies evaluated for the SQ, we used a modified version of the tool developed by Murad et al.,[22] which reviews the representativeness of the sample, adequacy of ascertainment of the exposure and outcome, whether design features support causal inference, and whether reporting permits replication or generalizable inference.

A senior investigator conducted initial ROB assessments for each study and a second senior investigator confirmed the assessment. We resolved disagreements by discussion. Individual study ROB assessments for included studies are included in ***Appendix D***.

2.6 Data Synthesis and Quality of Evidence Rating

We qualitatively synthesized study characteristics and results in tabular and narrative formats. We graded the certainty of evidence (COE) for each comparison using the Grading of Recommendations Assessment, Development, and Evaluation (GRADE) approach,[24] which requires ratings for ROB, consistency, directness, and precision. We based ROB ratings on RoB 2 and ROBINS-I ratings, as described above. Additionally, when we noted errors in reporting we attempted to contact study authors and downgraded the body of evidence for those outcomes when corrected data was not obtained.

To assess the consistency domain, we evaluated both the consistency in the direction and magnitude of effect for a statistically significant difference between intervention and comparator groups, or consistently supported no statistically significant difference.

To assess directness, we considered whether outcome measures were validated. Unvalidated measures were downgraded.

To assess precision, we used a minimally contextualized approach to grading precision, where the target of certainty was the null effect.[25] Outcomes with confidence intervals that excluded the null were not downgraded for precision unless the effect sizes were implausibly large (e.g., relative risk reduction or relative risk increase exceeding 30%). In such instances, we evaluated whether the ratio of the confidence interval exceeded 3 for relative risks and 2.5 for odds ratios for categorical outcomes, or if the sample size was smaller than 30% to 50% of the optimal information size for continuous outcomes. In such instances, we downgraded outcomes for precision by 2 levels. When outcomes had confidence intervals that included the null, we considered whether the confidence intervals included both appreciable benefit or appreciable harm (25% relative increase or reduction for categorical outcomes, 1 standard deviation for continuous outcomes). If this was the case we downgraded for precision by at least 2 levels. When confidence intervals were not available or calculable, we relied on reported information such as *p* values and sample sizes.

Two team members independently graded each body of evidence, and we resolved discrepancies through discussion. With GRADE, the COE can be graded as "very low," "low," "moderate," or "high." ***Table 2*** defines these levels.[104] We graded bodies of evidence from RCTs separately from other study designs. Bodies of evidence begin with a "high" certainty rating and are downgraded based on domains relating to study limitations (i.e., ROB), inconsistency, imprecision, indirectness, and other considerations, such as publication bias.

Table 2. Certainty of evidence grades and definitions (adapted from Berkman et al.)[104]

Certainty Grade	Definition
High	**We are very certain that the estimate of effect lies close to the true effect for this outcome.** The body of evidence has few or no deficiencies. We believe that the findings are stable, that is, another study would not change the conclusions.
Moderate	**We are moderately certain that the estimate of effect lies close to the true effect for this outcome.** The body of evidence has some deficiencies. We believe that the findings are likely to be stable, but some doubt remains.
Low	**We have limited certainty that the estimate of effect lies close to the true effect for this outcome.** The body of evidence has major or numerous deficiencies (or both). We believe that additional evidence is needed before concluding either that the findings are stable or that the estimate of effect is close to the true effect.
Very Low	**We have very limited certainty that the estimate of effect lies close to the true effect for this outcome.** The body of evidence has numerous major deficiencies. We believe that substantial additional evidence is needed before concluding either that the findings are stable or that the estimate of effect is close to the true effect.

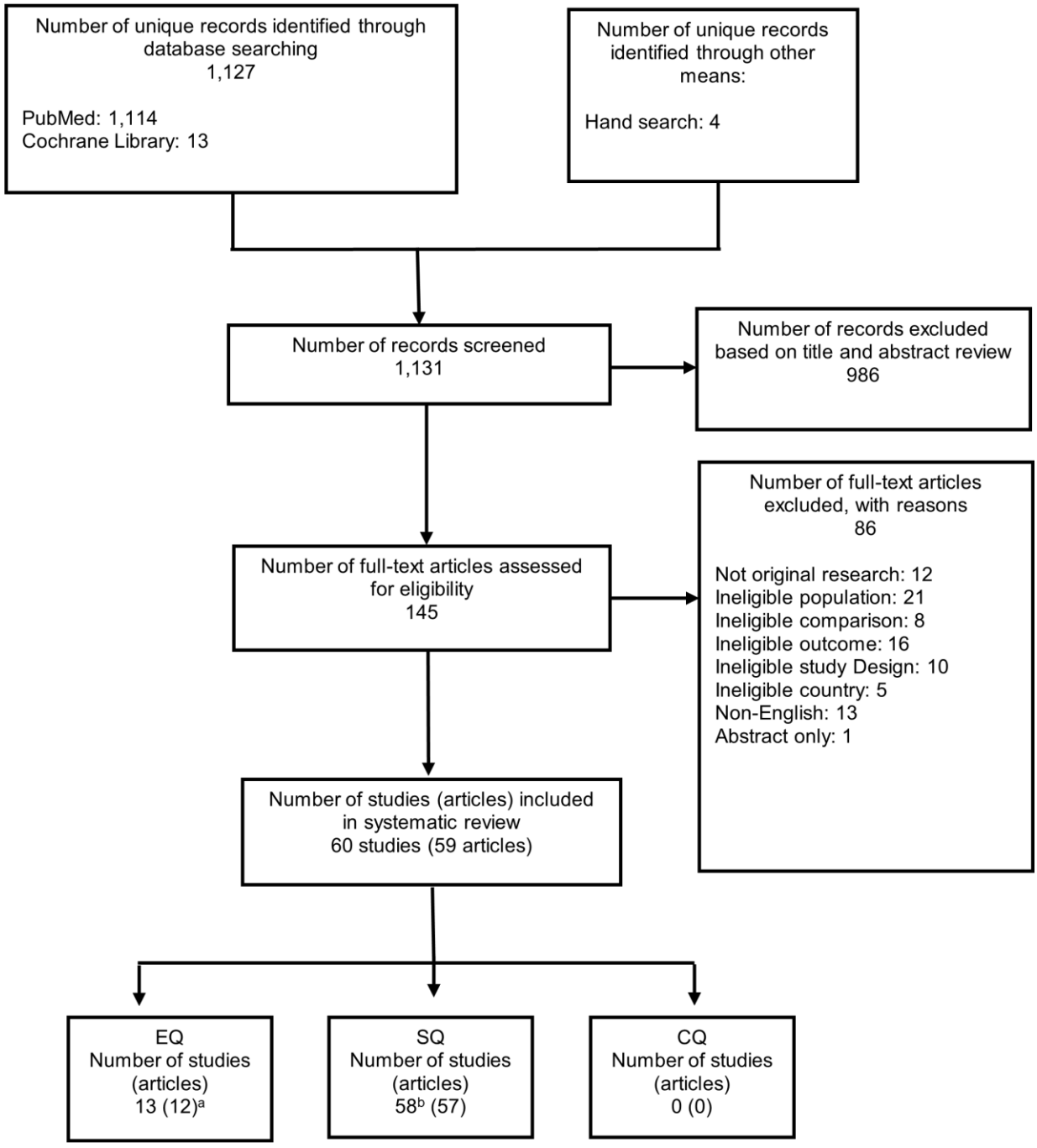
3. Results

3.1 Literature Search

Figure 2 depicts the study flow diagram summarizing our study identification and screening process. Of 1,131 unique citations, we included 60 studies reported in 59 articles representing 7

RCTs,[8,26-31] 6 cohort studies in 5 articles with comparison groups,[32-36] and 47 single-arm studies.[11,37-82] Thirteen studies[8,26-36] were relevant to the EQ, and 58 studies[8,11,26-32,35-82] were relevant to the SQ. No studies were identified for the CQ.

Figure 2. Study flow diagram for HTA on frenotomy and frenectomy with breastfeeding support



[a] Eleven of the EQ studies also contained data for the SQ.
[b] One article reported on 2 studies.

Abbreviations: CQ = cost question; EQ = efficacy question; RCT = randomized controlled trial; SQ = safety question.

Comparison groups included breastfeeding support,[8,28,30,31,33-35] immediate or delayed frenotomy,[8,28-30] sham,[26-28] and no frenotomy.[32-36] Six of the 7 RCTs offered frenotomy to the control group, either immediately (2 studies[26,28]) or within 2 weeks (4 studies[8,27,29,30]). The 2 RCTs that planned immediate crossovers had 100% crossover rates. For those offering delayed crossover, 67% to 96% of the control arm chose to have frenotomies. For these RCTs, we did not analyze outcomes reported after the planned crossover.

One trial (FROSTTIE) did not plan crossovers; the comparator was breastfeeding support only.[31] By the end of the trial, 73% of the control arm had received a frenotomy. We included all relevant outcomes and rated them as high ROB.

Of the 13 comparative studies, we assessed 3 as low ROB for some or all outcomes,[26-28] 0 as moderate ROB, and 10 as high ROB.[8,29-36] One RCT[26] had low ROB for breastfeeding pain and high ROB[27] for breastfeeding improvement.

Specific concerns in high ROB RCTs included flaws in randomization (such as unblinded allocation and differences at baseline), departures from intended interventions (unplanned crossover), bias in measurement (potential for bias in self-reported outcomes when participants were offered delayed frenotomy and were not blinded), and attrition. Specific concerns for high ROB in nonrandomized studies included confounding and attrition. Three sham trials[26-28] attempted to address bias in outcome measurement by blinding the mother to the procedure, but 2 of 3 reported at least some failure of blinding (in 1 of 26 participants[28] and 3 of 60 participants[26]). For the 47 single-arm studies that we included for harms data, only 1 ruled out alternative explanations for the outcome among those that appeared to have included representative populations and adequately measured exposures.[70] The majority of the single-arm studies (n=38) did not ascertain the outcome adequately or follow up patients long enough for harms to occur (n=38). As a result, the overall quality of these studies does not support a robust estimate of potential harms associated with frenotomy.

Key characteristics of these studies can be found in ***Table 3***, with additional study-level details summarized in ***Appendix C***.

Across the included studies, the age of infants at the time of the procedure ranged from a few hours after birth up to 52 weeks. More than half of the participants in included studies were male and had a tongue-tie, 100% for EQ studies and 67% of SQ studies. Twenty-two percent of SQ studies included participants with oral tie types other than tongue-tie only or did not describe the oral tie type that was being corrected (10%). Race and ethnicity of participants was not reported for the majority of included studies. Of those that did report race and ethnicity (23% of EQ and 24% of SQ studies), samples were majority White/European participants. The majority of studies (77% of EQ and 71% of SQ**)** were conducted in geographical settings outside of the United States, including Austria,[70] Australia,[11,37,38,77] Canada,[75,76] Denmark,[73] Germany,[34] Ireland,[65] Israel,[28,48] New Zealand,[32,42,54,59,60] Spain,[41,50] Thailand,[80] Turkey,[66] and the United Kingdom.[8,26,29,31,35,39,43-46,49,53,63,64,71,72,81] Study interventions included frenotomy with scissors, lasers, or unspecified methods. Studies reported that interventions were conducted by a range of

clinicians including pediatricians, ENTs, lactation consultants, midwives, dentists, oral surgeons, pediatric otolaryngologists, otolaryngology physician assistants, surgeons, and others.

Table 3. Key study characteristics

Study Characteristics	Subcharacteristics	EQ Number of Studies (%)	SQ Number of Studies (%)
Population characteristics			
Mean age at procedure	≤1 week	2 (15)	7 (12)
	≤2 weeks	1 (8)	6 (10)
	≤1 months	3 (23)	13 (22)
	≤2 months	2 (15)	14 (24)
	≤6 months	2 (15)	3 (5)
	NR[a]	3 (23)	15 (26)
Gender	Majority male (>50%)	10 (77)	41 (71)
	Majority female (>50%)	0 (0)	1 (2)
	NR	3 (23)	16 (28)
Race or ethnicity	Majority White/European (>50%)	3 (23)	14 (24)
	Majority non-White (>50%)	0 (0)	0 (0)
	Not reported	10 (77)	44 (76)
Oral tie type	Tongue-tie only	13 (100)	39 (67)
	Tongue-tie and/or other tie types	0 (0)	13 (22)
	NR[b]	0 (0)	6 (10)
Intervention characteristics			
Frenotomy method	Scissors	7 (54)	39 (67)
	Laser	1 (8)	8 (14)
	Unspecified	5 (38)	11 (19)
Provider	Various providers[c]	2 (15)	9 (16)
	Pediatrician/general practitioner	0 (0)	3 (5)
	ENT/Otolaryngology	1 (8)	8 (14)
	Lactation consultants/midwives	0 (0)	1 (2)
	Dentist/oral surgeon	0 (0)	5 (9)
	Surgeon specialty unspecified	2 (15)	6 (10)
	Physician specialty unspecified	0(0)	1(2)
	NR/unclear	8 (62)	25 (43)
Anesthesia/analgesia/anesthetic	Topical or other method	67 (46)	20 (34)
	None	1 (8)	15 (26)
	NR	6 (46)	23 (40)
Lactation consultant/contact	Varying intensity	9 (69)	42 (72)
	NR	4 (31)	16 (28)
Other study characteristics			
Design	RCT	7 (54)	7 (12)
	NRSI	6 (46)	4 (7)
	Single arm	NA	47 (81)
Comparator	Breastfeeding support	7(54)	5 (9)
	Sham	3(23)	3(5)
	Immediate or delayed frenotomy	4 (31)	4 (7)
	No frenotomy	6 (46)	4 (7)
	No comparison	NA	47 (81)
Geographical Setting	United States	3 (23)	17 (29)
	Outside the United States	10 (77)	41 (71)

Study Characteristics	Subcharacteristics	EQ Number of Studies (%)	SQ Number of Studies (%)
ROB	Low	3 (23)	3 (5)
	Moderate	0 (0)	0 (0)
	High	10 (77)	8 (14)
	Other[d]	NA	47 (81)
Funding	Industry	0 (0)	0 (0)
	No industry	6 (46)	13 (22)
	Unfunded	1 (8)	16 (28)
	Not reported	6 (46)	29 (50)

[a] Includes studies that did not provide information on mean or median age or only provided age ranges of participants.
[b] Includes studies that did not report breakdown of gender of participants or only provided age ranges.
[c] Included midwives, dentists, doctors, ENT consultants, general practitioners, lactation consultants, midwives, neonatal surgeons, neonatologists, nurses, otolaryngology surgeons, pediatricians, pediatric dentists, pediatric surgeons, pediatric surgical consultants, or specialist oral ENT/surgeons.
[d] The quality of single-arm studies was assessed using a tool by Murad et al.;[22] ROB was not assessed.

Abbreviations: ENT = ear, nose, and throat specialist; EQ = efficacy question; NA = not applicable; NR = not reported; NRSI = nonrandomized study of intervention; RCT = randomized controlled trial; ROB = risk of bias; SQ = safety question.

3.2 Findings: Efficacy Outcomes

This section provides the findings for the effectiveness and comparative effectiveness of frenotomy or frenectomy for tongue-tie on efficacy outcomes from 13 studies (12 publications). No relevant studies were identified that examined the effectiveness or comparative effectiveness of frenotomy or frenectomy on tongue-tie with concomitant lip-tie or lip-tie alone. ***Table 4*** summarizes instruments used in outcome measurements reported in included studies. ***Appendix C*** includes study-level details on all efficacy outcomes. The section is organized by outcomes specific to the mother or the dyad (mother and child) followed by infant outcomes.

Table 4. Instruments used in outcome measurements in included studies

Scale Name	Description	Scoring Range
Breastfeeding Self-Efficacy Tool–Short Form (BSES - SF)[29-31]	14-item validated survey that measures breastfeeding efficiency and confidence	Individual items ranging from 1 or 0[a] (not very confident) to 5 (very confident). Range of score is 0 or 5–70, with higher scores indicating lower breastfeeding impairment/higher confidence.
EuroQoL-5 Dimensions, 5-level version (EQ-5D-5L)[31]	5 dimensions assess health-related quality of life and anxiety and depression: mobility, self-care, usual activities, pain/discomfort, and anxiety/depression	Levels for each domain are as follows: no problems, slight problems, moderate problems, severe problems, and unable to.
Infant Breastfeeding Assessment Tool (IBFAT)[27,29,35]	4 domains assess breastfeeding behavior of infants: readiness to feed, rooting, fixing [latching on], and sucking pattern	Each domain is scored out of 3. A minimum score is 0 and a maximum score is 12, with a higher school indicating better breastfeeding.
Gastroesophageal Symptom Scale - Infant (GSQ-I)[30]	Times occurred and severity of 6 symptoms associated with infant reflux; scale covers the last 7 days	Time: respondents report the number of times the issue occurred. Severity: respondents rate symptoms between 1 (not at all severe) and 7 (most severe).
Latch, Audible swallowing, Type of nipple, Comfort, Hold (LATCH)[28,29,34]	5 domains assess breastfeeding quality: latch, audible swallowing, type of nipple, comfort, and hold	Each domain is evaluated from 0–2 points, with 10 being a score indicating highest quality breastfeeding.
Short-Form McGill Pain Questionnaire (SF-MPQ)[27,31]	3-section scale adapted from the MPQ. Section one: a set of 15 words describing pain Section two: a visual analog scale Section three: a list of descriptors comprising the present pain intensity measure	Section one: Each word is graded on a 0–4 scale. Section two: Scores range between 0 (mild pain) and 10 (worst possible pain). Section three: Graded on a 0–5 point list. The measures are combined with a total possible score of 50 indicating the most severe pain.
Visual Analog Scale (VAS)[26,28-30]	Scale of 0–10 laid out on a horizontal line	Pain is rated between 0 (no pain) and 10 (worst pain).

[a] The items are usually reported as scored starting with 1. However, Ghaheri et al. 2022[30] state the individual domains are scored starting at 0 for not very confident, with 0 being the lowest possible total score.

3.2.1 Maternal or Dyad (Maternal and Child) Outcomes

Thirteen studies in 12 articles provided data on the maternal outcomes of breastfeeding pain,[26-31] LATCH scores,[29] breastfeeding self-efficacy,[29-31] and breastfeeding status (e.g., any, exclusive, and change in breastfeeding).[8,26,27,29,31-34,36]

3.2.1.1 Breastfeeding Pain

Six RCTs (3 high ROB,[29-31] 3 low ROB,[26-28] total N=452) reported on breastfeeding pain. Differences in comparators and timing of outcome (immediately after the procedure, 1 to 2 weeks after the procedure, 3 months after the procedure) precluded quantitative synthesis. We assessed the COE for this outcome as *very low* (***Table 5***).

Pain immediately after frenotomy: Three RCTs compared frenotomy with sham and measured maternal pain immediately after the procedure using the Short-Form McGill Pain Questionnaire (SF-MPQ),[27] a visual analog scale,[28] and an undefined breast pain scale.[26] Of these 3 RCTs, 2 reported that mothers in the frenotomy arm experienced reduced pain compared to sham. One study[27] reported a statistically significant reduction in pain scores of -8.6 on a 0 to 50 scale for the SF-MPQ (calculated 95% CI, −12.56 to −4.64); the second study[28] reported a *p* value for the difference between study arms of 0.001 but did not report the numeric difference on a visual

analog scale. A third study[26] reported a statistically nonsignificant difference of -1.2 on a 1 to 10 scale (95% CI, -0.3 to 2.4; *p*=0.13) among the subset of randomized women that they noted did not report pain at a sample feed prior to the intervention. Although all 3 RCTs were rated low ROB for study conduct, 2 of the 3 studies had reporting inconsistencies or errors (reporting standard errors as standard deviations[27] and results described differently in the abstract and the main results[28]).

Pain 1 to 2 weeks after frenotomy: Three high ROB RCTS reported on pain 1 to 2 weeks after frenotomy. Of these, 2 RCTs compared immediate frenotomy with delayed frenotomy. Outcomes were reported at 5[29] and 10 days[30] post-intervention, respectively (prior to frenotomy in the control arm). Both RCTs used a 0 to 10 visual analog scale. One study[30] reported a statistically significant mean difference of -1.5 (calculated 95% CI, −2.62 to −0.38) at 10 days follow-up; the second study[29] reported median reductions of 2 points in both study arms at 5 days follow-up, with overlapping confidence intervals for median change, suggesting no difference between groups.

One RCT (FROSTTIE) compared frenotomy with breastfeeding support to breastfeeding support alone; no crossover was planned, but 63 of 87 (72%) infants in the breastfeeding support only arm had frenotomy by 1–2 weeks postrandomization.[31] At 1 to 2 weeks follow-up, pain scores were similar (2 on a modified 10-point SF-MPQ scale) with an adjusted effect estimate of 0 (95% CI, −0.9 to 0.9).

Pain 3 months after frenotomy: By 3 months, an additional 2 control participants had frenotomy (73%) in the FROSTTIE trial. Additionally, the study had serious issues with recruitment because of difficulties with recruitment and the COVID-19 pandemic; the final sample size was not of sufficient power to provide precise outcomes.[31] The study reported no statistically significant differences between study arms on the modified SF-MPQ (median pain score of 0 in both arms, adjusted effect estimate: −0.2, 95% CI, −0.6 to 0.3).

Table 5. Breastfeeding pain: Summary of findings and certainty of evidence ratings

№ of Studies (№. Participants)	Risk of Bias	Inconsistency	Indirectness	Imprecision	Summary of Findings	CERTAINTY
Breastfeeding pain						
6 RCTs[26-31] (452)	Very serious[a]	Serious[b]	Not serious	Serious[c]	Inconsistent changes in pain scores. Variations in outcome measures and comparators, and timing precluded additional syntheses	⊕◯◯◯ VERY LOW

[a] Very serious concerns for bias because of failure to account for baseline differences;[29] randomization issues;[30] high rate of crossover;[29,31] outcome measurement.[29-31] In addition to these potential biases in study conduct, the evidence base may have had errors or inconsistencies in study reporting. One study[27] was rated at low ROB for conduct of the study but reported standard deviations as standard errors. One study[26] reported limiting results to women without pain at baseline. For those same women, however, the authors reported pain scores at baseline but did not explain why pain scores of 4.1 and 4.2 at baseline (for intervention and control, respectively) were not considered to be pain. Nor did they report baseline pain scores for women excluded from the pain analyses who had pain at baseline. One study, rated low ROB for conduct of the study, reported a difference in pain scores in the abstract differently than in the main results. In the abstract, the results were described as a comparison between the arms (after frenotomy vs. after sham). In the main text, the manuscript described the results as an overall change for all participants.[28] For one 1 high ROB study, reported confidence intervals appeared incorrect (negative signs were missing), so we recalculated them.[30]

[b] Serious concerns for consistency because of differences in point estimates.
[c] Confidence spanning the null and inclusive of appreciable benefits and harms for some outcomes. For 1 study,[29] it was unclear whether sample size calculations when reported assumed normal distribution, non-normal distribution reported.

Abbreviations: RCT = randomized, controlled trial; ROB = risk of bias.

3.2.1.2 Breastfeeding Effectiveness: LATCH Scores

One high ROB RCT (105 term infants <2 weeks old) reported using LATCH, a measure of breastfeeding effectiveness ranging from 0 to 10.[29] The RCT compared immediate frenotomy with breastfeeding support to breastfeeding support alone (with a planned delayed frenotomy at 5 days). Results were reported at 5 days after the procedure. Both study arms reported a median LATCH score of 1 (interquartile ranges from 0 to 2, *P*=0.52 for difference). Failure to account for baseline differences led us to assess this study as high ROB. We graded the COE for this outcome as *very low (**Table 6**)*.

Table 6. Breastfeeding effectiveness: Summary of findings and certainty of evidence ratings

№ of Studies (№. Participants)	Risk of Bias	Inconsistency	Indirectness	Imprecision	Summary of Findings	CERTAINTY
Breastfeeding effectiveness						
1 RCT[29] (105)	Serious[a]	NA—single study	Not serious	Very serious[b]	No significant difference between frenotomy vs delayed frenotomy (LATCH score of 1 [interquartile ranges from 0–2] in both arms; *P*=0.52 at 5 days)	⨁◯◯◯ VERY LOW

[a] Serious concerns for bias because of failure to account for baseline differences.
[b] Unclear whether sample size calculations assumed normal distribution, non-normal distribution reported. IQR of difference between arms NR, nonsignificant *P* value.

Abbreviations: IQR = interquartile range; LATCH= Latch, Audible swallowing, Type of nipple, Comfort, Hold; NA = not applicable; NR = not reported; RCT = randomized, controlled trial.

3.2.1.3 Breastfeeding Self-Efficacy

Three high ROB RCTs[29-31] (320 term infants ≤4 months old) reported results using the Breastfeeding Self-Efficacy Scale–Short Form (BSES). Two RCTs had planned crossovers;[29,30] the third (FROSTTIE) had unplanned crossover occur during the trial.[31] As with breastfeeding pain, unblinded assignment coupled with the expectation of crossover may have biased outcome measurement. One RCT specified the use of lasers,[30] while others did not specify the approach used. The 2 studies with planned crossover reported outcomes before the crossover occurred (at 5[29] and 10 days[30] after the procedure respectively); both reported statistically significant differences favoring the intervention arm.[29,30] FROSTTIE reported no statistically significant differences at 3 months. We graded the COE for this outcome as *low* for benefit for outcomes reporting within 5 to 10 days of the procedure (***Table 7***).

Table 7. Breastfeeding self-efficacy: Summary of findings and certainty of evidence ratings

№ of Studies (№ of Participants)	Risk of Bias	Inconsistency	Indirectness	Imprecision	Summary of Findings	CERTAINTY
Breastfeeding self-efficacy scale						
3 RCTs[29-31] (312)	Very serious[a]	Not serious	Not serious	Not serious	Significantly larger changes for frenotomy vs. control (delayed or no frenotomy) in 2 of 3 studies; Study 1 (delayed frenotomy)[29]: median change [IQR]: 9 (1.8 to 12.3) vs. 1 (−4 to 7.5); *p*=0.002 at 5 days Study 2 (delayed frenotomy with breastfeeding support)[30]: mean change 13.4 vs. −1.0; 95% CI, 9.2 to 19.7; *P*<0.001 at 10 days Study 3 (No frenotomy with breastfeeding support)[31]: median difference at 3 months, −0.3; 95% CI, −5.2 to 5.8, after significant unplanned crossover	⨁⨁◯◯ LOW for benefit

[a] Very serious concerns for bias because of failure to account for baseline differences;[29] randomization issues;[30] high rate of unplanned[31] or early crossover;[29] potential bias in outcome measurement associated with delayed crossover or desired frenotomy.[29-31]

Abbreviations: BSES=Breastfeeding Self-Efficacy Scale; IQR = interquartile range; RCT = randomized controlled trial.

3.2.1.4 Breastfeeding Initiation, Exclusivity, and Change

We assessed initiation of any breastfeeding and exclusive breastfeeding at both short-term (≤2 months) and longer-term timeframes (>2 months). The longest-term follow-up for any breastfeeding or exclusive breastfeeding was 6 months post-intervention. The sections below address findings from RCTs and nonrandomized studies for any breastfeeding and exclusive breastfeeding in short- and longer-term follow-up. We present these outcomes by timeframe as initiation of breastfeeding is often immediately encouraged after birth but may not be continued at home.

Initiation of Any Breastfeeding

Two RCTs with high ROB[29,31] and 4 serious ROB cohort studies with comparison groups evaluated prevalence of any breastfeeding (e.g., exclusive or nonexclusive breastfeeding). The RCTs compared frenotomy with either breastfeeding support[31] or delayed frenotomy.[29] Both RCTs reported that all participants also received some level of breastfeeding support or monitoring. Authors assessed any breastfeeding at timepoints ranging from 5 days to 6 months post-intervention. In 1 RCT with unplanned crossover (FROSTTIE), most control group infants had received frenotomy at the time of assessment of this outcome at 3 months.[31] The other RCT reported outcomes at 5 days, before 83% of control group infants had received frenotomy.[29]

Four cohort studies (1 publication reports 2 studies[32]) with serious ROB reported on any breastfeeding;[32,33,36] All 4 studies compared frenotomy with no frenotomy, and none explicitly

reported providing breastfeeding support. The studies assessed report of any breastfeeding at varied timepoints: at 1, 3, and 6 months follow-up in 1 study[33] and at a median of 87 or 118 days in 2 others (reported in 1 publication).[32] One study did not report timing of follow-up (survey of mothers after procedure/no procedure).[36]

Any breastfeeding at 2 months or less follow-up

One RCT reported no significant group differences in any breastfeeding in short-term follow-up.[27,29] The RCT[29] reported outcomes at 5 days, by which time 9 of 52 control group infants had had an early frenotomy.[29] Report of any breastfeeding did not differ between groups (OR vs. bottle feeding: 0.57; 95% CI, 0.17 to 1.88).[29] We graded the COE for this outcome as *very low* certainty (***Table 8***).

One cohort study reported no difference in any breastfeeding in infants who received frenotomy compared with those who did not at 1 month (95% vs. 85%, calculated prevalence ratio [PR]: 1.12; 95% CI, 0.98 to 1.29).[33] We graded the COE for this outcome as *very low* certainty (***Table 8***).

Table 8. Initiation of any breastfeeding at 2 months or less follow-up: Summary of findings and certainty of evidence ratings

№ of Studies (№ of participants)	Risk of Bias	Inconsistency	Indirectness	Imprecision	Summary of Findings	CERTAINTY
Any Breastfeeding at 2 months or less						
1 RCT[29] (105)	Very serious[a]	NA—single study	Not serious	Very serious[b]	No significant differences at 5 days for frenotomy vs. delayed frenotomy (48/53 [91%] vs. 44/52 [85%], OR, 0.57; 95% CI, 0.17 to 1.88)	⊕◯◯◯ VERY LOW
1 cohort with comparison[33] (159)	Very serious[c]	NA-single study	Not serious	Serious[d]	No difference at 1 month follow-up for frenotomy vs. no frenotomy (114/120 [95%] vs.33/39 [85%]; calculated PR, 1.12; 95% CI, 0.98 to 1.29)	⊕◯◯◯ VERY LOW

[a] Very serious concerns for bias because of differences at baseline and early crossover.
[b] Confidence spanning the null and inclusive of appreciable benefits.
[c] Very serious concerns for bias in cohort study due to confounding, potential attrition bias, and potential bias in outcome measurement.
[d] Confidence spanning the null and inclusive of appreciable benefits and harms.

Abbreviations: OR = odds ratio; PR = prevalence ratio; RCT = randomized, controlled trial.

Any breastfeeding at more than 2 months follow-up

Any breastfeeding at more than 2 months follow-up did not differ by group in the FROSTTIE study.[31] The study only reported outcomes after most control group participants had received frenotomy as unplanned crossovers; report of any breastfeeding was not different between groups at 3-month follow-up (absolute risk ratio [aRR] in intention-to-treat [ITT] analysis: 1.02; 95% CI, 0.90 to 1.16, and 1.27; 95% CI, 0.99 to 1.64 in per protocol analysis) or 6-month

follow-up (aRR: 0.98; 95% CI, 0.84 to 1.14).[31] This RCT also reported an as-treated analysis (grouping infants according to the intervention received vs. the arm to which they were randomized) in which the frenotomy group had statistically significantly higher rates of any breastmilk feeding at 3 months (90.4% vs. 69.2%, aRR: 1.35; 95% CI, 1.05 to 1.74); however, this difference may be due to confounding not accounted for in the analysis.[31] Similarly, the lack of group differences in the ITT analysis may due to unplanned crossovers. We graded the COE as *low* for no difference ***(Table 9)***.

Four cohort studies reported no differences in breastfeeding.[32,33,36] One study reported similar rates of any breastfeeding in infants who received frenotomy compared with those who did not at 3 months (93% vs. 79%, calculated PR: 1.17; 95% CI, 0.99 to 1.39) and 6 months (92% vs. 79%, calculated PR: 1.15; 95% CI, 0.97 to 1.36).[33] Three other cohort studies also reported similar rates of any breastfeeding between those who received frenotomy and those who did not. In 1, 83% of frenotomy infants and 67% of control infants reported breastfeeding for a mean of 6 to 7 months (calculated RR: 1.24; 95% CI, 0.78 to 1.99).[36] Two studies (reported in 1 publication) did not report significant group differences in breastfeeding among infants receiving or not receiving frenotomy. In 1 study assessing conducted after the study hospital instituted use of standardized assessment to evaluate tongue-tie severity (Bristol Tongue-Tie Assessment Tool [BTAT]), 77% of infants receiving frenectomy and 82% who did not reported any breastfeeding at median of 87 days follow-up (calculated PR: 0.94; 95% CI, 0.76 to 1.17).[32] A second study conducted at the same hospital assessed breastfeeding after changes to the hospital's frenotomy process (including using the BTAT plus lactation consultant assessment depending on infant age and BTAT score). Fewer infants ultimately received frenotomy under the new pathway. At median 118 days, 71% of infants with frenotomy (n=24/34) and 100% (n=1/1) without frenotomy reported any breastfeeding (calculated Peto OR: 0.247; 95% CI, 0.003 to 18.89).[32] We graded the COE as *very low* (***Table 9***).

Table 9. Initiation of any breastfeeding at more than 2 months follow-up: Summary of findings and certainty of evidence ratings

№ of Studies (№ of Participants)	Risk of Bias	Inconsistency	Indirectness	Imprecision	Summary of Findings	CERTAINTY
Any breastfeeding at more than 2 months						
1 RCTs[31] (163)	Very serious[a]	NA—single study	Not serious	Not serious	No statistically significant differences for frenotomy vs. no frenotomy ITT (outcomes with significant unplanned crossover) 3 months: 67/80 (88%) vs. 75/89 (86%); aRR, 1.02; 95% CI, 0.90 to 1.16; *p*=0.73 6 months: 55/66 (83%) vs. 60/71 (85%), aRR, 0.98; 95% CI, 0.84 to 1.14 Per protocol (at 3 months) n=65/75 (90%) vs. 16/24 (27%); aRR, 1.27; 95% CI, 0.99 to 1.64; *p*=0.06	⊕⊕◯◯ LOW for no difference
4 cohorts with comparison[32,33,36]	Very serious[b]	Not serious	Not serious	Serious[c]	Similar prevalence between study arms (frenotomy vs. no frenotomy)	⊕◯◯◯ VERY LOW

№ of Studies (№ of Participants)	Risk of Bias	Inconsistency	Indirectness	Imprecision	Summary of Findings	CERTAINTY
(one publication reports 2 studies[32]) (471)					Study 1[33]: At 3 months, 112/120 [93%] vs. 31/30 [79%]; calculated PR, 1.17; 95% CI, 0.99 to 1.39) At 6 months, 110/120 [92%] vs. 31/39 [79%]; calculated PR, 1.15; 95% CI, 0.97 to 1.36 Study 2[36]: At mean 6 to 7 months, 68/82 (83%) vs. 6/9 (67%); calculated RR, 1.24; 95% CI, 0.78 to 1.99 Study 3(Dixon et al 2018, study 1)[32]: At median 87 days, 127/164 (77%) vs. 18/22 (82%); calculated PR, 0.94; 95% CI, 0.76 to 1.17 Study 4(Dixon et al 2018, study 2)[32]: At median 118 days, 24/34 (71%) vs. 1/1 (100%), calculated Peto OR, 0.247; 95% CI, 0.003 to 18.89	

[a] Very serious concerns for bias because of high crossover rate and lack of blinding.
[b] Very serious concerns for bias in cohort studies due to lack of controls for confounding, potential attrition bias, and potential bias in outcome measurement.
[c] Confidence spanning the null and inclusive of appreciable benefits and/or harms.

Abbreviations: aRR = adjusted risk ratio; ITT = intention to treat; NA = not applicable; OR = odds ratio; PR = prevalence ratio; RCT = randomized, controlled trial; RR = risk ratio.

Exclusive Breastfeeding

Two RCTs with high ROB[29,31] and 3 cohort studies (1 publication[32] reports 2 studies),[32,33] all with serious ROB, reported outcomes for exclusive breastfeeding at any follow-up timepoint. One RCT compared frenotomy with delayed frenotomy and assessed outcomes at 5 days before 83% of control group infants had received frenotomy; all participants received some level of breastfeeding support or monitoring.[29] The FROSTTIE trial compared frenotomy plus breastfeeding support to breastfeeding support alone.[31] Follow-up occurred at 1 to 2 weeks and 3 months; at each time point, most infants in the support only arm had also received frenotomy (i.e., 63 of 89 [71%] infants in the breastfeeding support only arm had frenotomy by 1 to 2 weeks postrandomization, and an additional 2 had frenotomy by the 3-month follow-up).[31]

Three serious ROB cohort studies compared frenotomy with no frenotomy, and none explicitly reported providing breastfeeding support.[32,33] The studies assessed exclusive breastfeeding at varied timepoints: at 1, 3, and 6 months in 1 study[33] and at a median of 87 or 118 days in 2 others.[32]

Exclusive breastfeeding at 2 months or less follow-up

Two RCTs reported no statistically significant differences in exclusive breastfeeding in short-term follow-up.[29,31] One RCT reported no group differences at 5 days (OR: 1.40; 95% CI, 0.60 to 3.22; p=0.43).[29] In FROSTTIE, authors reported no differences in exclusive direct breastfeeding

in the prior 24 hours (45% vs. 49%; aRR: 0.92; 95% CI, 0.59 to 1.45) at the 1- to 2-week follow-up.[31] We graded the COE for this outcome as *very low* (***Table 110***).

In 1 cohort study, exclusive breastfeeding prevalence in treated and untreated infants did not differ at 1 month (PR: 0.95; 95% CI, 0.78 to 1.17).[33] We graded the COE for this outcome as *low* (***Table 110***).

Table 10. Exclusive breastfeeding at 2 months or less follow-up: Summary of findings and certainty of evidence ratings

№ of Studies (№ of Participants)	Risk of Bias	Inconsistency	Indirectness	Imprecision	Summary of Findings	CERTAINTY
Exclusive breastfeeding at 2 months or less						
2 RCTs[29,31] (265)	Very serious[a]	NA—single study	Not serious	Very serious[b]	No statistically significant differences for frenotomy vs. no frenotomy Study 1 at 5 days follow-up: 35/53 [66%] vs. 38/52 [73%]; OR, 1.40; 95% CI, 0.60 to 3.22)[29] Study 2 at 1 to 2 weeks follow-up (35/80 [45%] vs. 43/89 [49%]; aRR, 0.92; 95% CI, 0.59 to 1.45)[31]	⊕◯◯◯ VERY LOW
1 cohort with comparison[33] (159)	Very Serious[c]	NA-single study	Not serious	Not serious	No difference at 1 month follow-up for frenotomy vs. control (88/120 [73%] vs. 30/39 [77%]; PR, 0.95; 95% CI, 0.78 to 1.17)[33]	⊕⊕◯◯ LOW No difference

[a] Very serious concerns for bias in RCTs because of failure to account for baseline differences, randomization issues, rate of crossover, and outcome measurement.
[b] Confidence spanning the null and inclusive of appreciable benefits and harms.
[c] Very serious concerns for bias in outcome measurement and because of confounding, outcome reporting, and attrition in cohort study.

Abbreviations: aRR = adjusted risk ratio; NA = not applicable; OR = odds ratio; PR = prevalence ratio; RCT = randomized controlled trial.

Exclusive breastfeeding at more than 2 months follow-up

One RCT, FROSTTIE, reported no significant differences in exclusive breastfeeding at longer-term follow-up. There were no differences at the 3-month follow-up, by which time most control arm infants received frenotomy (exclusive direct breastfeeding: 54% vs. 53%; aRR: 1.03; 95% CI, 0.65 to 1.62).[31] We graded the COE for this outcome as *very low* (***Table 11***).

In 3 cohort studies with longer-term follow-up, exclusive breastfeeding rates were similar between groups. Prevalence in treated and untreated infants did not differ at 3 months (PR: 0.94; 95% CI, 0.75 to 1.19) or 6 months (PR: 0.92; 95% CI, 0.72 to 1.16) in 1 study.[33] In 1 study conducted after the study hospital instituted use of the BTAT to evaluate tongue-tie severity, exclusive breastfeeding rates were 54% in the frenotomy arm compared with 46% in the no frenotomy arm (calculated RR: 1.19; 95% CI, 0.74 to 1.93) at median 87 days.[32] In a further study at the same hospital after a revised treatment pathway including the BTAT plus lactation

consultant assessment, exclusive breastfeeding rates were 56% in the frenotomy arm and 0% (n=0/1) in the no frenotomy arm (calculated Peto OR: 8.91; 95% CI, 0.17 to 455.73) at median 118 days.[32] We graded the COE for this outcome as *very low (**Table** 11**11**)*.

Table 11. Exclusive breastfeeding at more than 2 months follow-up: Summary of findings and certainty of evidence ratings

№ of Studies (№ of Participants)	Risk of Bias	Inconsistency	Indirectness	Imprecision	Summary of Findings	CERTAINTY
Exclusive breastfeeding at more than 2 months						
1 RCT[31] (163)	Very Serious[a]	NA-single study	Not serious	Very serious[b]	No difference at 3 months for frenotomy vs. no frenotomy (38/80 [54%] vs. 39/89 [53%]; aRR,1.03; 95% CI, 0.65 to 1.62); outcome had significant unplanned crossover[31]	⨁◯◯◯ VERY LOW
3 cohorts with comparison[32,33] (1 publication reports 2 studies[32]) (380)	Very Serious[c]	Not serious	Not serious	Very serious[b]	No difference for frenotomy vs. control Study 1 (Dixon et al. 2018 study 1[32]): 89/164 [54%] vs.10/22 [46%]; calculated RR, 1.19; 95% CI, 0.74 to 1.93 at median 87 days followup Study 2 (Dixon et al. 2018, study 2[32]): 19/34 [56%] vs. 0/1 [0%]; calculated Peto OR, 8.91; 95% CI, 0.17 to 455.73) at median 118 days Study 3[33]: at 3 months 81/120 [68%] vs. 28/39 [72%]; PR, 0.94, 95% CI, 0.75 to 1.19) and at 6 months (79/120 [66%] vs. 29/39 [7%]; PR, 0.92; 95% CI, 0.72 to 1.16)	⨁◯◯◯ VERY LOW

[a] Very serious concerns for bias in RCTs because of failure to account for baseline differences, randomization issues, rate of crossover, and outcome measurement.
[b] Confidence spanning the null and inclusive of appreciable benefits and harms.
[c] Very serious concerns for bias in outcome measurement and because of confounding, outcome reporting, and attrition in cohort study.

Abbreviations: aRR= adjusted risk ratio; PR = prevalence ratio; RCT = randomized controlled trial.

Change in Breastfeeding

Four studies reported outcomes (maternal-assessed improvement, cessation, breastfeeding problems) that we classified broadly as "change in breastfeeding." We graded the COE for this outcome as *very low* (***Table 12***)

Two RCTs with high ROB[8,26] comparing frenotomy with no frenotomy[26] or breastfeeding support[8] reported breastfeeding improvement post-intervention[26] or at an unspecified timepoint,[8] both using unvalidated measures. In both studies, significantly more mothers whose infants received frenotomy reported improvement compared with the control arms: 78% vs. 47% (calculated RR: 1.73; 95% CI, 1.13 to 2.65) in 1 study[26] and 96% vs. 3% (calculated RR: 28.0;

95% CI, 4.07 to 192.12) in the other study.[8] One of these 2 RCTs also reported objective observer ratings of improvement (using an unvalidated instrument), which were not significantly different between groups (50% vs. 40%, *p*=ns).[26]

Two cohort studies with serious ROB reported change in breastfeeding including stopping breastfeeding due to tongue-tie related difficulty or pain[36] or undefined breastfeeding problems.[34] Both studies compared frenotomy with no frenotomy and followed up participants at a mean of 2.5 weeks in 1 study[34] and at unspecified timepoint in the second.[36] In 1 study, individuals whose infants received frenotomy reported fewer breastfeeding problems than control participants (13% vs. 60%; calculated RR: 0.22; 95% CI, 0.07 to 0.70),[34] and in the other study, fewer individuals in the frenotomy arm stopped breastfeeding due to tongue-tie related difficulty or pain (17% vs. 33%; calculated RR: 0.51; 95% CI, 0.18 to 1.45).[36]

Table 12. Change in breastfeeding: Summary of findings and certainty of evidence ratings

№ of Studies (№ of Participants)	Risk of Bias	Inconsistency	Indirectness	Imprecision	Summary of Findings	CERTAINTY
Change in breastfeeding						
2 RCTs[8,26] (114)	Serious[a]	Not serious	Serious[b]	Serious[c]	Significant improvement in frenotomy arm vs. control in both RCTs Study 1: 78% (21/26) vs. 47% (14/30); *p*<0.02; calculated RR; 1.73; 95% CI: 1.13 to 2.65[26] Study 2:96% (27/28) vs. 3% (1/29); *p*<0.001; calculated RR: 28.0; 95% CI, 4.07 to 192.12[8]	⊕◯◯◯ VERY LOW
1 cohort study[34] (33)	Very serious[d]	NA—single study	Serious[b]	Serious[rc]	Fewer problems in frenotomy arm vs. control participants (13% [n=3/23] vs. 60% [n=6/10]; calculated RR: 0.22; 95% CI, 0.07 to 0.70)[34]	⊕◯◯◯ VERY LOW
1 cohort study[36] (91)	Very serious[d]	NA-single study	Not serious	Very serious[e]	Fewer individuals in frenotomy arm vs. control stopped breastfeeding due to tongue-tie related difficulty or pain (17%, [14/82] vs. 33% [3/9]; calculated RR: 0.51; 95% CI, 0.18 to 1.45)[36]	⊕◯◯◯ VERY LOW

[a] High ROB in RCTs due to issues with randomization and outcome assessment.[8,26]
[b] Serious indirectness due to unvalidated measures.
[c] Serious imprecision due to ratio of confidence intervals (≥3 for RR), suggesting that OIS was not met.
[d] Very serious ROB due to confounding and potential bias in outcome measurement.
[e] Very serious imprecision because confidence intervals do not exclude the null and include appreciable benefit and appreciable harm

Abbreviations: OIS=optimal information size; NA = not applicable; RCT = randomized, controlled trial; ROB = risk of bias; RR = risk ratio.

3.2.2 Infant Outcomes

Four studies, all RCTs, provided data on the outcomes of infant weight gain,[31] the IBFAT,[27,29] and the Gastroesophageal Symptom Questionnaire for Infants (GSQ-I).[30]

3.2.2.1 Infant Weight Gain

The high ROB FROSTTIE study (n=169 infant/mother dyads randomized) reported outcomes for infant weight gain.[31] The study compared differences in weight for age as a z-score[105] for infants who received a frenotomy with breastfeeding support with those who received only breastfeeding support and found no differences. We graded the COE for this outcome as *very low* (***Table 13***).

Table 13. Infant weight gain: Summary of findings and certainty of evidence ratings

№ of Studies (№ of Participants)	Risk of Bias	Inconsistency	Indirectness	Imprecision	Summary of Findings	CERTAINTY
Infant Weight Gain						
1 RCT[31] (163)	Very serious[a]	NA-single study	Not serious	Serious[b]	No significant difference at 3 months, z-score for weight for age, −1.0 (SD 1.6) vs. −1.1 (SD 1.3); adjusted mean difference in z-score: 0.10 (95% CI, −0.83 to 1.03; *p*=0.83)	⨁◯◯◯ VERY LOW

[a] Very serious concerns for bias because of high crossover rate and lack of blinding.
[b] Confidence intervals span the null and exceed +1 standard deviation, thus including both no difference and appreciable benefit.

Abbreviations: NA = not applicable; RCT = randomized, controlled trial; SD = standard deviation.

3.2.2.2 Infant Breastfeeding Behavior

Two RCTs reported findings using the Infant Breastfeeding Assessment Tool (IBFAT, scores range from 0 to 12), one with low ROB in the conduct of the study, but with potential errors in reporting results[27] and the second with high ROB.[29] The low ROB study compared infants who received a frenotomy with those who received a sham frenotomy.[27] After we corrected for errors in reporting, results from the low ROB study showed a significant improvement in IBFAT scores of 3.53 immediately after the procedure.[27] The high ROB study did not measure IBFAT immediately after the procedure but found no differences five days after procedure but prior to crossover (0 [95% CI, −1.8 to 1.0] vs 0 [95% CI 0–1], p=0.36)[29]. We graded the COE for this outcome as *Very Low* (***Table 14***).

Table 14. IBFAT scores: Summary of findings and certainty of evidence ratings

№ of Studies (№ of Participants)	Risk of Bias	Inconsistency	Indirectness	Imprecision	Summary of Findings	CERTAINTY
Infant Breastfeeding Assessment Tool (IBFAT)						
2 RCTs[27,29] (165)	Serious[a]	Serious[b]	Not serious	Serious[c]	Study 1[27]: Significant improvement in score immediately after procedure (calculated mean difference: 3.53; 95% CI, 1.22 to 5.84) Study 2[29]: No change in median score 5 days after procedure: 0 (95% CI, −1.8 to 1) vs. 0 (95% CI, 0 to 1); *p*=0.36	⨁◯◯◯ VERY LOW

[a] One study[27] was rated low ROB for conduct of the study but reported standard deviations as standard errors. One study[29] was rated high ROB because of failure to account for baseline differences.
[b] Inconsistent evidence of benefit arising from multiple possible sources including ROB, timing of outcome measurement, and errors in reporting.

[c] One study reported a large effect size. The sample size of the study (N=58) did not reach 30% to 50% of the optimal information size of 336, based on a baseline value of 8.5 in the control arm and 9.3 in the intervention arm, 1 standard deviation of 3.7, alpha=0.05, and power of 0.80.[27] The second study reported nonoverlapping confidence intervals for median scores, suggesting lack of precision.[29]

Abbreviations: RCT = Randomized, controlled trial; ROB = risk of bias.

3.2.2.3 Other Infant Outcomes

One RCT reported on gastroesophageal symptoms, as measured by the GSQ-I.[30] The RCT had high ROB arising from deviations from unintended interventions and lack of blinding. Infants received a frenotomy either immediately after randomization or were placed on a 10-day wait list. No crossover between groups was reported. Results were compared between groups at day 10 for the number of symptoms occurring and severity of occurring symptoms for 12 measures. Calculated effects showed statistically significant differences between groups when comparing events between day 0 and day 10 for 2 of 12 measures (***Table 15***Table 15). We graded the COE for this outcome as *very low*.

Table 15. Other infant outcomes: Summary of findings and certainty of evidence ratings

№ of Studies (№ of Participants)	Risk of Bias	Inconsistency	Indirectness	Imprecision	Summary of Findings	CERTAINTY
Gastroesophageal Symptom Questionnaire for Infants (GSQ-I)						
1 RCT[30] (48)	Very serious[a]	NA—single study	Not serious	Serious[b]	No significant differences (after Bonferroni corrections for multiple comparison) for calculated mean differences and CIs for 10 of 12 domain measures in the GSQ-I;[c,d] 4 domains did not exclude the null (refusal to feed [times], episodes of hiccups [times], choking/gagging [severity], irritability/fussiness [times]). Six; 6 domains excluded the null but had *p* values that did not meet the threshold for multiple comparisons (*p*=0.004) (arching back [times and severity], episodes of hiccups [severity], irritability/fussiness [severity], refusal to feed [severity], and vomiting [times]) Statistically significant differences for number of times infants experienced choking/gagging (−9.8, 95% CI, −-16.9 to −3.7;, uncorrected *p*=0.002) and severity of vomiting/regurgitation (−1.9;, 95% CI, −3.0 to −-0.8;, uncorrected *p*=0.001))	⨁◯◯◯ VERY LOW

[a] Very serious concerns for bias because of method of randomization and measurement of outcomes.
[b] Confidence intervals for most domain measures do not exclude the null.
[c] Reported confidence intervals appeared incorrect (negative signs were missing), so we calculated them for each domain.
[d] Mean differences (CIs and *p* values) for (1) vomiting (times): −7.9 (−15.68 to −0.12; 0.057); (2) vomiting (severity): −1.9 (−2.99 to −0.81; 0.001); (3) irritability/fussiness (times): −5.5 (−13.77 to 2.77, 0.19); (4) irritability/fussiness (severity): −1.5 (−2.73 to −0.27; 0.02); (5) refusal to feed (times): −2.3 (−4.62 to 2.48; 0.05); (6) refusal to feed (severity): −1.3 (−2.39 to −0.21; 0.02); (7) choking/gagging (times): −9.8 (−15.93 to −3.67; 0.002); (8) choking/gagging (severity): −0.8 (−1.72 to 0.12; 0.09); (9) arching back (times): −9.7 (−17.03 to −2.37; 0.01); arching back (severity): −1.4 (−2.37 to −0.43; 0.005); (10) episodes of hiccups (times): −4.2 (−8.76 to 0.36; 0.07); and (11) episodes of hiccups (severity): −1.1 (−1.88 to −0.32; 0.006).

Abbreviations: NA = not applicable; RCT = randomized controlled trial.

3.3 Findings: Safety Outcomes

This section provides an overview of the findings on harms of frenotomy for tongue-tie and/or lip-tie for breastfeeding support. ***Appendix C*** includes detailed tables on all harm outcomes. We describe results for 58 studies (57 publications) for harms below. The section is organized by study design, frenotomy method, and oral tie type. Seven of the included studies were RCTs, four were NRSIs, and 47 were single-arm studies. Because harms may vary by frenotomy method, we organized the findings by method: scissors, lasers, or unspecified. ***Table 16.*** *Error! Reference source not found.* presents the number of studies and designs addressing each procedure and indication. Of the included studies, the majority reported harms that were not severe or related to the procedure itself such as minor bleeding, crying, or pain. However, more serious complications were sometimes reported, including damage to other structures in the mouth (e.g., salivary ducts), weight loss and increased feeding difficulties following the procedure, and hospital readmission. Adverse event rates varied significantly across studies owing to inconsistencies in how they were assessed and reported and therefore do not enable comparisons by frenotomy method.

Table 16. Harms of frenotomy for tongue-tie by procedure type

Method of Frenotomy Procedure	Indication	Comparative Studies Reporting Specific Complications	Comparative Studies Reporting No Complications	Comparative Studies Reporting Complications	Single-Arm Studies Reporting No Complications	Single-Arm Studies Reporting Overall Complications
Scissors	**Tongue-tie only**	NA	3[8,27,35]	4[26,32,36]	14[38,40,45,48,50,62,64,66,71,72,76,78,80,81]	14[11,37,39,41,43,53,58,59,63,65,70,75,77,79]
	Tongue-tie and/or lip-tie	NA	NA	NA	2[69,79]	3[42,67,82]
	Unspecified tie type	NA	NA	NA	1[73]	NA
Laser	**Tongue-tie only**	NA	1[30]	NA	NA	2[47,74]
	Tongue-tie and/or other specified tie type	NA	NA	NA	2[51,55]	3[56,68,74]
Unspecified Method	**Tongue-tie only**	1[31]	NA	3[28,29,31]	2[44,49]	1[46]
	Tongue-tie and/or unspecified tie type	NA	NA	NA	1[57]	1[54]
	Unclear/ unspecified tie type	NA	NA	NA	NA	1[60]

Abbreviations: NA = not applicable

A single high ROB RCT compared rates of complications between frenotomy with unspecified methods for tongue-tie and breastfeeding support (1/80 [1.25%] vs. 2/89 [2.2%]).[31] Potential for bias, few events, and small sample sizes resulted in *very low* certainty of evidence **(*Table 17*)**.

Table 17. Harms of frenotomy with unspecified methods for tongue-tie: Summary of findings and certainty of evidence ratings

№ of Studies	Risk of Bias	Inconsistency	Indirectness	Imprecision	Summary of Findings	CERTAINTY
Incidence of Complications						
1 RCT[31] (169)	Very serious[a]	NA—single study	Not serious	Extremely serious[b]	No significant difference (1/80 [1.25%] vs. 2/89 [2.2%]; OR: 0.55; 95% CI: 0.05 to 6.19)[31]	⨁◯◯◯ VERY LOW

a. Serious concerns for bias because of high crossover rate and lack of blinding.
b. Extremely wide confidence intervals including appreciable benefit and appreciable harm and suggest very different inferences.

Abbreviations: OR = odds ratio; RCT = randomized controlled trial.

No other study compared complications by arm; as a result, we did not grade the evidence for the other studies. Some studies reported that they found no complications without specifying additional details (3 comparative studies, 22 single-arm studies). The remainder reported incidence of specific complications in 1 arm (7 comparative studies, 25 single-arm studies).

3.3.1 Frenotomy with Scissors

One study of participants with unspecified tie types explicitly reported that no complications or harms occurred based on patient files and parent report.[73] Results for infants with tongue-tie only or tongue-tie and/or lip-tie are in Sections 3.4.1.1 and 3.4.1.2, respectively.

3.3.1.1 Tongue-Tie Only

Three RCTs,[8,26,27] and 4 NRSIs,[32,35,36] reported on harms of frenotomy with scissors for tongue-tie (***Table 18***). Comparisons used in the comparative studies included sham frenotomy,[26,27] no frenotomy,[26,27] and no frenotomy with breastfeeding support.[32,36] Of the comparative studies, 3 studies explicitly reported that there were no complications or harms.[8,35] Between 0.5% to 10% of participants experienced minor bleeding;[8,27,35] there were 2.6% cases of tongue-tie recurrence;[36] and 0.3% to 8% of participants needing repeat surgery.[32,36] A single participant required paracetamol for analgesia.[32]

Table 18. Summary of harms comparing frenotomy with scissors to no frenotomy (i.e., sham, no frenotomy, or breastfeeding support) for tongue-tie

№ of Studies	N at Follow-up	Summary of Findings
Explicitly reported that no complications occurred		
2 RCTs[8,27] 1 Cohort Study[35]	94	No complications or harms from the procedure
Minor bleeding		
1 RCT[26] 2 Cohorts[32]	225	0.5% to 10% of participants reported minor bleeding
Recurrence of tongue-tie		
1 Cohort[36]	82	2.6% of ties reoccurred secondary to scarring
Revisions/repeat surgery		
3 Cohort[32,36]	280	0.3% to 8% of participants needed a repeat surgery
Required paracetomol for analgesia		
1 Cohort[32]	259	0.3% of participants required paracetamol for analgesia

Abbreviations: N=number; RCT = randomized, controlled trial.

Twenty-eight single-arm studies[11,37-41,43,45,48,50,53,58,59,61-66,70-72,75-78,80,81] reported on harms of frenotomy with scissors for tongue-tie (***Table 19***).

Table 19. Summary of harms from single-arm studies of frenotomy with scissors for tongue-tie only

№ of Studies	N at Follow-up	Summary of Findings
Explicitly reported that no complications occurred		
14 Single arm[38,40,45,48,50,62,64,66,71,72,76,78,80,81]	1,292	No complications or harms were reported from the procedure.
Adverse events		
1 Single arm[37]	474	4.4% reported adverse events.[a]
Bleeding		
5 Single arm[37,39,53,63,75,77]	1,060	2.8% to 100% reported bleeding during or after the procedure.[a]
Brown posset due to swallowed blood		
1 Single arm[53]	215	0.5% reported "brown posset" due to swallowed blood.
Feeding deteriorated		
1 Single arm[59]	175	0.5% reported that feeding deteriorated after the procedure.
Fever		
1 Single arm[70]	126	0.8% reported infant having a fever for 1 day.
Need a Syringe for feeding		
1 Single arm[59]	175	0.57% of previously breastfeeding infants required syringe feeding after the procedure.
Irritability/Crying		
5 Single arm[11,37,41,53,59]	921[a]	3% to 60% reported irritability or crying after the procedure.[a,b]
Need for repeat procedure		
4 Single arm[39,43,58,79]	792	0.66% to 6.5% reported needing a repeat procedure.
Readmission		
1 Single arm[37]	474	1.1% of participants had to be readmitted.[a]
Refusal to drink from breast of bottle		
1 Single arm[70]	126	0.8% of infants refused to drink from breast or bottle for 2 hours after the procedure.
Reoccurrence/reattachment		
2 Single arm[59,65]	264	1.5% to 2% reported reattached of tongue-tie or reoccurrence.
Scarring		
1 Single arm[37]	474	38% reported scarring.[a]
Soreness/discomfort		
2 Single arm[53,63]	251	0.5% to 5.6% reported soreness or discomfort.
Swelling		
2 Single arm[37,59]	649	4.1% to 5% reported swelling.[a]
Ulcers		
4 Single arm[37,53,63,77]	984	2% to 100% reported ulcers.[a]
Worse pain and latch difficulties		
1 Single arm[59]	175	0.57% reported worse pain and latch difficulties at follow-up.

Notes:[a] Geddes et al. 2008[11] reported 24 participants at baseline; the number of participants at followup was unclear.
[b] Akbari et al. 2023[37] reported adverse events in general as well as specific adverse events (bleeding, ulcers, swelling, irritability, scar tissue). It is unclear if the 4.4% experiencing adverse events is inclusive of the other events.
[c] Geddes et al. 2008[11] was not included in the estimate due to a lack of specificity.

Abbreviations: N=number.

3.3.1.2 Tongue-Tie and/or Lip-Tie (Specified)

Five single-arm studies of frenotomy with scissors reported on harms in populations with tongue-ties and/or lip-ties (***Table 20***Table 20).[42,67,69,79,82]

Table 20. Summary of harms for single-arm studies of frenotomy with scissors for tongue-tie and/or lip-tie

№ of Studies	N at Follow-up	Summary of Findings
Explicitly reported that no complications occurred		
2 Single arm[69,79]	715	Reported no complications or harms.
Frenotomies were revised		
1 Single arm,[42]	33	6% of frenotomies were revised.
Need for cauterization with silver nitrate		
1 Single arm[67]	157	1% needed cauterization with silver nitrate for persistent oozing.
Pain		
1 Single arm[82]	41	24.4% reported notable postoperative pain.

Abbreviations: N=number.

3.3.2 Frenotomy with Laser

3.3.2.1 Tongue-Tie Only

One RCT (n=47)[30] of frenotomy with laser for tongue-tie compared with delayed frenotomy reported no adverse events or unanticipated problems.[30]

Two single-arm studies[47,74] reported on harms of frenotomy with laser for tongue-tie only (***Table 21***Table 21).

Table 21. Summary of harms from single-arm studies of frenotomy with laser for tongue-tie only

№ of Studies	N at Follow-up	Summary of Findings
Bleeding		
1 Single arm[47]	56	30.4% reported bleeding during the procedure and 1 case had punctiform bleeding due to accidental trauma 7 days after the procedure.
Carbonization of the irradiated site		
1 Single arm[47]	56	19.6% needed the irradiated site carbonized during procedure.
Crying		
1 Single arm[47]	56	96.4% had a high pitched, easily consolable cry after procedure.
Frequently awake		
1 Single arm[47]	56	26.8% were reported to be frequently awake.
Heart rate		
1 Single arm[47]	56	83.9% heart rate increase <20% after procedure. Heart rate return to baseline after procedure, n (%): 9 (16.1)
Need for repeat procedure		
1 Single arm[74]	146	4.6% had a second lingual frenotomy within 1 month.
Pain (C.R.I.E.S. Scale)[a]		
1 Single arm[47]	56	Pain intensity raised significantly during procedure, mean difference = 5 points; $p<0.001$ C.R.I.E.S. score after procedure, mean (SD): 4.4 (1.1) C.R.I.E.S. score 30 minutes after procedure, mean (SD): 0.7 (0.8)
Refusal of pacifier		
1 Single arm[47]	56	69.9% refused pacifier at 7-day follow-up.

[a] C.R.I.E.S. includes 5 domains related to distress or pain in infants: crying, requires oxygen to reach saturation over 95%, increased vital sings, facial expression, and sleeplessness. Each domain is rated on a scale from 0 to 2, with 2 indicating higher pain intensity and a total score ranging from 0 to 10.

Abbreviations: C.R.I.E.S. = Crying, Requires increased oxygen administration, Increased vital signs, Expression, Sleeplessness; N/n = number; SD = standard deviation.

3.3.2.2 Tongue-Tie and/or Other Tie Types (Specified)

Five single-arm studies[51,55,56,68,74] reported on harms of frenotomy with laser for tongue-tie and/or other tie types (e.g., lip-tie, buccal) (***Table 22***).

Table 22. Summary of harms from single-arm studies of frenotomy with laser for tongue-tie and/or other tie types (specified)

№ of Studies	N at Follow-up	Summary of Findings
Explicitly reported that no complications occurred		
2 Single arm[51,55]	157	No complications were reported following procedure.
Crying		
1 Single arm[56]	25	56% participants were crying and 44% were not crying after the procedure.
Pain		
1 Single arm[68]	22	82% reported local pain.
Reoccurrence/reattachment		
1 Single arm[68]	22	9% reported recurrence of lip-tie.
Temporary hypergranulation of wound tissue		
1 Single arm[74]	146	0.7% reported temporary hypergranulation of wound tissue.

Abbreviations: N = number.

3.3.3 Frenotomy with Unspecified Methods

3.3.3.1 Tongue-Tie Only

Three RCTs[28,29,31] reported on harms of frenotomy with unspecified methods for tongue-tie only (***Table 23***Table 23). One study (n=25) reported that there were no significant side effects but reported minimal bleeding (e.g., 1 or 2 drops) and crying that lasted only a few seconds in all participants, events that were classified as harms and complications by other studies.[28] In an RCT of 169 participants, there was a single case of bleeding; a single case of salivary duct damage; and a single case of cut to tongue and salivary duct damage based on mother and clinician report.[31] In a third study (n=107), it was reported that 2.5% of participants needed a repeat procedure.[29]

Table 23. Summary of harms comparing frenotomy with unspecified methods to no frenotomy (i.e., sham, breastfeeding support, or delayed frenotomy) for tongue-tie only

№ of Studies	N at Follow-up	Summary of Findings
Bleeding		
2 RCTs[28,31]	194	0.6% to 100% reported bleeding.
Crying		
1 RCT[28]	25	100% infant crying lasting a few seconds following procedure.
Salivary duct damage		
1 RCT[31]	169	0.6% reported salivary duct damage following procedure.
Accidental cut to tongue and salivary		
1 RCT[31]	169	0.6% reported an accidental cut to tongue and salivary duct damage following procedure.

№ of Studies	N at Follow-up	Summary of Findings
Complications		
1 RCT[31]	169	1.8% frenotomies with complications.
Need for repeat procedure		
1 RCT[29]	107	2.5% of participants needed a repeat procedure.

Abbreviations: N = number; RCT = randomized, controlled trial.

Three single-arm studies[44,46,49] reported on harms of unspecified methods for frenotomy for tongue-tie only (***Table 24***Table 24).

Table 24. Summary of harms from single-arm studies of frenotomy with unspecified methods for tongue-tie only

№ of Studies	N at Follow-up	Summary of Findings
Explicitly reported that no complications occurred		
2 Single arm[44,49]	145	No complications were reported following procedure.
Need for repeat procedure		
1 Single arm[46]	158	4% of participants needed a repeat procedure.

Abbreviations: N = number.

3.3.3.2 Tongue-Tie and/or Other Tie Types (Specified and Unspecified)

Two single-arm studies[54,57] reported on harms of frenotomy with unspecified release methods for tongue-tie and/or other tie types (e.g., lip-tie, buccal tie, 2 or more unspecified types) (***Table*** 25***25***).

Table 25. Summary of harms from single-arm studies of frenotomy with unspecified methods for tongue-tie and/or other tie types (specified and unspecified)

№ of Studies	N at Follow-up	Summary of Findings
Explicitly reported that no complications occurred		
1 Single arm[57]	84	No complications were reported by 99% of the sample (1 unsure).
Apnea, ALTE/BRUE, or other breathing difficulties		
1 Single arm[54]	16	25% reported apnea, ALTE/BRUE, or other breathing difficulties.
Bleeding		
1 Single arm[54]	16	19% reported bleeding.
Feeding		
1 Single arm[54]	16	44% reported poor feeding.
Grayish black stools		
1 Single arm[54]	16	6% reported grayish black stools.
Hypernatremia, hypothermia and 20% weight loss		
1 Single arm[54]	16	6% reported severe hypernatremia, hypothermia, and 20% weight loss.
Pain		
1 Single arm[54]	16	19% reported pain.
Pallor/anemia		
1 Single arm[54]	16	13% reported pallor/anemia.
Scarring		
1 Single arm[54]	16	13% reported excess scarring.
Ulcer		
1 Single arm[54]	16	6% reported ulcer.
Unsettledness		
1 Single arm[54]	16	6% reported unsettledness.

№ of Studies	N at Follow-up	Summary of Findings
Weight loss		
1 Single arm[54]	16	19% reported weight loss.

Abbreviations: ALTE/BRUE = Apparently Life-Threatening Event/Brief Resolved Unexplained Event; N = number.

3.3.3.3 Unspecified Ties

One single-arm study[60] reported on harms of frenotomy with an unspecified method for unspecified tie types (***Table 26***).

Table 26. Summary of harms from single-arm studies of frenotomy with unspecified methods for unspecified ties

№ of Studies	N at Follow-up	Summary of Findings
Unplanned visits		
1 Single arm[60]	414	27% reported unplanned visits[a] after the procedure (total of 132).
Bleeding		
1 Single arm[60]	414	0.2% reported bleeding.
Need for repeat procedure		
1 Single arm[60]	414	23% had a repeat frenulotomy performed and 3.1% had more than 2 frenulotomies performed.

[a] Reasons for visit included infection concerns (1.0%), bleeding (0.2%), continued poor feeding (0.4%), continued nipple pain (0.4%), and concern that tongue-tie persisted (29.5%).

Abbreviations: N = number.

3.4 Findings: Cost-Effectiveness

No studies that met criteria for the cost-effectiveness question were identified.

3.5 Clinical Practice Guideline Synthesis

We synthesized Clinical Practice Guidelines (CPGs) to review the guidance that different organizations have provided on frenotomy and frenectomy when applied for breastfeeding support. We searched for relevant CPGs and attempted to appraise each guideline using the Appraisal of Guidelines for Research & Evaluation II (AGREE II) instrument. The authors of these documents characterized them as clinical consensus statements,[16] management policies,[83] position statements,[85,106] scopes of practice,[86] reviews,[107] and clinical reports with recommendations.[84]

We identified 7 clinical practice guidelines related to frenotomy and frenectomy from organizations including the American Academy of Otolaryngology—Head and Neck Surgery, the American Academy of Pediatric Dentistry, American Academy of Pediatrics, The Academy of Breastfeeding Medicine, International Board of Lactation Consultant Examiners, the Canadian Paediatric Society, and the Canadian Agency for Drugs and Technologies in Health. None can be characterized as guidelines that explicitly relied on systematic reviews. ***Table 27*** summarizes references from relevant professional and clinical organizations related to frenotomy and frenectomy when applied for breastfeeding support.

Table 27. Clinical practice statements relating to frenotomy and frenectomy with breastfeeding support

Title/Organization Guideline Quality[a]	Year Published	Excerpts of Findings	Rating/Quality of Evidence Narrative Assessment Used
American Academy of Otolaryngology—Head and Neck Surgery[16] Quality rating: NA	2020	**For frenotomy:** A survey of expert pediatric otolaryngologists agreed that frenotomy in infants with ankyloglossia can lead to an improvement in breastfeeding, not all infants with ankyloglossia need a frenotomy, and there are more common conditions that may impede breastfeeding. The academy recommends further study to refine evidence.	Based on 2 systematic reviews. Quality of evidence assessment not performed.
American Academy of Pediatric Dentistry[83] Quality rating: NA	2022	**For surgical interventions on the frenulum:** Recognizes that difficulties with breastfeeding may have another cause and not all infants with ankyloglossia require surgical intervention. Recommends a team-based approach to treatment planning. The academy supports further research in the causative association between ankyloglossia and difficulties in breastfeeding.	Based on a nonsystematic review of the literature. Quality of evidence assessment not performed.
American Academy of Pediatrics[84] Quality rating: NA	2024	**For frenotomy:** It is unclear if release of a tight lingual frenulum in neonates improves breastfeeding. Because symptoms of ankyloglossia overlap those of other breastfeeding difficulties, a team partnership is necessary.	Based on a nonsystematic review of the literature. Quality of evidence assessment not performed.

Title/Organization Guideline Quality[a]	Year Published	Excerpts of Findings	Rating/Quality of Evidence Narrative Assessment Used
		Frenotomy may decrease maternal nipple pain. Further research is necessary.	
The Academy of Breastfeeding Medicine[85] Quality rating: NA	2021	**For surgical tongue-tie release:** If there is the presence of a restrictive sublingual frenulum, frenotomy can be an effective way to increase maternal comfort and milk transfer and may prevent premature breastfeeding cessation. The academy recommends more research on clear definitions of "tongue-tie," optimal surgical methods, and long-term outcomes.	Based on a nonsystematic review of the literature. Quality of evidence assessment not performed.
International Board of Lactation Consultant Examiners[86] Quality rating: NA	2017	Members of the International Board of Lactation Consultant Examiners should not diagnose tongue-tie but may refer parents to a clinician who can diagnose.	Overview of International Board of Lactation Consultant Examiners scope of practice, clinical competencies, code of conduct, and advisory opinions. Quality of evidence assessment not performed.
Canadian Paediatric Society[106] Quality rating: NA	2015; Reaffirmed 2024	**For frenotomy:** Does not recommend for all infants with ankyloglossia. Infants who experience significant breastfeeding difficulties may benefit from frenotomy. Frenotomy should be performed by a clinician "experienced with the procedure and using appropriate analgesia."	Based on a nonsystematic review of the literature. Quality of evidence assessment not performed.
Canadian Agency for Drugs and Technologies in Health[107] Quality rating: NA	2016	**For frenectomy:** Frenectomy is a safe procedure with demonstrated short-term breastfeeding effectiveness as perceived by the mother. There is less evidence on objective and long-term breastfeeding measurements.	Based on a nonsystematic limited literature search. Critical appraisals of the included studies were performed.

[a] We attempted to apply the AGREE guideline appraisal instrument to these documents. The instrument focuses on evaluating the process through which guidelines are developed, which did not apply to the included documents. The tool does not assess how well the evidence included in each guideline is evaluated, interpreted, or whether the conclusions were consistent with the evidence. Due to these concerns a quality rating of NA is reported.

Abbreviations: NA = not applicable.

4. Discussion

4.1 Summary of the Evidence

As depicted in ***Figure 3***, the level of certainty across all outcomes was low or very low.

Figure 3. Evidence map—frenotomy and frenectomy with breastfeeding support

Outcome category	Specific outcome	Favors frenotomy	No difference	Favors control	Cannot determine a difference
Maternal outcomes	Breastfeeding pain				6 RCTs, N=452
	Breastfeeding effectiveness				1 RCT, N=105

	Breastfeeding self-efficacy	3 RCTs, N=312			
	Any breastfeeding at ≤2 months				1 RCT, N=105 1 NRSI, N=159
	Any breastfeeding at >2 months		1 RCT, N=163		4 NRSIs, N=471
	Exclusive breastfeeding at ≤2 months		1 NRSI, N=159		2 RCTs, N=265
	Exclusive breastfeeding at >2 months				1 RCT, N=163 3 NRSIs, N=380
	Improvement in breastfeeding				2 RCTs, N=114
	Breastfeeding problems				1 NRSI, N=33
	Cessation of breastfeeding				1 NRSI, N=91
Infant outcomes	Infant weight gain				1 RCT, N=163
	Infant breastfeeding assessment				2 RCTs, N=165
	Gastroesophageal symptoms				1 RCT, N=48
Safety	Complications				1 RCT, N=163

[a] We did not grade the safety evidence from 47 single-arm studies, 6 RCTs, and 4 NRSIs.

Abbreviations: N = number; NRSI = nonrandomized study of interventions; RCT = randomized controlled trial.

Table legend

	High certainty of evidence
	Moderate certainty of evidence
	Low certainty of evidence
	Very low certainty of evidence

The level of certainty across all graded evidence examining the effectiveness and comparative effectiveness of frenotomy for tongue-tie only was rated as low or very low COE. Among infants with tongue-tie, frenotomy was associated with improvements in breastfeeding self-efficacy (low certainty of evidence). No differences were found between frenotomy and control for exclusive breastfeeding at 2 months or less in cohort studies, or any breastfeeding at greater than two months in RCTs (low certainty of evidence). For maternal breastfeeding and dyad outcomes of breastfeeding pain, breastfeeding effectiveness, any breastfeeding at 2 months or less, exclusive breastfeeding at more than 2 months, improvement in breastfeeding, breastfeeding problems, and cessation of breastfeeding, we could not determine the direction of effect (very low COE). For infant breastfeeding outcomes including infant weight gain, infant breastfeeding assessment

scores (measure of infant feeding behaviors), and infant gastroesophageal symptoms, we could not determine the direction of effect (very low COE).

The inconclusive evidence for the benefits of frenotomy on breastfeeding outcomes arises from study design limitations. Ethical and pragmatic considerations around the widespread belief among participant and some clinicians that frenotomy will be helpful ("lack of equipoise")[31] meant that most RCTs offered frenotomy to the control arm, either immediately or after a delay. The only RCT without planned frenotomy in the control arm reported very high rates of unplanned crossover;[31] even trials with planned delayed crossover (at 5 days) reported early crossover.[29] The expectation of frenotomy in the control may have influenced patient-reported outcomes. The expectation of frenotomy in the control may have influenced patient-reported outcomes. Sham RCTs[26-28] attempted to address this issue by blinding participants in both arms during the procedure but could only provide blinded outcomes immediately after the procedure. Authors of 1 study noted that they "do not foresee a way to prevent mothers from looking in their infants' mouths."[27] Additionally, blinding was not always successful during the sham procedure. All these considerations limited the utility of trial evidence. Nonrandomized, retrospective cohort studies did not have these specific design issues, but the potential for confounding also seriously limited their utility. Beyond these design considerations, there is also an unclear understanding of the typical time to achieve "good" breastfeeding, and thus the appropriate time to consider surgical or other intervention for breastfeeding problems may vary. Other reasons for lack of certainty are related to small underpowered studies lacking precision and the use of unvalidated measures.

We identified no evidence that examined the effectiveness or comparative effectiveness of frenotomy for tongue-tie with concomitant lip-tie or lip-tie alone.

Various harms were reported for frenotomy using scissors, lasers, and unspecified methods across examined oral tie types.

The level of severity for reported harms ranged from studies reporting no harms or adverse events, minor harms including bleeding or crying, and more serious harms including accidental cut to the tongue and salivary duct damage. Only 1 study provided comparative data on harms of frenotomy for tongue-tie and reported no differences in the incidence of complications between frenotomy and breastfeeding support (very low certainty of evidence). Other included comparative safety studies reported no harms from frenotomy or only overall study complication rates (i.e., across all study groups). All other reported safety data was from single-arm studies, which were not designed to provide causal inference or provide clear associations between frenotomy and harms.

No evidence was captured that examined the cost-effectiveness or cost-utility of frenotomy for tongue-tie with or without lip-tie or lip-tie alone.

4.2 Limitations of the Evidence Base

The evidence we identified for inclusion in this HTA has several limitations.

The limitations of the efficacy studies included small sample sizes, the inability to maintain randomization and concealment, and poor outcome measurement. Almost all of the included efficacy studies were underpowered and were limited in their ability to maintain blinding and group allocation due to significant crossover of study participants between study arms. Follow-up times for included studies were short due to the small window of time for mother and infant dyads to achieve breastfeeding efficacy. In head-to-head longer-term studies, it was difficult to determine the level of exposure to other interventions that could impact outcomes (i.e., interaction with lactation consultants or other breastfeeding assistance).

For safety studies, study limitations included small sample size, poor study design, and poor outcome measurement. The majority of safety studies were single-arm studies or comparative studies that only provided overall harms data, but not by group. The majority of safety studies also lacked detailed measurement information and lacked consistency in how harms and complications were classified across studies.

4.4 Selected Payer Coverage Policies

We conducted a scan of commercial payer coverage documents for frenotomy and frenectomy; overviews are provided in ***Table 28***Table 28 and ***Table 29***Table 29. Three payers had coverage policies for labial frenotomy/frenulotomy, labial frenoplasty/frenuloplasty, lingual frenoplasty/frenuloplasty, labial frenectomy/frenulectomy, and lingual frenectomy/frenulectomy.[88-90] The clinical criteria for coverage varied across payers and procedures ***(Table 29***). One policy covered buccal/labial frenectomy, lingual frenectomy, and frenuloplasty without any specific requirements,[89] and 1 policy covered anesthesia and facility services related to frenulectomy for congenital ankyloglossia when performed by oral surgeons or by a DDS, DMD, MD, or DO but did not cover the frenulectomy procedure itself.[88] Two policies required the procedure to be medically necessary to address newborn feeding difficulties,[90,108] and 1 was specific to only being medically necessary.[87]

Table 28. Overview of payer coverage policies for frenotomy and frenectomy with breastfeeding support

Condition	Washington Apple Health (Medicaid)[87]	Cigna	Kaiser Permanente	Premera Blue Cross[88]	Regence BlueShield[89]	UnitedHealth[90]
Labial frenotomy/ frenulotomy	—	—	—	—	—	—
Lingual frenotomy /frenulotomy	—	—	—	—	—	—
Labial frenoplasty/ frenuloplasty	✓	—	—	—	✓	✓
Lingual frenoplasty/ frenuloplasty	✓	—	—	—	✓	—
Labial frenectomy/ frenulectomy	✓	—	—	✓	✓	✓
Lingual frenectomy/ frenulectomy	✓	—	—	✓	✓	—

Notes: ✓ = covered; X = not covered; — = no policy identified.

Table 29. Payer coverage policies for frenotomy and frenectomy with breastfeeding support

Payer (Effective Date)	Coverage Policy
Aetna (1996, reviewed 2024)[108]	**Covered** Lingual or labial frenectomy, frenotomy, or frenuloplasty medically necessary for newborn feeding difficulties or childhood articulation problems **Not covered (Experimental)** Prophylactic frenectomy, frenotomy, or frenuloplasty to promote speech development Lingual frenuloplasty with myofunctional therapy for treatment of dental clenching, mouth breathing, myofascial tension, or snoring Oro-myofunctional therapy following frenectomy
Premera Blue Cross (2024)[88]	**Covered** Anesthesia and facility services related to frenulectomy for congenital ankyloglossia (tongue-tie only) when performed by oral surgeons or by a DDS, DMD, MD, or DO The procedure itself is not mentioned as covered or not covered
Regence Blue Shield (2024)[89]	**Covered** • Buccal/labial frenectomy • Lingual frenectomy • Frenuloplasty
UnitedHealthcare Dental (2023)[90]	**Covered** Frenulectomy or frenuloplasty for ankyloglossia or papillary penetrating attachment of maxillary labial frenum in newborns when there is interference with feeding
Washington Apple Health (2024)[87]	**Covered** Frenuloplasty/frenulectomy for clients aged 6 and younger with documented medical necessity do not require prior authorization; includes buccal/labial and lingual frenectomy

4.5 Limitations of This HTA

This HTA has several limitations related to the scoping and the processes we used to conduct the HTA. We limited the scope to English-language publications conducted only in countries rated as "very high" on the 2023/2024 UN Human Development Index. Also, the literature search was limited to 2 databases and studies published from database inception through August 2024. We did not seek unpublished data and did not use data presented only in conference abstracts. We did not consider efficacy outcomes from uncontrolled studies, and we did not use GRADE to evaluate the body of evidence consisting of uncontrolled studies for safety outcomes.

4.6 Ongoing Research and Future Research Needs

We identified 1 relevant clinical trial registered in clinicaltrials.gov (***Table 30***).[91] The crossover RCT consisted of group 1 receiving sham frenotomy followed by a lingual frenotomy, and group 2 receiving a lingual frenotomy followed by a sham frenotomy. Newborns who continued having feeding difficulties received a third intervention. May 2018 was listed as the trials completion date, but no published results are currently available. The trial record was last updated June 12, 2019.

Table 30. Summary of ongoing frenotomy and frenectomy with breastfeeding support studies

Registration Number	Sponsor	Description	Number of Participants	Status	Estimated Completion Date
NCT02141243[91]	University of South Florida	Infants at Tampa General Hospital with Class III or IV ankyloglossia (as identified via the HATLFF) were randomly assigned to 2 groups. Group 1 received a sham frenotomy followed by an actual lingual frenotomy procedure. Group 2 received an actual lingual frenotomy followed by a sham procedure. Infants with continued feeding difficulties will undergo a labial frenotomy. The goal of the studies is to determine when lingual frenotomies, labial frenotomies, or both are required to improve outcomes. Maternal pain will be measured using the Wong-Baker FACES Pain Rating Scale and a change in LATCH score.	120	Completed	2018-05

Abbreviations: FACES = Facial Action Coding System; HATLFF = Hazelbaker Assessment Tool or Lingual Frenulum Function; LATCH = Latch, Audible swallowing, Type of nipple, Conform, Hold.

Future efficacy, safety, and cost-effectiveness studies should seek to address the limitations of the current evidence base. Due to the ethical concerns of randomizing mother and infant dyads to intervention versus control, the difficulty of blinding, and lack of equipoise because most mothers likely believe that frenotomy will be helpful as evidence by high crossover rates, future studies should focus on comparing frenotomy methods (i.e., scissors vs. lasers), timing (e.g., frenotomy performed soon after birth vs. later) including assessing improvement in breastfeeding over time absent frenotomy, and whether the benefits of frenotomy vary based on contextual factors such as the availability of intensive and comprehensive breastfeeding support. Future studies should attempt to report on longer-term outcomes and should consider using large health care system data in an effort to include larger sample sizes. For efficacy, future studies should look to explore the impact of frenotomy on the outcomes of nipple excoriations, nipple infections (mastitis), aerophagia, swallowing function, failure to thrive, milk transfer, low milk supply, and other feeding issues that appear to be absent in the literature based on the current review's inclusion criteria. For harms specifically, future studies should use more intentional approaches for collecting and reporting harms data, including using predetermined and well-defined measures and collecting data from various sources (e.g., parents, provider, medical records).

5. Conclusion

We identified methodologically limited evidence for evaluating the efficacy and safety of frenotomy for breastfeeding support in infants up to 1 year of age with tongue-tie and/or lip-tie and no evidence reporting on cost-effectiveness.

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Appendix A. Search Strategy

PubMed Searches (August 30, 2024)

#	Query	Filters	Results
1	("Mouth Abnormalities"[Mesh:noexp] OR "Tongue Diseases/congenital"[Mesh:noexp] OR "Tongue/abnormalities"[Mesh] OR "Lingual Frenum"[Mesh] OR "Lip Diseases/congenital"[Mesh:noexp] OR "Lip/abnormalities"[Mesh] OR "Labial Frenum"[Mesh] OR "Ankyloglossia"[Mesh] OR "ankyloglossia"[tiab] OR (("tongue"[tiab] OR "lip"[tiab] OR "lingual"[tiab] OR "linguae"[tiab] OR "labial"[tiab] OR "maxillary"[tiab]) AND ("frenum"[tiab] OR "fraenum"[tiab] OR "frenulum"[tiab] OR "frena"[tiab] OR "frenula"[tiab])) OR (("tongue"[tiab] OR "lip"[tiab] OR "maxillary"[tiab]) AND ("tie"[tiab] OR "tied"[tiab])))		4,697
2	"Oral Surgical Procedures"[Mesh] OR "surgical"[tiab] OR "surgery"[Subheading] OR "surgery"[tiab] OR "frenulotomy"[tiab] OR "frenulectomy"[tiab] OR "frenotomy"[tiab] OR "frenectomy"[tiab] OR "frenuloplasty"[tiab] OR "z-plasty"[tiab] OR "h-plasty"[tiab] OR "laser"[tiab]		3,935,364
3	#1 AND #2		1,903
4	((child OR children OR childhood OR pediatric* OR paediatric* OR infant* OR baby OR newborn* OR babies OR neonate*))		4,392,867
5	#3 AND #4		1,115

CDSR Searches (August 30, 2024)

#1 MeSH descriptor: [Ankyloglossia] explode all trees 23 (1 Systematic review, 22 RCTs)

Appendix B. Excluded Articles

List of Exclusion Codes

X1: Not original research

X2: Ineligible population

X3: Ineligible intervention

X4: Ineligible or no comparator

X5: Ineligible or no outcome

X6: Ineligible study design

X7: Wrong Timing

X8: Ineligible setting

X9: Meets criteria but ineligible country

X10: Non-English publication

X11: Abstract only

X12: Irretrievable

X13: Data uninterpretable

1. Araujo M, Freitas RL, Lima MGS, et al. Evaluation of the lingual frenulum in newborns using two protocols and its association with breastfeeding. *J Pediatr (Rio J)*. 2020 May-Jun;96(3):379-85. doi: 10.1016/j.jped.2018.12.013. PMID: 31029684. Exclusion Code: X9.
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4. Baxter RT, Zaghi S, Lashley AP. Safety and efficacy of maxillary labial frenectomy in children: A retrospective comparative cohort study. *Int Orthod*. 2022 Jun;20(2):100630. doi: 10.1016/j.ortho.2022.100630. PMID: 35283058. Exclusion Code: X2.
5. Bettinsoli AR. [Treatment of lingual frenulum]. *Arch Argent Pediatr*. 2015 Apr;113(2):e136. PMID: 25915970. Exclusion Code: X10.
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9. Caloway C, Hersh CJ, Baars R, et al. Association of Feeding Evaluation With Frenotomy Rates in Infants With Breastfeeding Difficulties. *JAMA Otolaryngol Head Neck Surg*. 2019 Sep 1;145(9):817-22. doi: 10.1001/jamaoto.2019.1696. PMID: 31294774. Exclusion Code: X6.
10. Carríon Zabaraín E, Ornelas F. [Modification of the Z-plasty technic for short lingual frenum]. *Adm (1986)*. 1987 Jan-Feb;44(1):15-8. PMID: 3483581. Exclusion Code: X10.
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Appendix C. Evidence Tables

Table C-1. Included EQ studies sample characteristics

Author, Year Design Registration ROB Sponsorship	Setting	Criteria	Mean Age (SD)	Gender	Race/ Ethnicity	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method	Other
Berry, 2012[26] RCT ISRCTN24575506 High for breastfeeding improvement Low for other outcomes NR	Mothers and babies were recruited from referrals to one author (U.K.)	Inclusion: Infant <4 months old, symptoms of a breastfeeding problem, and presence of tongue-tie Exclusion: Bottle fed or infant would not feed	Age at randomization, mean IG: 33 days (range: 6–115 days) CG: 28 days (range: 5–111 days)	IG Male: 21 Female: 9 CG Male: 19 Female: 11	NR	Difficulty with latch, N (%) IG: 23 (77) CG: 24 (80) All: 47 (78) Nipple pain/trauma, N (%) IG: 20 (67) CG: 19 (63) All: 39 (65) Inefficient feeding, N (%) IG: 19 (63) CG: 18 (60) All: 37 (62) All 3 indications, N (%) IG: 10 (33) CG: 9 (30) All: 19 (32)	Tongue-tie: 60 (100) NR	Breastfed: 100%. NR if mixed breast and bottle feeding was permissible	NR
Buryk, 2011[27] RCT NCT00967915 Low Presumably the U.S. military: "Dr Buryk is a military	Naval Medical Center Portsmouth, VA, U.S., a military medical center with ~350 newborn	Inclusion: Infants with significant ankyloglossia as detected by certified lactation consultants, according to HATLFF, with	Age, days Frenotomy: 6.2 (6.9) Sham: 6.0 (7.0)	Frenotomy Girls: 11 Boys: 19 Sham Girls: 9 Boys: 19	NR	Short-Form McGill Pain Questionnaire (SF-MPQ), Mean (SD) IG: 16.8 (10.6) CG: 19.2 (9.9) p=0.36 Hazelbaker Assessment Tool	Tongue-tie: 100% per inclusion criteria Presence of other ties NR	NR	NR

Author, Year Design Registration ROB Sponsorship	Setting	Criteria	Mean Age (SD)	Gender	Race/ Ethnicity	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method	Other
service member; this work was prepared as part of her official duties."	deliveries a month	report of maternal nipple pain or difficulty breastfeeding Exclusion: Infants older than 30 days, craniofacial abnormalities, neurologically compromised infants, or other contraindica-tions to breastfeeding				for Lingual Frenulum (HATLFF) Appearance score, Mean (SD) IG: 6.0 (1.6) CG: 5.7 (2.2) *p*=0.63 HATLFF Function score, Mean (SD) IG: 9.4 (2.6) CG: 8.4 (2.0) *p*=0.08	NR		
Dixon, 2018[32] Cohort NR Serious Canterbury District Health Board	Outpatient clinic at Christchurch Women's Hospital in Christchurch, New Zealand	Inclusion: Audit 2016 (Study 1): BTAT score ≤5 for frenotomy Audit 2017 (Study 2): Within first 48 hours: severe feeding difficulties, medical review. Between 48 hours and 8	Median 2016 Audit Released: 4 days (IQR 1–31) Not released: 10.5 days (3–48) 2017 Audit Released: 13 days (IQR 5–61) Not released:	2016 Audit Released Male: 173 (65.5) Female: 91 (34.4) Not released Male: 21 (47) Female: 24 (53) 2017 Audit Released	NR	2016 Audit Released Poor latch: 160 (60.6) Nipple pain trauma: 149 (56.4) Constant feeding-poor milk transfer: 40 (15.2) Weight gain failure: 41 (15.5) Not released Poor latch: 21 (46.6) Nipple pain	Audit 2016 Tongue-tie: 264 (100) Audit 2017 Tongue-tie: 55 (100) Ankyloglossia type NR	2016 Audit Released Exclusive or fully breastfed: 103 (39.0) Bottle: 9 (3.4) Mixed: 126 (47.7) Not released Exclusive or fully breastfed: 16 (35.6) Bottle: 4	Family history of tongue-tie 2016 Audit Released: 8 (22.0) Unreleased: 9 (20.0) 2017 Audit Released: 23 (41.8) Unreleased: 2 (66.6)

Author, Year Design Registration ROB Sponsorship	Setting	Criteria	Mean Age (SD)	Gender	Race/ Ethnicity	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method	Other
		weeks: breastfeeding difficulties and assessment by a lactation consultant (or midwife with additional training), BTAT score ≤4 for frenotomy with a neonatologist; otorhinolaryngology surgeon would see infants older than 8 weeks following a breastfeeding assessment by a lactation consultant (or midwife with additional training) and a BTAT score ≤4, or infants with an anatomical abnormality Exclusion: NR	104 days (63–106)	Male: 34 (61.8) Female: 21 (38.2) Not released Male: 3 (100) Female: 0 (0)		trauma: 28 (62.2) Constant feeding-poor milk transfer: 15 (33.3) Tongue-tie type Released Coryllos type 1: 69 (26.1) Coryllos type 2: 118 (44.7) Coryllos type 3: 49 (18.6) Coryllos type 4: 1 (0.4) Not released Coryllos type 1: 1 (2.2) Coryllos type 2: 8 (17.8) Coryllos type 3: 11 (24.4) Coryllos type 4: 1 (2.2) 2017 Audit Released Poor latch: 31 (56.3) Nipple pain trauma: 32 (58.2) Constant feeding-		(8.89) Mixed: 19 (42.2) 2017 Audit Released Exclusive or fully breastfed: 27 (49.1) Bottle: 4 (7.3) Mixed: 24 (43.6) Not released Exclusively or fully breastfed: 2 (66.6) Bottle: 0 (0) Mixed: 1 (33.3)	

Author, Year Design Registration ROB Sponsorship	Setting	Criteria	Mean Age (SD)	Gender	Race/ Ethnicity	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method	Other
						poor milk transfer: 15 (27.3) Weight gain failure: 12 (21.8) Not released Poor latch: 2 (66.6) Nipple pain trauma: 2 (66.6) Constant feeding-poor milk transfer: 2 (66.6) Weight gain failure: 2 (66.6) Tongue-tie type Released Coryllos type 1: 16 (29.1) Coryllos type 2: 24 (43.6) Coryllos type 3: 2 (3.6) Coryllos type 4: 0 (0) Not released Coryllos type 1: 0 (0) Coryllos type 2: 1 (33.3) Coryllos type 3: 0 (0)			

Author, Year Design Registration ROB Sponsorship	Setting	Criteria	Mean Age (SD)	Gender	Race/ Ethnicity	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method	Other
						Coryllos type 4: 0 (0)			
Dollberg, 2006[28] RCT Low NR NR	Lis Maternity Hospital, Israel	Inclusion: Full-term healthy, appropriate-for-gestational age infants aged 1 to 21 days Exclusion: NR	Range 1 to 21 days	NR	NR	Ankyloglossia defined as "inability of the infant to protrude the tip of the tongue over the lower gum line while the tip was tied to the floor of the mouth by a tight cord of a frenulum, and the tongue became heart-shaped when lifted up" Pain or nipple trauma: 25 (100)	Ankylo-glossia: 25 (100) (tongue-tie) Anterior crease of the tongue: 15 (60)	NR	Family history of ankyloglossia in the first degree: 4 (16)
Emond, 2014[29] RCT ISRCTN 73554751 High National Institute for Health	Southmead Hospital in Bristol, U.K.; mothers were referred by their hospital or midwife to Southmead for examination by lactation consultants	Inclusion: Mothers with term babies with a tongue-tie experiencing breastfeeding difficulties (usually poor attachment and sore nipples). Eligible infants had a	NR[b]	NR[b]	NR[b]	"Breastfeeding difficulties"	Tongue-tie: 105 (100) NR	Bottle only By bottle IG: 1 (1.8%) CG: 0 (0%) By bottle and breast IG: 10 (18.2%) CG: 5 (9.6%) By breast only (exclusive) IG: 44 (80%)	

Author, Year Design Registration ROB Sponsorship	Setting	Criteria	Mean Age (SD)	Gender	Race/ Ethnicity	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method	Other
Research (NIHR)	for tongue-tie[a]	HATLFF-short form score of 6–12 (mild-moderate tongue-tie) and a LATCH score of ≤8 Exclusion: Infant < 2 weeks old, premature (<37 weeks), congenital orofacial malformations, infant weight loss >10% birth weight, and HATLFF score <6						CG: 47 (90.4%)	
Ghaheri, 2022[30] RCT NCT03793414 High Crowd-sourced on	Participants were enrolled "when scheduling appointments" at a private practice (Oregon, U.S.)	Inclusion: Infants 3 weeks to 4 months, ≥36 weeks gestational age, already partially bottle feeding, had surgical correction for posterior tongue-tie (at	Enrollment Age, days IG: 39.6 (25.2) CG: 47.4 (25.4)	Male IG: 13 (56.6) CG: 16 (66.7) Female IG: 10 (43.5) CG: 8 (33.3)[c]	White IG: 22 (95.7) CG: 23 (95.8) Asian IG: 0 (0) CG: 1 (4.2) Other IG: 1 (4.3)	Coryllos classification: type 3, N (%) IG: 15 (65.2) CG: 20 (83.3) Coryllos classification: type 4, N (%) IG: 8 (34.8) CG: 4 (16.7) Targeted	Tongue-tie: 48 (100) Posterior: 48 (100)	Breast and bottle fed: 48 (100) Infants were required to be both breast and bottle fed at enrollment as part of the inclusion criteria	NA

Author, Year Design Registration ROB Sponsorship	Setting	Criteria	Mean Age (SD)	Gender	Race/ Ethnicity	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method	Other
www.experiment.com		randomization or 10 days after) Exclusion: Comorbid diagnoses of the heart, lungs, or brain; multiple births; diagnoses that may impair breastfeeding (e.g., neurological conditions, cleft lip/palate, abnormalities of the throat/ esophagus); mother with insufficient glandular tissue; infants with anterior tongue-tie or lip-tie			CG: 0 (0) Ethnicity Hispanic/ Latino IG: 1 (4.3) CG: 0 (0) Non-Hispanic/ Latino IG: 22 (95.7) CG: 24 (100)	examination for palpably restricted frenulum with lingual elevation, limitation of lateral movement, abnormal floor of mouth elevation with tongue elevation, and renulum attachment location; sucking evaluations were performed, noting gum/lip grip, cupping/seal of the tongue against the finger, and the nature of the sucking tongue movements			
Guinot, 2022[33] Cohort NCT04703946	Hospital Senyora de Meritxell, Andorra	Inclusion: Patients born at the Hospital Nosra Senyora de Meritxell from January 2016 to	NR but frenotomy performed for consenting families within	N with ankylo-glossia Male: 95 (51.95) Female: 88 (48.05)	NR	In the intervention group only (n=136): Coryllos classification, N (%) Type 1: 13 (9.5)	Ankylo-glossia: 183 (100) (Tongue-tie)	NR	Family history of ankyloglossia: 41 (22.4)

Author, Year Design Registration ROB Sponsorship	Setting	Criteria	Mean Age (SD)	Gender	Race/ Ethnicity	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method	Other
Serious No external funding		December 2020 Exclusion: Neonates transferred for complex neonatal pathology or with missing medical records	maximum 7 days post-failure of trial of breastfeed-ing support measures in first 72 hours post-birth			Type 2: 114 (83.9) Type 3: 8 (5.9) Type 4: 1 (0.7)	NR		
Hogan, 2005[8] RCT NR High NR	Princess Anne Hospital, Southampto n, and Hythe, Romsey, and Lymington Birth Centres, U.K.	Inclusion: Infants with tongue-tie identified at delivery with feeding problems possibly due to tongue-tie Exclusion: NR	Age (at randomiza-tion), mean days IG: 20; median 14 CG: 18; median 15 Age (at randomiza-tion), mean days Breastfed IG+CG: 18 (median 19; range 3–51) Bottle fed IG+CG: 24 days (median 19	NR. Male: Female ratios: IG: 1:1 CG: 1.3:1	NR	Infants were evaluated at birth; those with feeding issues related to tongue-tie were re-examined to confirm issues and randomized For breastfed babies, N (%) (total n=40) Latching problems IG: 17 (85) CG: 16 (80) Sore nipples IG: 16 (80) CG: 16 (80) Continuous feeds IG: 9 (45)	Based on criteria, tongue-tie: 57 (100) NR	Breastfed: 40 (70.2) IG: 20 (50) CG: 20 (50) Artificially fed: 17 (29.8) IG: 8 (47.1) CG: 9 (52.9)	NR

Author, Year Design Registration ROB Sponsorship	Setting	Criteria	Mean Age (SD)	Gender	Race/ Ethnicity	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method	Other
			days, range 5–70) Mean age (days) at randomization, overall IG+CG: 20 days (median 15, range: 3–70) SD NR			CG: 12 (60) Top-up feeds IG: 6 (30) CG: 8 (40) For bottle-fed babies, N (%) (total n=17) Slow bottle feeds IG: 5 (62) CG: 8 (88) Dribbling IG: 5 (62) CG: 7 (77) Excess wind IG: 2 (25) CG: 2 (25)			
Knight, 2023[31] RCT ISRCTN 10268851 High National Institute for Health and Care Research	12 infant feeding services in England	Inclusion: Infants aged <10 weeks referred by parent or breastfeeding support service to an infant feeding difficulties and judged to have tongue-tie	Age at randomization <2 weeks IG: 30 (37.5) CG: 34 (38.2) ≥2 and <4 weeks IG: 24 (30.0) CG 24	Male IG: 51 (63.8) CG: 49 (55.1) Female IG: 29 (36.2) CG: 40 (44.9)	Infant race/ ethnicity NR Mother's ethnic group White, n (%) IG: 72 (92.3) CG: 84 (95.5)	Degree of tongue-tie (BTAT), n (%) 0–4 IG: 29 (36.3) CG: 26 (29.6) 5–6 IG: 21 (26.3) CG: 32 (36.4) 7–8 IG: 30 (37.5) CG: 30 (34.1)	NR (169 Tongue-tie) Anterior only IG: 11 (14.7) CG: 1 (1.8) Anterior and	Exclusive breastmilk feeding in the previous 24 hours, n (%) Yes IG: 51 (63.8) CG: 60 (67.4) No IG: 29 (36.2) CG: 29 (32.6)	Phototherapy for jaundice, n (%) Yes IG: 10 (12.5) CG: 13 (14.6) No IG: 70 (87.5) CG: 76 (85.4) NICU admission, n (%) IG: 9 (11.3) CG: 10 (11.2) 1–2 nights, n (%)

Author, Year Design Registration ROB Sponsorship	Setting	Criteria	Mean Age (SD)	Gender	Race/ Ethnicity	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method	Other
(NIHR) Health Technology Assessment Programme		Exclusion: Infant older than 10 weeks, born at <34 weeks gestation, had a congenital anomaly known to interfere with breastfeeding (e.g., cleft palate, Down syndrome), known bleeding diathesis, or had a frenotomy prior to recruitment	(27.0) ≥4 and <10 weeks IG: 26 (32.5) CG: 31 (34.8)		Asian, n (%) IG: 5 (6.4) CG: 2 (2.3) Black, n (%) IG: 0 (0.0) CG: 0 (0.0) Mixed, n (% IG: 1 (1.3) CG: 2 (2.3) Other, n (%) IG: 0 (0.0) CG: 0 (0.0) Missing, n IG: 2 CG: 1	Missing, n IG: 0 CG: 1 Maternal pain while feeding in previous 24 hours, median (IQR) IG: 4 (1-7) CG: 4 (2-7) Missing , n IG: 2 CG: 1	posterior IG: 54 (72.0) CG: 48 (87.3) Posterior only IG: 8 (10.7) CG: 6 (10.9) Data missing: IG=0; CG=10	Exclusive direct breastfeeding in the past 24 hours, n (%) Yes IG: 30 (37.5) CG: 37 (41.6) No, n (%) IG: 50 (62.5) CG: 52 (58.4) Use of infant formula, n (%) Yes IG: 28 (35.0) CG: 29 (32.6) No IG: 52 (65.0) CG: 60 (67.4)	IG: 4 (50.0) CG: 4 (40.0) 3–4 nights, n (%) IG: 2 (25.0) CG: 1 (10.0) >4 nights, n (%) IG: 2 (25.0) CG: 5 (50.0) Missing, n IG:1 CG: 0 Baby is 1 of a multiple pregnancy Yes IG: 1 (1.2) CG: 0 (0.0) No IG: 70 (87.5) CG: 76 (85.4) Previous live birth(s), n (%) Yes IG: 39 (48.7) CG: 42 (47.2) No IG: 41 (51.3) CG: 47 (52.8) Breastfed before

Author, Year Design Registration ROB Sponsorship	Setting	Criteria	Mean Age (SD)	Gender	Race/ Ethnicity	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method	Other
									Yes, n (%) IG: 33 (89.2) CG: 40 (95.2) No, n (%) IG: 4 (10.8) CG: 2 (4.8) NA—no previous live birth, n IG: 41 CG: 47 Missing, n IG: 2 CG: 0 Pretrial breastfeeding support received Yes, n (%) IG: 66 (84.6) CG: 74 (84.1) No, n (%) IG: 12 (15.4) CG: 14 (15.9) Missing, n IG: 2 CG: 1
Schlatter, 2019[34] Cohort	Tertiary maternity united affiliated to a	Inclusion: Newborns born at the center between September	NR	Among babies with tongue-tie (n=116) and ATLFF	NR	Among babies with tongue-tie (n=116) and ATLFF score <11, N (%) Breastfeeding	Tongue-tie: 116 (100) NR	NR	Among babies with tongue-tie (n=116) and ATLFF score <11, N (%) Family history of tongue-tie: 6/33 (18)

Author, Year Design Registration ROB Sponsorship	Setting	Criteria	Mean Age (SD)	Gender	Race/ Ethnicity	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method	Other
360/14, 16.10.2014 Serious NR	German University	2014 and June 2015 Exclusion: Born <35 weeks gestation, congenital oropharyngeal malformations, floppy infant syndrome, infants who needed intensive or intermediate care, those with congenital heart defects or sepsis, and primary ablactation		score <11, N (%), Male: 22/33 (68)		problems: 18/33 (55) Severe breastfeeding problems: 10/33 (29) An indication for frenulotomy was an ATLFF score of <11 and difficulties breastfeeding but was not the case in all infants who received one			
Sharma, 2015[35] Cohort NR Serious NR	Medical records of infants (U.K.)	Inclusion: Neonates and infants diagnosed with tongue-tie between June 2013 and July 2014 Exclusion: NR	38 days (range, 15–178 days) (not clear if this is age at frenotomy or phone survey)	Male: 23 (55) Female: 19 (45)	NR	Poor latch: 28 (67) Maternal nipple pain: 20 (48)	Tongue-tie: 42 (100) Other types of tie NR NR	NR	NR
Steehler, 2012[36]	Medical records were	Inclusion: Neonates and	Mean at time of	Males: 216 (58.9)	Caucasian: 258 (70.3)	Coryllos type 1: 64 (17.4)	Tongue-tie:	NR	Family history of ankyloglossia

Author, Year Design Registration ROB Sponsorship	Setting	Criteria	Mean Age (SD)	Gender	Race/ Ethnicity	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method	Other
Cohort NR Serious NR	consulted; the source of the records was NR; data from 2006–2011	infants suspected to have ankyloglossia Exclusion: NR	procedure: 18 days (SD NR)	Females: 151 (41.1)	African American: 57 (15.5) Hispanic: 16 (4.4) Multiethnic: 12 (3.3) Indian: 10 (2.7) Asian: 6 (1.6) Arabic: 4 (1.1) Persian: 2 (0.5) Filipino: 1 (0.3) Unknown: 1 (0.3)	Coryllos type 2: 167 (45.5% Coryllos type 3: 93 (25.3) Coryllos type 4: 18 (4.9) Insufficient data: 25 (6.8)	100%, based on proce-dure descript-tion Other types of tie NR NR		Yes: 127 (34.6) No: 194 (52.9) Unknown: 46 (12.5)

Notes: [a] This hospital has an established service for treatment of tongue-tie led by midwife lactation consultants and good support for breastfeeding in the hospital and the community midwifery service.
[b] Demographics between groups were "similar." Compared to the general profile of women delivering at the hospital, trial mothers had a higher education level and were less likely to have an ethnic minority background.
[C] Percentages calculated by Evidence-based Practice Center staff.

Abbreviations: ATLFF = Assessment Tool for Lingual Frenulum Function; BTAT = Bristol Tongue Assessment Tool; CG = control group; EQ = efficacy question; HATLFF = Hazelbaler Assessment Tool for Lingual Frenulum Function; IG = intervention group; IQR = interquartile range; LATCH = Latch, Audible swallowing, Type of nipple, Comfort, Hold; N = number; NA = not applicable; NR = not reported; RCT = randomized controlled trial; SD = standard deviation; U.K. = United Kingdom.

Table C-2. Included EQ Study Intervention Characteristics

Author, Year Design ROB	Treatment N (Follow-up N)	Setting	Treatment Professional	Intervention Anesthesia/analgesia/anesthetic	Other Therapy	Lactation Consult Involvement
Berry, 2012[26] RCT High for breastfeeding improvement Low for other outcomes	IG: Frenotomy with scissors 30 (27) CG: Sham frenotomy 30 (30)	Hospital	IG: NR; one of the authors. CG: NA	IG: NR CG: NA	IG: None CG: None	Both groups: Not specified but "parents were given written study information on arrival and had the usual consultation with either M.G. or C.W. [study authors] where the tongue-tie and feeding difficulties were confirmed."
Buryk, 2011[27] RCT Low	IG: Frenotomy with scissor 30 (30) CG: Sham frenotomy 28 (28)	ENT clinic	IG: ENT surgeon CG: NA	IG: NR CG: NA	IG: NR CG: NR	Both groups: Mothers who were noted to have nipple pain or difficulty breastfeeding were referred to certified lactation consultants. Lactation consultants routinely examined the infants' mouths as part of their assessment. Infants were enrolled if the lactation consultants detected significant ankyloglossia, according to the HATLFF.
Dixon, 2018[32] Cohort Serious	IG: Frenotomy with scissors, 2016 audit (Study 1): 264 released (164) 45 not released Frenotomy with scissors, 2017 audit (Study 2)	Hospital outpatient clinic	IG: NR CG: Otorhinolaryngology surgeon if infant was older than 8 weeks; other ages NR	IG: Sucrose or expressed breast milk used for analgesia CG: Sucrose or expressed breast milk used for analgesia	IG: No post-frenotomy bodywork or oral exercises were advised CG: No post-frenotomy bodywork or oral exercises were advised	IG: NR CG: Required infants between 48 hours and 8 weeks of age with breastfeeding difficulties to be assessed by a lactation consultant (or midwife with additional training).

Author, Year Design ROB	Treatment N (Follow-up N)	Setting	Treatment Professional	Intervention Anesthesia/analgesia/anesthetic	Other Therapy	Lactation Consult Involvement
	CG: 55 released (34) 3 not released					
Dollberg, 2006[28] RCT Low	IG: Frenotomy (method unclear), breastfeeding, sham, breastfeeding 14 (14) CG: Sham, breastfeeding, frenotomy, breastfeeding 11 (11)	Hospital	IG: Neonatologist or pediatric dentist CG: Neonatologist or pediatric dentist	IG: NR CG: NR	IG: NR CG: NR	Both groups: Data were obtained by lactation consultant. No mention of consultation.
Emond, 2014[29] RCT High	IG: Immediate frenotomy (method unclear) 55 (52) CG: Delayed frenotomy 52 (50)	Hospital	IG: NR CG: NA	IG: NR CG: NA	IG: Breastfeeding support from midwives CG: Breastfeeding support from midwives	Both groups: Mothers were referred to a lactation consultant by their hospital or midwives. Lactation consultants identified tongue-tie using the HATLFF-short form and LATCH scales. Lactation consultants were not mentioned as part of the intervention.
Ghaheri, 2022[30] RCT High	IG: Frenotomy with laser immediately after enrollment 24 (23)	Private practice	IG: NR. Possibly "lead author" CG: NR. Possibly "lead author"	IG: Topical anesthetic gel (3% lidocaine/3% tetracaine) CG: Topical anesthetic gel (3% lidocaine/3% tetracaine)	IG: None CG: Therapies before frenotomy varied	Both groups: Mandatory evaluation by lactation consultants before referral was a prerequisite for consultation. Latch assessment was considered in decision making to determine

Author, Year Design ROB	Treatment N (Follow-up N)	Setting	Treatment Professional	Intervention Anesthesia/analgesia/anesthetic	Other Therapy	Lactation Consult Involvement
	CG: Delayed treatment. Continuation of non-frenotomy therapies with offer of frenotomy with laser ~10 days after randomization 24 (24)					whether frenotomy was offered.
Guinot, 2022[33] Cohort Serious	IG: Received frenotomy with scissors 136 (136) CG: No frenotomy 47 (47)	NR	IG: NR CG: NR	IG: Surgical intervention was performed using 3 drops of a gluco-saline solution (0.9 mL sodium chloride and 5% glucose) applied topically in the oral cavity CG: NA	IG: Support was provided for breastfeeding by pediatricians and nurses CG: Support was provided for breastfeeding by pediatricians and nurses from the Maternal and Infant Unit of the hospital	Both groups: NR, but during the first 72 hours after birth, the Maternal and Infant Unit, made up of pediatricians and nurses, was responsible for reevaluating the morphological and functional aspect of the lingual frenulum, offering support measures and reinforcement of the breastfeeding technique (the existence or not of difficulties with breastfeeding was evaluated).
Hogan, 2005[8] RCT High	IG: Frenotomy with scissors 28 (28) CG: Intensive support, advice, and	NR	IG: NR. "The authors," at least 2 of whom were lactation consultants CG: Lactation consultant	IG: No anesthetic or analgesic was used CG: NA	Both groups: All the mothers in the study were monitored weekly for 4 weeks to assess feeding	IG: NR. Midwives and health visitors provided initial advice; lactation consultants were involved in study conduct. CG: Lactation consultants gave advice and help with positioning and attachment, along with a plan of care with

Author, Year Design ROB	Treatment N (Follow-up N)	Setting	Treatment Professional	Intervention Anesthesia/analgesia/anesthetic	Other Therapy	Lactation Consult Involvement
	help from lactation consultant 29 (29)					the mother. If symptoms did not alleviate after 48 hours, a frenotomy was offered.
Knight, 2023[31] RCT High	IG: Frenotomy (method unclear) with standard breastfeeding support 80 (78) CG: Breastfeeding support 89 (88)	Hospital	IG Midwife, N (%) IG: 61 (81.3) CG: 56 (90.3) Nurse, N (%) IG: 0 CG: 0 Doctor, N (%) 13 (17.3) 6 (9.7) Other, N (%) IG: 1 (1.3) CG: 0 Missing IG: 0 CG: 11 CG Midwife, N (%) IG: 61 (81.3) CG: 56 (90.3) Nurse, N (%) IG: 0 CG: 0 Doctor, N (%)	IG: NR CG: NA	Both groups: Breastfeeding support included at a minimum: Assessment, such as using the LATCH tool, advice on positioning and attachment, and at least one follow-up visit, as well as drop-in clinic advice as required (available more than 1 day a week); support was provided in person or virtually	IG: Pretrial breastfeeding support received Yes, n (%) IG: 66 (84.6) CG: 74 (84.1) No, n (%) IG: 12 (15.4) CG: 14 (15.9) Missing, n IG: 2 CG: 1 CG: Pretrial breastfeeding support received Yes, n (%) IG: 66 (84.6) CG: 74 (84.1) No, n (%) IG: 12 (15.4) CG: 14 (15.9) Missing, n IG: 2 CG: 1 For both groups, following referral to the infant feeding service, infant feeding was

Author, Year Design ROB	Treatment N (Follow-up N)	Setting	Treatment Professional	Intervention Anesthesia/analgesia/anesthetic	Other Therapy	Lactation Consult Involvement
			13 (17.3) 6 (9.7) Other, N (%) IG: 1 (1.3) CG: 0 Missing IG: 0 CG: 11			observed (either in person or via video conferencing), tongue assessment conducted, and mothers received advice on positioning and attachment. Initial discussions may have taken place in person or virtually via telephone or videoconferencing if this was what was being offered as part of routine care.
Schlatter, 2019[34] Cohort Serious	IG: Frenulotomy with scissors 30 (30) CG: Tongue-tie but no frenulotomy 10(10)	NR	IG: Maxillofacial surgeon CG: NR	IG: No anesthesia. 20% glucose was administered CG: NR	IG: NR CG: NR	Both groups: Support was provided by a lactation consultant if breastfeeding problems were reported.
Sharma, 2015[35] Cohort Serious	IG: Frenotomy with scissors 36 (36) CG: No frenotomy, infant feeding support 6 (6)	NR	IG: Surgeon CG: Infant feeding coordinator	IG: NR[a] CG: NA	IG: NR; possibly received support before frenotomy CG: Support from infant feeding coordinator	IG: NR CG: NR
Steehler, 2012[36] Cohort Serious	IG: Frenotomy with scissors 302 (82)	NR	IG: NR CG: NA	IG: Yes; topical viscous lidocaine is applied to the lingual frenulum CG: NA	IG: NR CG: NA	IG: NR CG: NA

Author, Year Design ROB	Treatment N (Follow-up N)	Setting	Treatment Professional	Intervention Anesthesia/analgesia/anesthetic	Other Therapy	Lactation Consult Involvement
	CG: No frenotomy 65 (9)					

Notes: [a] Note from authors on not using anesthetic is in discussion: "With our technique, we do not advocate the use of local anesthetic."

Abbreviations: CG = control group; ENT = ear, nose, and throat; EQ = efficacy question; HATLFF = Hazelbaker Assessment Tool for Lingual Frenulum Function; IG = intervention group; LATCH = Latch, Audible swallowing, Type of nipple, Comfort, Hold; N = number; NA = not applicable; NR = not reported; RCT = randomized controlled trial; ROB = risk of bias.

Table C-3. EQ outcome results

Author, Year Design ROB	Breastfeeding Outcomes	Related Outcomes	Comment
Berry, 2012[26] RCT High for breastfeeding improvement Low for all other outcomes	Pain, change in score from baseline to immediately after division, visual analog scale score, change (SD) (among 28 mothers with baseline pain) IG: −2.5 (1.9) (n=14) CG: −1.3 (1.5) (n=14) *p*=0.13 (95% CI, −0.3 to 2.4) Improved feeding, immediately after division, per protocol (IG=27; CG=30), N (%) IG: 21 (78) CG: 14 (47%) *p*<0.02 (95% CI, 6 to 51) Mothers noted better latch, reduced pain, baby sucking "differently," feeding feeling "more effective," and a less frantic, more relaxed feed	NA	Outcomes only reported on a per-protocol basis. Observer and mothers both also recorded if they thought the procedure had been completed.
Buryk, 2011[27] RCT Low	SF-MPQ, after intervention, ITT (IG=30; CG=28), mean score (SD) IG: 4.9 (1.46) CG: 13.5 (1.5) Effect size: 0.38 Length of breastfeeding No difference between groups; *p*=0.43 IBFAT score, after intervention, ITT (IG=30; CG=28), mean score (SD) IG: 11.6 (0.81) CG: 8.07 (0.86) Effect size: 0.31; *p*=0.029	NA	All but 1 infant in the sham group received a frenotomy by the 2-week follow-up. The authors continued to measure results until 12-month follow-up but only reported significant results from measurements taken during breastfeeding session immediately after procedure/sham procedure.
Dixon, 2018[32] Cohort	Feeding method at follow-up, N (%) 2016 audit (Study 1), released (N at follow-up=164) Exclusive or fully breastfed: 89 (54) Bottle: 37 (23) Mixed: 38 (23) 2016 audit (Study 1), not released (N at follow-up=22) Exclusive or fully breastfed: 10 (46) Bottle: 4 (18) Mixed: 8 (36) *P*=0.40	NA	NA

Author, Year Design ROB	Breastfeeding Outcomes	Related Outcomes	Comment
	2017 audit (Study 2), released (N at follow-up=34) Exclusively or fully breastfed: 19 (56) Bottle: 10 (29) Mixed: 5 (15) 2017 audit (Study 2), not released (N at follow-up=1) Exclusively or fully breastfed: 0 (0) Bottle: 0 (0) Mixed: 1 (100) *P* not calculated due to low N		
Dollberg, 2006[28] RCT Low	LATCH score, mean (SD), both groups pre- and post-real frenotomy score (n=25) Before frenotomy: 6.4 (2.3) After frenotomy: 6.8 (2.0) *p*=0.06 Pain, VAS, mean (SD), both groups pre- and post-real frenotomy score (n=25) Before frenotomy: 7.1 (1.9) After frenotomy: 5.3 (2.2) *p*=0.01	NA	
Emond, 2014[29] RCT High	Change in pain (VAS) from 0–5 days, (IG=53; CG=52), median (IQR) IG: −2 (−3 to 0.4) CG: −1 (13.5 to 1) *p*=0.09 Feeding method, 5 days, (IG=53; CG=52), N (%) 5 days By bottle IG: 5 (9.4%) CG: 8 (15.5%) By bottle and breast IG: 13 (24.5%) CG: 6 (11.5%) By breast only (exclusive) IG: 35 (66%)	NA	An additional qualitative survey was completed at 8 weeks but was not abstracted here as the outcomes were ineligible.

Author, Year Design ROB	Breastfeeding Outcomes	Related Outcomes	Comment
	CG: 38 (73%) OR (95% CI) (by bottle and by bottle and breast as reference): 1.40 (0.60 to 3.22); *p*=0.43 By breast at all IG: 48 (91%) CG: 44 (85%) OR (95% CI) (by bottle as reference group): 0.57 (0.17 to 1.88); p=0.35 Change in LATCH score from 0–5 days, (IG=53; CG=52), Median (IQR) 5 days IG: 1 (0–2) CG: 1 (0–2) *p*=0.52 Change in BSES score from 0–5 days, (IG=53; CG=52), median (IQR) IG: 9 (1.8 to 12.3) CG: 1 (−4 to 7.5) *p*=0.002 IBFAT Score, change from baseline, median (IQR) (Ns, IG=53; CG=52) 5 days IG: 0 (−1.8 to 1.0) CG: 0 (0‑1) *p*=0.36		
Ghaheri, 2022[30] RCT High	VAS for breastfeeding pain, day 10, mean (SD) IG: −2.3 [2.4] CG: −0.8 [1.4] BSES-SF, day 10, mean (SD) IG: 13.4 (10.1) CG: -1.0 (7.8) 95% CI: 9.2 to 19.7; *p*<0.001	GSQ-I results, between day 0 and day 10, mean (SD) Vomiting/regurgitation (times) IG: −4.0 (17.3) CG: 3.9 (8.7) 95% CI: −0.1 to 15.8, *p*=0.006 Vomiting/regurgitation (severity)	Measurements gathered by the infant feeding solution device are not reported in this table as the device is bottle based.

Author, Year Design ROB	Breastfeeding Outcomes	Related Outcomes	Comment
		IG: −1.1 (1.9) CG: 0.8 (1.9) 95% CI: 0.8 to 3.0; *p*=0.001 Irritability/fussiness (times) IG: −6.3 (18.2) CG: −0.8 (9.6) 95% CI: −3.0 to 14.0; *p*=0.08 Irritability/fussiness (severity) IG: −1.0 (2.1) CG: 0.5 (2.2) 95% CI: 0.2 to 2.7; *p*=0.01 Refusal to feed (times) IG: −1.5 (4.5) CG: 0.8 (3.6) 95% CI: −0.1 to 4.7; *p*=0.02 Refusal to feed (severity) IG: −0.7 (1.9) CG: 0.6 (1.9) 95% CI: 0.2 to 2.4; *p*=0.03 Choking/gagging (times) IG: −8.7 (13.9) CG: 1.1 (6.3) 95% CI: 3.5 to 16.1; *p*=0.001 Choking/gagging (severity) IG: −1.1 (1.7) CG: −0.3 (1.5) 95% CI: − −0.1 to 1.8; *p*=0.06 Arching back (times) IG: −7.2 (15.0) CG: 2.5 (10.3))	

Author, Year Design ROB	Breastfeeding Outcomes	Related Outcomes	Comment
		95% CI: 2.2 to 17.3; *p*=0.003 Arching back (severity) IG: −1.3 (1.7) CG: 0.1 (1.7) 95% CI: 0.4 to 2.4; *p*=0.01 Episodes of hiccups (times) IG: −4.6 (8.8) CG: −0.4 (7.1) 95% CI: −0.5 to 8.9; *p*=0.05 Episodes of hiccups (severity) IG: −1.2 (1.6) CG: −0.1 (1.1)) 95% CI: 0.3 to 1.9; *p*=0.01	
Guinot, 2022[33] Cohort Serious	Method of feeding, n (%) 1 month By exclusive bottle feeding IG: 6 (5.0) CG: 6 (15.39) Prevalence ratio (PR): 0.32 (95% CI, 0.11 to 0.95); *p*=0.03 By mixed feeding IG: 26 (21.7) CG: 3 (7.69) PR: 2.82 (95% CI, 0.90 to 8.80); *p*=0.05 By exclusive breastfeeding IG: 88 (73.3) CG: 30 (76.92) PR: 0.95 (95% CI, 0.78 to 1.17); *p*=0.656 3 months By exclusive bottle feeding IG: 8 (6.67)	NA	NA

Author, Year Design ROB	Breastfeeding Outcomes	Related Outcomes	Comment
	CG: 8 (20.51) PR: 0.32 (95% CI, 0.13 to 0.81); *p*=0.013 By mixed feeding IG: 31 (25.83) CG: 3 (7.69) PR: 3.36 (95% CI, 1.09 to 10.38); *p*=0.016 By exclusive breastfeeding IG: 81 (67.5) CG: 28 (71.8) PR: 0.94 (95% CI, 0.75 to 1.19); *p*=0.616 6 months By exclusive bottle feeding IG: 10 (8.33) CG: 8 (20.51) PR: 0.41 (95% CI, 0.17 to 0.96); *p*=0.037 By mixed feeding IG: 31 (25.83) CG: 3 (7.69) PR: 3.36 (95% CI, 1.09 to 10.38); *p*=0.016 By exclusive breastfeeding IG: 79 (65.84) CG: 28 (71.8) PR: 0.92 (95% CI, 0.72 to 1.16); *p*=0.491		
Hogan, 2005[8] RCT High	Breastfed for at least 4 months, N (%), (breastfeeding infants, IG=20; CG=20) IG: 12 (60) CG: NR; all but 1 CG infant had a division by 48 hours Improvement, timepoint NR (all babies) IG: 27 CG: 1	NA	

Author, Year Design ROB	Breastfeeding Outcomes	Related Outcomes	Comment
	Improvement, timepoint NR (breastfed babies, n=40) IG: 19 CG: 1 Improvement, timepoint NR (bottle-fed babies, n=17) IG: 8 CG: 0 All mothers in CG who did not experience improvement requested frenotomy at 48 hours		
Knight, 2023[31] RCT High	Mother's pain while feeding during previous 24 hours, SF-MPQ, median (IQR) 1–2 weeks postrandomization IG: 2 (0–4) CG: 2 (0–4) Missing or not breastfeeding: IG=4; CG=3 RR: 0.0 (95% CI: −0.9 to 0.9); aRR: 0.0 (95% CI, −0.9 to 0.9); *p*=0.99 3 months of age IG: 0 (0-1) CG: 0 (0-2) Missing or not breastfeeding: IG=19; CG=26 EE: 0 (−0.5 to 0.5); aEE: −0.2 (−0.6 to 0.3); *p*=0.45 Any breastmilk feeding at 3 months, N (%) IG: 67/80 (88.2) CG: 75/89 (86.2) Missing: IG=4; CG=2 RR: 1.02 (95% CI, 0.91 to 1.15); aRR: 1.02 (95% CI, 0.90 to 1.16); *p*=0.73 Any breastmilk feeding at 3 months, per protocol analysis IG: 65/75 (90.3) CG: 16/24 (27.3) aRR: 1.27 (95% CI, 0.99 to 1.64; *p*=0.06) Exclusive breastmilk feeding in the last 24 hours, 1–2 weeks postrandomization, N (%)	Infant weight gain between birth and 3 months of age (z-score), mean (SD) IG: -1.1 (2.3) CG: -1.2 (1.1) EE: 0.10 (−0.62 to 0.82); aEE: 0.17 (−0.60 to 0.95); p=0.65 Infant postrandomization weight gain between baseline and 3 months of age (z-score), mean (SD) IG: -1.0 (1.6) CG: -1.1 (1.3) EE: 0.04 (−0.82 to 0.90); aEE: 0.10 (−0.83 to 1.03), p=0.83	Results adjusted for center, infant's age at randomization, and parity.

Author, Year Design ROB	Breastfeeding Outcomes	Related Outcomes	Comment
	IG: 51/80 (65.4) CG: 66/89 (75.9) Missing: IG=2; CG=2 RR: 0.86 (95% CI, 0.60 to 1.24); aRR: 0.86 (95% CI, 0.59 to 1.24); *p*=0.42 3 months of age IG: 45 /80(63.4) CG: 50/89 (66.7) Missing: IG=9; CG=14 EE: 0.95 (0.64 to1.42); aEE: 0.92 (0.61 to 1.39); *p*=0.69 Exclusive direct breastfeeding in the last 24 hours, N (%) 1–2 weeks postrandomization, IG: 35/80 (44.9) CG: 43/89 (49.4) Missing: IG=2; CG=2 RR: 0.91 (95% CI, 0.58 to 1.42); aRR: 0.92 (95% CI, 0.59 to 1.45); *p*=0.73 3 months of age IG: 38/80 (53.5) CG: 39/89 (52.7) Missing: IG=9; CG=15 EE: 1.02 (0.65 to 1.59); aEE: 1.03 (0.65 to 1.62); *p*=0.90 Mother's breastfeeding self-efficacy, 3 months, median (IQR) (BSES) IG: 60 (47–65) CG: 56.5 (47–65) EE: 4 (−1.8 to 9.8); aEE 0.3 (−5.2 to 5.8); *p*=0.92 Amount of breastfeeding support used (number of contacts) at 3 months, median (IQR) IG: 3 (2-5) CG: 2 (1-4) Missing or NA: IG=30; CG=27 EE: 1 (0.1 to 1.9); aEE: −0.3 (−1.5 to 1.0); *p*=0.68 Maternal anxiety and depression, N (%)		

Author, Year Design ROB	Breastfeeding Outcomes	Related Outcomes	Comment
	1–2 weeks postrandomization IG: 23/80 (29.9) CG: 29/89 (33.3) Missing: IG=3; CG=2 RR: 0.90 (95% CI, 0.52 to 1.55); CG: 0.92 (95% CI, 0.53 to 1.63); *p*=0.79 3 months of age IG: 29/80 (39.7) CG: 26/89 (34.7) Missing: IG=7; CG=14 EE: 1.15 (0.67 to 1.95); aEE: 1.12 (0.65 to 1.93); *p*=0.69 Maternal anxiety and depression, 1–2 weeks postrandomization, score median (IQR) IG: 0.9 (0.8 to 1.0) CG: 0.9 (0.8 to 1.0) RR: 0.0 (95% CI, −0.1 to 0.1); aRR: 0.0 (95% CI, −0.1 to 0.1); *p*=0.94 Maternal anxiety and depression, EQ-5D-5L overall index value, 3 months, median (IQR) IG: 0.9 (0.8-1.0) CG: 0.9 (0.8-1.0) EE:0.0 (−0.1 to 0.1); aEE: 0.0 (−0.1 to 0.1); *p*=0.94		
Schlatter, 2019[34] Cohort Serious	LATCH score change, mean, from baseline to 2.5 weeks IG: 6.9 to 9.5 CG: 7.5 to 9.5 *P*=0.044 Breastfeeding problems, reduction 2.5 weeks after frenulotomy, Fisher's exact test 23 of 33 infants with indication for frenulotomy had fewer breastfeeding problems at follow-up *P*=0.010 Breastfeeding problems after the procedure among dyads with a ATLFF score of <11, N (%) (IG=23; CG=10) IG: 3 (13.0) CG: 6 (60)	NA	NA

Author, Year Design ROB	Breastfeeding Outcomes	Related Outcomes	Comment
Sharma, 2015[35] Cohort Serious	Infant Breastfeeding Assessment Tool, post-intervention, mean (SD) IG: 9.19 (2.44), within-group change from baseline *p*=0.0001 CG: 6.00 (1.73), within-group change from baseline *p*=0.16 Mother report of General Improvement in Breastfeeding, N (%) All infants IG: 29 (81) CG: 1 (17) *p*=0.0074 Mother report of improvement in breastfeeding, Infants aged <30 days, n=17 IG: 16 (94) Infants aged >30 days, n=19 IG: 13 (68)	NA	Follow-up dates NR for either group.
Steehler, 2012[36] Cohort Serious	Stopped breastfeeding due to difficulty or pain due to ankyloglossia, N (%) (IG=82; CG=9) IG: 14 (17.1) CG: 3 (33.3) Continued to breastfeed following frenotomy/diagnosis of ankyloglossia, N (%) (IG=82; CG=9) IG: 68 (82.9) CG: 6 (66.7) Average duration of breastfeeding, months IG: 7.09 CG: 6.28 No significant difference on t-test Average age for starting solid foods, months IG: 5.8 CG: 6	NA	NA

Notes: [a] Recruitment was halted due to poor recruitment success and the COVID-19 pandemic.

Abbreviations: ATLFF = Assessment Tool for Lingual Frenulum Function; AEE = adjusted effect estimate; ARR = adjusted risk ratio; BSES = Breastfeeding Self-Efficacy Scale; BSES-SF = Breastfeeding Self-Efficacy Scale–Short Form; CG = control group; EE = effect estimate; EQ = efficacy question; EQ-5D-5L = EuroQoL-5 Dimensions, five-level

version; GSQ-I = Gastroesophageal Symptom Scale – Infant; IBFAT = Infant Breastfeeding Assessment Tool; IG = intervention group; IQR = interquartile range; ITT = intention to treat; LATCH = Latch, Audible swallowing, Type of nipple, Comfort, Hold; N = number; NA = not applicable; NR = not reported; PR = prevalence ratio; RCT = randomized controlled trial; RR = risk ratio; SD = standard deviation; SF-MPQ = Short-Form McGill Pain Questionnaire; VAS = Visual Analog Scale.

Table C-4. Comparative studies examining frenotomy/frenectomy: SQ results

Author, Year Design ROB	Intervention N Comparator N	Study Reported No Harms Occurred	Reported Harms
Berry, 2012[26] RCT High for breastfeeding improvement Low for other outcomes	Frenotomy with scissors: 30 Sham frenotomy: 30	NA	Small amount of bleeding, N (%) 3 (5)
Buryk, 2011[27] RCT Low	Frenotomy with scissor: 30 Sham frenotomy: 28	There were no complications from the procedure in any of the infants.	NA
Dixon, 2018[32] Cohort	Frenotomy with scissors, 2016 audit (Study 1): 264 released 45 not released Frenotomy with scissors, 2017 audit (Study 2):55 released 3 not released	NA	1 infant required repeat surgery 1 had a minor bleeding 1 required paracetomol for analgesia
Dollberg, 2006[28] RCT Low	Frenotomy (method unclear), breastfeeding, sham,	In all cases, there was minimal blood loss, that is, no more than a drop or 2, collected on sterile gauze, and infant crying lasted a few seconds only. There was no significant side effect of the frenotomy,	NA

Author, Year Design ROB	Intervention N Comparator N	Study Reported No Harms Occurred	Reported Harms
	breastfeeding: 14 Sham, breastfeeding, frenotomy, breastfeeding: 11	and bleeding (a few drops) was controlled within seconds in all cases.	
Emond, 2014[29] RCT High	Immediate frenotomy (method unclear): 55 Delayed frenotomy: 52	NA	Repeated procedure (Total N=99) 4 (4%) "Small white patch at base of the frenulum reported at 5 days post procedure" (Total N=99) 63 (64%) Days to heal, Median (Range): 7 (1–30)
Ghaheri, 2022[30] RCT High	Frenotomy with laser immediately after enrollment: 24 Delayed treatment. Continuation of non-frenotomy therapies with offer of frenotomy with laser: ~10 days after randomization: 24	No adverse events or unanticipated problems were experienced during the study duration.	NA
Hogan, 2005[8] RCT	Frenotomy with scissors: 28	There were no problems with infection or bleeding, either primary or secondary. Most babies cried for only a few seconds until they were given a feed. Division of tongue-ties is not an operation but a procedure, and	NA

Author, Year Design ROB	Intervention N Comparator N	Study Reported No Harms Occurred	Reported Harms
High	Intensive support, advice, and help from lactation consultant: 29	mothers of older babies commented that it was much less traumatic than immunization.	
Knight, 2023[31] RCT High	Frenotomy (method unclear) with standard breastfeeding support: 80 Breastfeeding support: 89	NA	Bleeding: N=1 Salivary duct damage: N=1 Accidental cut to the tongue and salivary duct damage: N=1 Frenotomies with complications IG: 1/80 (1.25%) CG: 2/89 (2.2%)
Sharma, 2015[35] Cohort Serious	Frenotomy with scissors: 36 No frenotomy, infant feeding support: 6	There were no surgical complications within the group of patients that underwent frenotomy.	NA
Steehler, 2012[36] Cohort Serious	Frenotomy with scissors: 302 No frenotomy: 65	NA	Recurrent ankyloglossia secondary to scarring: n=8 (2.6%) Repeat intervention successfully treated all 8 of these patients

Abbreviations: CG = control group; IG = intervention group; N = number; NA = not applicable; RCT = randomized controlled trial; ROB = risk of bias; SQ = safety question.

Table C-5. Single-arm studies examining frenotomy/frenectomy with scissors: SQ study characteristics

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
Akbari, 2023[37] No specific grant from funding agencies	Large regional hospital in Central Australia that serves a rural and remote community	Inclusion: Births occurring in hospital between January 2013 and December 2018 with codes for ankyloglossia and frenotomy, <24 months of age. Exclusion: NR	Diagnosis: 3 days (SD NR)	NR for those with frenotomy (n=474) Overall Sample (n=478) Male: 280 (58.6) Female: 198 (41.4)	NR	Latching issues: 334 (70) Maternal nipple pain: 187 (39) Sucking issues: 138 (29) No feeding issues: 87 (18)	Ankyloglossia/ tongue-tie: 474 (100) NR for those with frenotomy; among all infants diagnosed with ankyloglossia: Posterior: 118 (25) Anterior: 69 (14) Anterior and posterior: 4 (0.8) Other: 11 (2) Not specified: 278 (58)	NR	NR
Amir, 2005[38] NR	Breastfeeding clinic in a tertiary maternity hospital (Royal Women's Hospital) in Melbourne, Australia between August 2002 and July 2003	Inclusion: Presenting to breastfeeding service and HATLFF indicated an impaired lingual function and frenulum was visualized to be a thin membrane. Exclusion: NR	18 days (range: 3 to 98, median 12.5)	Assessed Male: 29 (63) Female: 17 (37) Received tongue-tie release Male: 22 (63)	NR	Based on total assessed for tongue-tie Difficulty attaching baby to breast: 21 (31.8) Nipple pain: 13 (19.7) Nipple damage: 4 (6.1) Frequent feeding: 7 (10.6)	Based on total assessed Tongue-tie: 66 (100) NR	Not clearly reported but assume 100% breastfed based on sample; bottle fed: NR	Based on total assessed Family history of tongue-tie: 7 No family history: 36

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
				Female: 13 (37)		Prolonged feeding: 2 (3.0) Poor weight gain: 8 (12.1) Not breastfeeding or data missing: 5 (7.5)			
Argiris, 2011[39] NR	Patients at the Royal Free Hospital Neonatal Tongue Tie service run by the Department of Otolaryngology in London, U.K.	Inclusion: Patients with breastfeeding difficulties and diagnosed by either a lactation nurse consultant or ENT consultant and undergoing tongue-tie division between August and October 2008. Exclusion: NR	4 weeks (NR); range: 1 day to 12 weeks	Male: 33 (71.7) Female: 13 (28.2)	NR	Poor latch: 31 (67) Sore nipples: 29 (63) Damaged nipples: 20 (43) Coming on/off the breast: 24 (50) Baby not satisfied after feeding: 15 (30) Poor infant weight gain: 10 (22)	Tongue-tie: 46 (100) NR	46 (100) (attempted breast-feeding)	NR
Ballard, 2002[40] March of Dimes, Ohio Chapter, Cincinnati Children's Hospital Medical Center, and University Hospital, Inc,	Cincinnati Children's Hospital between January 1, 1998, and June 30, 2001	Inclusion: Full-term breastfeeding infants, either inpatient or outpatient (presenting with breastfeeding problems), function score of <11 out of a possible 14 on HATLFF. Participants were assessed for latch and maternal nipple pain and, when	Median age with poor latch: 1.2 days (range: 0.7 to 2.0) Median age with maternal nipple pain: 2.0 days (range: 1.0 to 12.0)	Total population ratio of boys to girls: 1.5:1 NR for ankyloglossia group	NR	Infants were examined for ankyloglossia and significant ankyloglossia was defined as a function score of less than 11 out of 14 or an appearance score of less than 8 out of 10 on HATLFF. Latch and maternal nipple	Ankyloglossia: 127 (100) NR	Breast-feeding (per criteria): 100	Family history of ankyloglossia: 26 (21)

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
members of the Health Alliance of Greater Cincinnati		appropriate, frenuloplasty was offered. Exclusion: NR				pain assessed if ankyloglossia was identified, pain measured on a scale from 1 to 10, with 1 meaning extremely mild discomfort and 10 meaning severe or intolerable pain. Of the 88 infants with ankyloglossia identified in the hospital: Poor latch: 56 (63.6) Nipple pain: 32 (36.3) Of the 35 infants with ankyloglossia identified in outpatient lactation center: Poor latch: 14 (40.0) Nipple pain: 21 (60.0) 6 outpatients presented with failure to thrive (4 poor latch and 2 maternal			

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
						nipple pain) 1 infant presented with poor latch and failure to thrive, presented at 4.5 months and required hospitalization for fatty infiltration of the liver secondary to prolonged starvation 1 infant at 1 month presented with ulcer of the posterior hard palate			
Barberá-Pérez, 2021[41] None	Babies born in the maternity ward of a baby-friendly hospital (Spain, February–August 2019)	Inclusion: Frenotomies performed in "the first days of life" before discharge, breastfed, and "with no associated comorbidities." Exclusion: Newborns with artificial feeding and those who underwent frenotomy after discharge from the maternity ward.	1 day; range 4 hours to 6 days	Male: 18 (54.4) Female: 15 (45.5)	NR	Early frenotomy was advised for infants who presented with significant ankyloglossia with Coryllos type 1 frenulums; functional evaluation and maternal symptoms were considered for other infants Coryllos typeType 1: 4 (12.1) Type 2: 15 (45.4) Type 3: 8 (24.2) Type 4: 6 (18.2)	Ankyloglossia: 33 (100) per protocol NR	Breastfed: 33 (100) per protocol; unclear if supplementing with formula was permitted	Jaundice in newborns: 6 (18.2)

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
						Maternal symptoms Difficulty with latching: 18 (54.5) Weight loss >10%: 3 (9.1) Pain: 29 (87.9) Cracked nipples: 18 (54.5) Mastitis: 0 (0) Pain intensity (VAS), Mild (0-2): 0 Moderate (3–7): 19 (57.6) Sever (8–10): 10 (30.3)			
Benoiton, 2016[42] NR	Prospective audit of ENT outpatient clinic in New Zealand between May 2014 and September 2015	Inclusion: Referred by a lactation consultant with ankyloglossia and/or lip-tie with breastfeeding (or occasionally bottle feeding) concerns. Exclusion: Patients with comorbidities including known coagulopathy and significant craniofacial anomaly with risk of	Median: 6.6 weeks, range: 2–10	Male: 21 (62) Female: 13 (38)	New Zealand European: 25 (24) Other race/ ethnicities NR	Infants were assessed by lactation consultants and referred for outpatient frenotomy. Symptoms Latching issues: 29 (85) Painful nipples: 22 (65) Poor weight gain: 7 (21) Clicking noises: 4	Posterior ankyloglossia and lip-tie: 10 (29) Upper lip-tie only: 3 (9) Ankyloglossia only: 21 (62) Anterior and posterior: 1 (3) Posterior: 20 (59)	Breast-feeding: 34 (100) per protocol	Previous anterior frenotomy: 14 (41)

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
		tongue base obstruction.				(12) Unsettled feeding: 2 (6) Mean Hazelbaker score Appearance 4.7 (range 1 to 8) Function 8.1 (range 4 to 14)			
Bhandarkar, 2022[43] No sources of funding	Retrospective review in a tertiary children's hospital (London, U.K.)	Inclusion: Less than 60 days corrected gestational age referred to tongue-tie service. Exclusion: Infants >60 days corrected gestational age were excluded.	Median: 14 days; range 7 to 58 days	NR	NR	Referral after conservative methods to promote latching and breastfeeding failed	Ankyloglossia: 599 (100) per protocol NR	NR	NR
Blenkinsop, 2003[45] NR	Audit at St Peter's Hospital in Chertsey Surrey, U.K.	Inclusion: Retrospective audit of babies referred for frenulotomy between January and June 2002, and prospective audit of 21 babies in 2003. Exclusion: NR	Diagnosis range: 1 day to 6 weeks	NR	NR	Feeding problems apparently caused by tongue-tie	Tongue-tie: 21 (100) NR	Bottle feeding only: 3 (14.3)	NR

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
Dollberg, 2014[48] NR	Unclear, bedside (presumably hospital) and offices in Israel between March 2010 and October 2010	Inclusion: Term infants with breastfeeding difficulties who underwent lingual frenotomy. Exclusion: Congenital anomalies	Median: 14 days (range 1 to 135)	Female: 101 (41) [a] Male: 143 (59)	Israeli Jewish: 244 (100)	Infants were referred after examination by lactation consultant rules out other reasons for breastfeeding difficulties. Coryllos type of tongue-tie, % Type 1: 13% Type 2: 31% Type 1: 24% Type 4: 32% Presenting symptom, N (%) Sore maternal nipples: 203 (83) Pain with bruising: 152 (62) Pain without bruising: 92 (38) Latching difficulties: 134 (55) Repeated, frequent detachment of the infant from the breast: 64% Inability to feed: 7% Falling asleep on the	Lingual tie: 244 (100) per protocol NR	Breastfed: 244 (100) per protocol	Problems with breastfeeding with previous offspring (n=104), N (%) 64 (61)

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
						breast: 7% Visual description of frenular thickness, N (%) Thin: 105 (43) Thick: 96 (40) Graded (thin distally and thick proximally): 42 (17) Notched tongue tip: 78 (32) Tongue elevation above midmouth: 51 (21)			
Ferrés-Amat, 2017[50] NR	Infants referred by the pediatric service to the Suction Pathology Unit ( ) at Hospital de Nens in Barcelona, Spain	Inclusion: Infants 0 to 6 months of age, healthy ASA I, without a diagnosis of systemic disease or syndrome, diagnosed with ankyloglossia associated with inefficient suction. Exclusion: NR	NR	Male: 62 (70.5) Female: 26 (29.5) [b]	NR	Coryllos type, N (%) Type 1–2: 33 (37.5) Type 3: 52 (59.1) Type 4: 3 (3.41)[b] Ankyloglossia was diagnosed using the Coryllos classification as well as consideration of poor weight gain (less than 100 grams a week), excessively long breastfeeds (>60 minutes), and maternal pain	NR NR	Maternal: 36 (40.9) Mixed: 52 (59.1)[b]	Previous breastfeeding: 12 (13.6)[b] Family history of ankyloglossia: 24 (27.3)[b]

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
Geddes, 2008[11] Medela AG provided scholarship to first author but did not sponsor this specific study	Breastfeeding Centre at King Edward Memorial Hospital (the study site) or private health centers in Western Australia	Inclusion: Mothers who had received lactation advice and follow-up, yet breastfeeding difficulties had not resolved Exclusion: NR	33 days (28), range 4–131 days	NR	NR	Breastfeeding difficulties unresolved after lactation advice and follow-up	Tongue-tie: 24 (100) NR	NR, based on sample 100% were breastfeeding, NR on bottle feeding	NR
Griffiths, 2004[53] NR	One Center between December 1999 and December 2001 (No other information provided, assume in the U.K.)	Inclusion: Infant < 3 months; mother wanting to breastfeed but experiencing difficulty despite professional support Exclusion: NR	19 days	Male: Female ratio 2:1	NR	Difficulty latching: 192 (88) Painful, sore, or bleeding nipples: 167 (77) "Continuous" feeding cycle: 156 (72) All three symptoms: 112 (52)	Tongue-tie: 215 (100) NR	Breast-feeding: 100% (per protocol) Had tried Bottle feeding: 111; breast milk n=104; formula n=7	Family history of tongue-tie: 44%
Hong, 2010[58] NR	Retrospective chart review of outpatient pediatric otolaryngology clinic for ankyloglossia from July 2007 to July 2009	Inclusion: Healthy infants with no other significant medical issues Exclusion: NR	In weeks Anterior: 2.4 Posterior: 2.9 Median age, overall: 2.7 weeks; range 1 day to 24 weeks	Female Anterior: 101 Posterior: 12 Male Anterior: 221 Posterior: 7 Overall Male: 228	NR	In infants with posterior tongue-tie Maternal nipple pain and bloody nipple discharge: 17 (89) Latching-on difficulties: 16 (84) Prolonged feeds: 15 (80) Poor weight gain: 3 (16)	Tongue-tie: 341 (100) Anterior ankyloglossia: 322 (94) Posterior ankyloglossia: 19 (6)	Breastfed: 341 (100), based on sample included Bottle fed: NR	NR

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
				(67) Female: 113 (33)					
Illing, 2019[59] NR	Medical center with a GP and lactation consultant focused on ankyloglossia who accept referrals (New Zealand, 2016–2017)	Inclusion: Infant <6 months with confirmed ankyloglossia, who received a frenotomy for feeding-related issues Exclusion: Prior frenotomy with reattachment and requesting a second frenotomy	44 days (35)	Male: 109 (62) Female: 67 (38)	Pakeha/ New Zealand European: 126 (72) Maori: 27 (15) European (other): 10 (6) Asian: 7 (4) Other: 6 (3)	Issues with latching: 115 (65) Nipple pain when breastfeeding: 84 (48) Slow to feed: 52 (30) Falling asleep breastfeeding: 46 (26) Unsettled/fussy baby: 37 (21) Poor weight gain: 36 (20) Leaking milk while breastfeeding: 36 (20) Not tolerating breastfeeding: 22 (13) Windy: 20 (11) Very frequent feeding: 18 (10) Reflux issues: 11 (6) Breastfeeding so painful nipple shields required: 8 (5) Maternal milk supply issues: 8 (5)	NR NR	Not only breast-feeding: 93 (53)	NR

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
						Recurrent mastitis: 6 (3) Cosmetic concerns: 5 (3) Told to by a professional: 136 (77) Parental concern of loud clicking while feeding: 94 (53)			
Masaitis, 1996[61] NR	Mother-Baby program, including all mothers with problems at Bess Kaiser Medical Center, Portland, Oregon	Inclusion: Infants where ankyloglossia caused breastfeeding problems Exclusion: NR	Average: 5.7 days Median: 3 days (1–24 days)	Male: 20 (55.6) Female: 16 (44.4)	NR	Tongue does not cross alveolar ridge: 29 (80.6) Heart-shaped tongue: 29 (80.6) Poor attachment at breast: 27 (75) Injured nipples (maternal): 27 (75) Frenulum attached to the tip of the tongue: 24 (66.7) Previous breastfeeding failure (maternal): 9 (25) Inadequate weight gain: 7 (19.4) Clicking sound with nursing: 4 (11.1) Breast abscess (maternal): 1 (2.8)	Ankyloglossia: 36 (100) NR	NR	Family history of ankyloglossia: 21 (58) First-time mothers: 22 (61.1) Two children: 12 (33.3) Three children: 1 (2.8) Four children: 1 (2.8)

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
McKenna, 2024[62] Authors disclose no funding	Tertiary care, free-standing children's hospital (New York). Data was EMR data; participants were not recruited.	Inclusion: Infants diagnosed with CPT code 41010 (lingual frenectomy) or ICD-10 Q38.1 (ankyloglossia); eligible infants were also identified via billing codes or EMR. All records identified at this hospital during the time period of the study (2018–2021) were included Exclusion: NR; frenotomy was not completed if infant had an acute infection or disease or if caregivers declined or procedure not indicated	NR	Male: 52 (50.5) Female: 34 (39.5)	Race White or Caucasian: 71 (83.5) Black or African American: 8 (9.4) Other: 6 (7.1) Ethnicity Hispanic or Latino: 4 (5.0)	Coryllos type 1: 23 (27.1) Coryllos type 2: 46 (54.1) Coryllos type 3: 9 (10.6) Coryllos type 4: 7 (8.2) (n=85) Lateralization 0: 11 (14.3) Lateralization 1: 55 (71.4) Lateralization 2: 11 (14.3) (n=77) Tongue lift 0: 27 (34.2) Tongue lift 1: 47 (59.5) Tongue lift 2: 5 (6.3) (n=79) Extension 0: 4 (5.1) Extension 1: 73 (92.4) Extension 2: 2 (2.5) (n=79)	NR NR	NR	Number of comorbiditie s, Mean (SD) 0.7 (1.1) Range: 0–5

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
						aHATTLF 0: 0 (0) aHATTLF 1: 10 (13.0) aHATTLF 2: 19 (24.7) aHATTLF 3: 36 (46.8) aHATTLF 4: 9 (11.7) aHATTLF 5: 2 (2.6) aHATTLF 6: 1 (1.3) (n=77) Pre-frenotomy tip-to-frenulum length (mm), mean (SD) 2.9 (2.0) Range: 0–8 (n=85)			
Mettias, 2013[63] NR	Outpatients hospital ENT clinic in the U.K. (Glan Clwyd Hospital) between May and June 2011	Inclusion: Babies who had a tongue-tie division Exclusion: NR	4.1 weeks (3.2) (based on overall)[c]	Based on overall Male: 49.2% Female: 50.8%	NR	Mothers were referred to the clinic by midwives and health visitors Difficulty breastfeeding: 66.7% Poor growth: 11.1% Limitation in tongue movement 22.2% Breast problems such as cracking and sore nipples: 27.7%	Tongue-tie: 63 (100) NR	Breastfed: Unclear, 67% had difficulty Bottle fed: NR	NR

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
						Asymptomatic: 13.9%			
Miranda, 2010[64] Study carried out without any funding sources or commercial associations	Outpatient departments	Inclusion: Neonates with diagnosis of ankyloglossia and breastfeeding difficulty resistant to initial lactation consultant management Exclusion: Nonplastic surgery pediatric patients	Range 12–36 days	NR	NR	In the 51 infants who returned for follow-up: Poor latch: 28 (55) Nipple pain: 27 (53) Nipple cracking: 19 (37) Nipple bleeding: 11 (22)	Ankyloglossia: 62 (100) NR	Unclear, based on sample 100% breast-feeding, NR for bottle feeding	NR
Muldoon, 2017[65] No funding	Seven health care/GP clinics in Ireland that perform frenotomy from March 2016 to July 2016	Inclusion: Breastfed infants and attending a health care clinic for a planned frenotomy Exclusion: NR	7 weeks, 3 days (6 weeks, 2 days)	NR	NR	Difficulty latching baby to the breast: 37 (38) Nipple pain: 19 (20) Baby unsettled post-feed: 10 (10) Concern over later speech: 6 (6) Difficulty maintaining latch: 5 (5) Mastitis: 3 (3) Breast feeling full post-feed: 1 (1) Concern over weight gain: 3 (3) Previous infant had tongue-tie and feeding improved	Tongue-tie: 89 (100) based on included sample NR	Exclusive breastfeeding: 57 (58) Expressing breast milk: 2 (2) Combination breastfeed and expressed breast milk: 17 (17) Combination breastfeeding and formula: 12 (12) Formula	First baby Yes: 37 (38) No: 61 (62) Family history of tongue-tie Yes: 40 (41) No: 58 (59)

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
						after frenotomy: 3 (3) Concern over milk supply: 2 (2) Coryllos type Type 1: 11 (11) Type 2: 45 (46) Type 3: 15 (15) Type 4: 24 (24)		feeding: 6 (6)	
Narsat, 2022[66] Received no external funding	Kastamonu Training and Research Hospital, tertiary training and research hospital in Turkey	Inclusion: Infants born at the hospital between 1 January and 30 June 2022 with ankyloglossia and breastfeeding difficulties Exclusion: Infants with problems such as prematurity, swallowing dysfunction, choanal atresia, swallowing disorders due to neurological developmental disorders, anatomical problems besides ankyloglossia, and conditions that prevent breastfeeding	NR	NR for frenotomy group Overall sample (N=234) Male: 124 (52.0) Female: 110 (47.0)	NR for frenotomy group	Coryllos type 1: 14 (6.1) Coryllos type 2: 32 (13.7) Coryllos type 3: 15 (6.4) Coryllos type 4: 6 (2.5)	NR NR	NR	NR

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
O'Callahan, 2013[67] Middlesex Hospital	Referrals to primary care practice in central Connecticut and western Massachusetts between December 2006 and March 2011	Inclusion: Infants who underwent a frenotomy for ankyloglossia[d] Exclusion: NR	Median: 41 days, range NR[b]	Male: 80 (51) Female: 77 (49)[b]	NR	Referrals for the frenotomy were made by lactation consultants, physicians, and craniosacral practitioners. Assessment included maternal report of breastfeeding difficulties, examination of infant looking for swallowing and neurological defects, and a suck evaluation. The frenulum was also visually examined. Type I and Type II combined ankyloglossia: 18 (12) Type III ankyloglossia: 52 (33) Type IV ankyloglossia: 87 (55)[b]	Maxillary tie Yes: 44 (44) No: 55 (56)[b] Anterior: 18 (12)[b]	Breastfed: 157 (100) based on included sample Bottle fed: NR	NR

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
Pransky, 2015[69] NR	Patient records from a dedicated ankyloglossia clinic at Rady Children's Hospital in San Diego, California, January 2014 to December 2014	Inclusion: Healthy infants with ankyloglossia and/or lip-tie and no other significant medical issues Exclusion: NR	Infant age NR	Male: 362 (59) Female: 256 (41)[e]	African Americans 7 (1) Asian American: 11 (2) Caucasians 338 (55) Hispanics 157 (25) Did not specify their ethnicity: 105 (17)[e]	Breastfeeding difficulties and oral cavity anomaly determined by physical examination.	Tongue-tie only: 410 Lip-tie only: 14 Tongue-tie and lip-tie: 67 Anterior ankyloglossia: 324 (290 + 34 also with lip-tie) (77.9) Posterior ankyloglossia: 153 (120 + 33 also with lip-tie) (32.1)	NR, assuming 100% attempted	Family history of ankyloglossia: 207 (33)[e]
Ramoser, 2019[70] Study did not receive specific funding	Clinic for Pediatrics at at the Medical University of Innsbruck in Austria between February 2011 and February 2017	Inclusion: Infants who underwent a frenotomy at the study location[f] Exclusion: NR	Median: 6 weeks, range 0.5–52 weeks	Male: 168 (56.9) Female: 127 (43.1)	NR	Diagnosis based on clinical symptoms, signs such as latch, number of meals a day, and weight, and the HATLFF Inadequate latch (breastfeeding): 198 (67.1) Inadequate latch (bottle feeding): 46 (15.6) Painful nipples: 131 (44.4)	Tongue-tie:295 (100) based on included sample Posterior: 214 (72.5) Anterior: 17 (5.8)	Unclear, 198 had inadequate breastfeeding latch (67.1) and 46 had inadequate bottle feeding latch (15.6)	NR

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
						Sore nipples: 103 (34.9) Poor weight gain: 68 (23.1) Dribbling of milk from the corner of the mouth: 63 (21.4) Breast milk insufficiency: 47 (15.9) Mastitis: 22 (7.5) Hindered intake of solid foods: 7 (2.4) Articulation disorder: 1 (0.34) Misaligned teeth: 1 (0.34) Other symptoms (deformed nipples, mamillar vasospasm after breastfeeding, agitation, aerophagia, increased salivation, colics, vomited and hematemesis, problems with swallowing, and problems licking ice cream): 23 (7.8)			

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
Ridgers, 2009[71] NR	Frimley Park hospital lactation and breastfeeding clinic located in the U.K.	Inclusion: NR. Appears to report on all frenotomies done in the clinic during a 24-month time period Exclusion: NR	Median: 10 days (range 3–70)	Male: 141 (64) Female: 79 (36)	NR	Difficult attachment: 95 (43) Nipple soreness: 86 (39) Frequent feeds: 57 (26) Infant not attaching: 40 (18) Protracted feeds: 40 (18) Dribbles on bottle: 18 (8) Poor milk supply: 17 (8) Infant never attached: 9 (4) Mastitis: 9 (4)	Tongue-tie: 220 (100%) NR	Breast-feeding: 130 (59) Artificially feeding: 35 (16) Mix of both: 55 (25)	Family history of tongue-tie: 90 (41)
Sethi, 2013[72] NR	ENT outpatient department (Pinderfields Hospital) in Wakefield, West Yorkshire, England (U.K.) between February 2008 and February 2011	Inclusion: Infants who underwent frenotomy in the outpatient clinic Exclusion: NR	19 days (NR), range 3–120 days[g]	Male: 35 (67) Female: 17 (33)[g]	NR	Referrals originated from midwives, lactation consultants; pediatricians, and general practitioners. Senior authors assessed presence of tongue-tie based on history of breastfeeding difficulties, family history of tongue-tie, and full oral examination.	Tongue-tie: 85 (100) per protocol NR	Breast-feeding exclusively: 28 (53.8) Supplementing with expressed breast milk: 22 (42.3) Formula-fed exclusively: 2 (3.8) [g]	NR

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
						Poor latch: 49 (94) Continual feeding: 18 (34) Poor weight gain: 10 (19) Sore nipples: 6 (12) Excess wind: 2 (4)			
Siggard, 2022[73] NR	Private ENT clinics (Denmark)	Inclusion: NR Exclusion: NR	NR	In follow-up group (n=163) Males: 103 (63)	NR	Dysfunctional breastfeeding and ENT specialist assessment Preoperative symptoms Infant breastfeeding difficulty: 110 (67) Insufficient infant weight gain: 33 (20) Infant abdominal pain/flatulence: 41 (25) Maternal nipple/breast pain during breastfeeding: 66 (40) Other symptoms: 67 (41)	NR NR	Breast-feeding: 230 (total seen over study period)	Comorbiditie s (such as premature birth, reflux syndrome, icterus, congenital heart condition, or asthmatic bronchitis): 35 (21) Infants with siblings who needed a frenotomy: 27 (17)
Srinivasan, 2019[75]	Herzl–Goldfarb Breastfeeding Clinic in	Inclusion: Infants <12 weeks of age, with posterior tongue-tie	Mean age at frenotomy: 37.9 days	Male: 20 (66.7)	NR	Posterior tongue-tie diagnosed by one of the clinic physicians,	Tongue-tie: 30 (100)	Breastfed: 30 (100)	Previous breastfeedin g

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
NR	Montreal, Canada between April 2014 and April 2015	and having persistent breastfeeding problems (e.g., poor latch, nipple pain, poor infant gain and/or poor milk supply) despite lactation support Exclusion: Anterior tongue-tie), gestational age <37 weeks, neurodevelopmental anomalies (e.g., Down syndrome, cleft lip/palate), and previous frenotomy	(21.1) (median=34.5 days, IQR=34, range=9–80)	Female: 10 (33.3)		corresponding to Coryllos types 3 and 4 Frenotomy Decision Tool for Breastfeeding Dyads, median (IQR): 5.0 (3.0), range 2.5-9.0 LATCH, median (IQR): 7.5 (2.0) Pain, left nipple , median (IQR): 3.0 (5.0) (n mothers=26) Pain, right nipple, median (IQR): 3.25 (5.0) (n mothers=24)	Posterior 30 (100)	Bottle fed: NR	experience, n (%) 9 (30) Previous breastfeeding problems, n (%) 7 (77.8)
Srinivasan, 2006[76] No financial support for this project	Recruited through the Goldfarb Breastfeeding Program at the Jewish General Hospital in Montreal, Canada from August 2004 and February 2005; referrals solicited through mailed	Inclusion: Infant <12 weeks, mothers intending to begin or continue breastfeeding Exclusion: Congenital anomalies or developmental delay	19 days (19) Median: 10 days; range 2–71 days	Male: 18 (67.7) Female: 9 (33.3)	NR	Referral for ankyloglossia due to maternal nipple pain, trauma, latching difficulties, and/or poor infant weight gain. N=1 vasospasm of the nipple. N=2 decreased milk supply. Dyads were	N at enrollment NR (probably tongue-tie) NR	NR	NR

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
	information to various local hospitals, local community health centers, and word of mouth					evaluated using the following decision tree: Mother with nipple pain/trauma while breastfeeding *and/or* inability to maintain latch *and/or* poor weight gain in the infant (<15 g/d), *and* A visible membrane anterior to the base of the tongue, which restricts tongue movement, leading to: An inability to touch the roof of the mouth, *or* an inability to cup an examining finger, *or* an inability to protrude the tongue past the gum line			
Todd, 2015[77] NR	Hospital neonatal department in Australia in 2008 or 2011[h]	Inclusion: Record audit of infants who had a tongue-tie division Exclusion: NR	At time of division, days 2011: 9.7 (6.2) 2008: 6.5 (4.5)	2011 Male: 91 (63.2) Female: 53 (36.8) 2008 Male: 77	NR	Tongue-tie types, both groups Types 1 and 2 anterior: approx. 1/3 of group Type 3: approx. 1/3 of group Type IV and V:	Tongue-tie 2011: 144 (100) per protocol 2008: 115 (100) per protocol NR	NR	NR

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
				(67.0) Female: 38 (33.0)		approx. 1/3 of group Sore nipples 2011: 140 (97.2) 2008: 98 (85.4)			
Toner, 2014[78] NR	Otorhinolaryngology office at a children's hospital, both main campus and satellite offices in the suburbs of Philadelphia between 2003 and 2008	Inclusion: Infants who had a frenotomy preformed in the otorhinolaryngology office of the Children's Hospital of Philadelphia Exclusion: NR	NR	NR	NR	Difficulty latching: 76% Other reported problems: pain with breastfeeding, prolonged feeding, and combination of symptoms. One patient had no pre-op symptoms but had the procedure performed because of parental preference.	NR; probably tongue-tie based on description of procedure NR	Breastfed: 19 (76) Bottle fed: 4 (16) Both: 2 (8)	NR
Towfighti, 2022[79] No funding	Retrospective chart review of infants at an otolaryngology-head and neck surgery department (Washington, DC, U.S.)	Inclusion: Infants ~0-3 months of age who presented with feeding difficulties who had a release of the lingual frenulum Exclusion: Infants who did not receive any intervention at the first clinic visit, infants with	23.6 days; range 2 to 108 days (n=316 infants presenting for evaluation)	Male: 134 (59.8) Female: 90 (40.2) (n=224 infants receiving lingual frenotomy or lingual and	Race/ethnicity Asian: 12 (5.4) Black: 23 (10.3) Hispanic: 15 (6.7) White: 160 (71.4) Other: 14	Breastfeeding difficulties based on evaluation from lactation consultant. For lip-tie, Kotlow lip-tie classification was recorded. For tongue-tie, Coryllos tongue-tie classification was recorded.	Lip-tie: 224 (100) Tongue-tie: 224 (100) Had a maxillary and lingual frenulum release: 13 (5.8) Had only a lingual release: 211 (94.2)	NR	NR

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
		a prior frenulum release at a different institution, infants who did not continue breastfeeding, or did not have a maxillary frenulum classified		maxillary frenotomy)	(6.3) (n=224 infants receiving lingual frenotomy or lingual and maxillary frenotomy)	Mother-infant feeding symptoms Cracked/flattened/creased/bleeding nipples: 97 (43.3) Pain: 169 (75.4) Inefficient/poor feeding/poor latch: 185 (82.6) Coryllos lingual frenulum classification Lingual frenotomy only (n=207), mean: 2.48 Maxillary+lingual frenotomy (n=13), mean: 3.38 Kotlow upper lip-tie classification Lingual frenotomy only (n=170), mean: 2.22 Maxillary+lingual frenotomy (n=8), mean: 3.33	Ankyloglossia type NR		
Wakhanrittee, 2016[80]	Thammasat University	Inclusion: Infants born in Thammasat	>24 hours: 267 (81.41)	Male: 194 (59.15)	NR	All infant-mother pairs were assessed	Tongue-tie: 328 (100)	Exclusive breast-	Numbers of children in

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
Thammasat University Hospital	Hospital from January 2014 to March 2015 in Thailand	University Hospital, <1 month, tongue-tie confirmed by pediatric surgeon, and presence of breastfeeding problem Exclusion: Infants or mothers with contraindications for breastfeeding, emergency conditions, or severe critical illness	≤24 hours: 61 (18.59) Median: 50 hours (IQR: 29–120)	Female: 134 (40.85)		for breastfeeding problems by postpartum nurses or lactation clinic nurses and referred to pediatric surgeons to re-assess the problems and assess the infant for tongue-tie. Frenotomy was performed in infants who had both tongue-tie and breastfeeding problems. Maternal problems Maternal nipple pain during the infant's suckling: 292 (89.02) Nipple trauma/sore nipples from the infant's suckling: 93 (28.35) The infant could not suck: 28 (8.54) The infant could not form an appropriate seal while suckling and sucked in air: 151 (46.04)	NR	feeding: 328 (100) per protocol	family 1: 172 (52.44) 2: 114 (34.76) 3: 34 (10.37) ≥4: 8 (2.43)

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
						The infant's tongue could not reach the areola: 238 (72.56) Nipple pain score, numeric pain scale (0=no pain; 10=highest pain), median (IQR): 5 (3–7) No pain (pain score = 0): 36 (10.98) Mild pain (pain score = 1–3:) 56 (17.07) Moderate pain (pain score = 4–6): 149 (45.43) Severe pain (pain score = 7–10:) 87 (26.52) Nipple characteristics Inverted nipple: 7 (2.13) Short nipple: 83 (25.30) Everted/normal nipple: 238 (72.56) Nipple sensation during suckling			

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
						No latch on: 9 (2.74) Tongue touch at nipple: 231 (70.43) Tongue touch at areola: 88 (26.83) Severity of tongue-tie Severe: 142 (43.29) Moderate: 180 (54.88) Mild: 6 (1.83)			
Wallace, 2006[81] NR	Lactation consultants referred to ENT consultants between August 2003 and February 2005 at the Mid Yorks NHS Trust, England	Inclusion: Infants who underwent tongue-tie division for feeding difficulties[i] Exclusion: NR	11.7 days (10.8) Median: 10 days; range 2–31 days	Male: 8 (80) Female: 2 (2)	NR	Poor latch: 9 (90) Sore nipples: 6 (60) Continual feeding cycles: 5 (50)	Tongue-tie: 10 (100) NR	Breast-feeding only: 3 (30) Breast-feeding and bottle feeding with breast milk: 4 (40) Breast-feeding and bottle feeding with formula: 1 (10) Cup feeding with expressed breast milk: 2 (20)	NR

Author, Year Funder	Recruitment Setting	Inclusion/ Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/ Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N (%)	Feeding Method N (%)	Other
Wen, 2022[82] None	Otolaryngology service (Illinois); mother-infant dyads underwent evaluation and breastfeeding education by a lactation consultation prior to evaluation at the service	Inclusion: Mothers who were actively breastfeeding, infant <4 months of age with ankyloglossia; at least 35 weeks gestation and no comorbidities Exclusion: Babies born ≤34 weeks gestation or with comorbidities	Range: 7 days to 4 months	Male: 22 (53.7) Female: 19 (46.3)	NR	Conservative breastfeeding education by lactation consultants over at least 3 visits must be documented as ineffective before infants were evaluated for frenotomy. Ankyloglossia was evaluated using the Coryllos scale, and frenulum length and distance to base and tip of the tongue were measured. LATCH scale, pre-procedure, mean (SD) 5.5 (2.9)	Underwent lingual frenulum release: 41 (100) Underwent maxillary labial frenulum release: 3 (7.3) NR if any infants with lip-tie did not undergo release NR	Per protocol, breastfeeding: 41 (100) Supplemental bottle feeding is possible but NR	NR

Notes: [a] Female N is noted as 100 in table 1, but 101 in the text. Assuming 143 male infants is correct, 101 females is correct.
[b] Data abstracted for surgery group only.
[c] Demographics, except age and gender, are for the 36 patients who responded to follow-up.
[d] 43 infants received multiple frenotomies.
[e] Based on who was seen at clinic, not final sample who were assessed for ties (n=618).
[f] Study included infants and children, but only data for infants are abstracted here.
[g] Demographic data was only provided for the 52 infants who successfully complete follow-up.
[h] Hospital standards for timing on division surgery changed in 2011. The 2 groups were those who were recommended division before 7 days of life (2008) and those recommended division after 7 days of life (2011).
[i] Data are for those reached at follow-up (10/11).

Abbreviations: aHATTLFF = Abbreviated Hazelbaker Tool for Lingual Frenulum Function; approx. = approximately; ASA I=American Society of Anesthesiologists [physical classification system,] one; CPT = Current Procedural Terminology; ENT = ear, nose, and throat; EMR = electronic medical records; g/d = grams per day; GP = general practitioner; ICD-10=International Classification of Diseases, 10th Revision; IQR = interquartile range; HATLFF = Hazelbaker Assessment Tool for Lingual Frenulum Function; LATCH = Latch, Audible swallowing, Type of nipple, Comfort, Hold; N = number; NA = not applicable; NR = not reported; SD = standard deviation; SQ = safety question; U.K. = United Kingdom; U.S. = United States.

Table C-6. Single-arm studies examining frenotomy/frenectomy with scissors: SQ intervention characteristics

Author, Year	Intervention N	Follow-up N	Provider/Setting	Anesthesia/ Procedure Duration	Other Therapy	Lactation Consult	Follow-up Time	Other
Akbari, 2023[37]	Frenotomy with scissors for 99% of sample (N=471), unclear procedure used for those not completed with scissors 474	474	Lactation consultants who were also midwives/hospital (at birth admission)	NR/NR	Nonsurgical management with a midwife or lactation consultant was provided to 40 of the infants with ankyloglossia who also received frenotomy	49% were diagnosed by lactation consultations; frenotomies were completed by lactation consultants; and nonsurgical management was provided by a midwife or lactation consultant to 40 infants with ankyloglossia who also received frenotomies.	Unclear, once discharged from hospital, follow-up was not recorded in clinical files; adverse events reported post-frenotomy throughout the study target years	NA
Amir, 2005[38]	Frenotomy with scissors 66 (35 received division)	46 (35 who received division)	NR/4 hospital ward; 31 breastfeeding clinic	None/NR	NR	NR but infants most commonly referred for tongue-tie assessment by 1 of the hospital lactation consultants; lactation consultants conducted structured interview with the mother by	Mean: 26 weeks (range 12–46, median 24)	NA

Author, Year	Intervention N	Follow-up N	Provider/Setting	Anesthesia/ Procedure Duration	Other Therapy	Lactation Consult	Follow-up Time	Other
						telephone at 3 months.		
Argiris, 2011[39]	Tongue division with scissors 46	46	ENT consultant or lactation consultant/ pediatric ward of hospital	No anesthetic or analgesia was used/NR	NR but 44 mothers felt they had been given sufficient support and advice about breastfeeding prior to the operation and that difficulties were experienced despite this advice	Infants were diagnosed and treated by a lactation nurse consultant or ENT consultant. Audit questionnaire was completed by lactation consultant on admission, immediately after the procedure, and 6 weeks post-procedure.	6 weeks	NA
Ballard, 2002[40]	Frenuloplasty with scissors 123	NR	NR/NR	Clear use NR, but suggested anesthesia is required for infants over 4 months of age but not for infants in early infancy/NR	NR	NR but outpatient participants were recruited from lactation center.	Approximately 3 days, although results also mention data from 5 days after frenuloplasty	NA
Barberá-Pérez, 2021[41]	Frenotomy [a] 33	33	NR/hospital	NR/NR	NR	NR	1 month	NA
Benoiton, 2016[42]	Frenotomy with scissors 34	33	Pediatric otolaryngologist/ ENT outpatient clinic	None were prescribed; 1 mL of sucrose was given via a 1 ml syringe to settle the baby post-procedure if required/NR	Parents were given a handout of stretches to perform for 2 weeks post-frenotomy	Patients were referred by lactation consultants. Provided guidance after procedure and followed up	2 weeks	NA

Author, Year	Intervention N	Follow-up N	Provider/Setting	Anesthesia/ Procedure Duration	Other Therapy	Lactation Consult	Follow-up Time	Other
						within 24 hours after the procedure.		
Bhandarkar, 2022[43]	Frenotomy (clipping and dividing) 599	194	Pediatric surgical consultant or breastfeeding midwife/clinic in children's hospital	None/NR	Before a referral for frenotomy, infants were required to be examined by a lactation consultant and to attempt conservative methods to promote latching and breastfeeding	Infants were examined by a lactation consultant before being approved for frenotomy.	Up to 1 year. Average period of 6 month; range 2–10 months	NA
Blenkinsop, 2003[45]	Frenulotomy with scissors 21	20	Pediatric and neonatal surgeon or a lactation consultant trained by pediatric and neonatal surgeon/hospital	NR/ NR; "very quick"	Before frenulotomy, midwives and lactation consultants offered help to maximize attachment to the breast	Before frenulotomy, midwives and lactation consultants offered help to maximize attachment to the breast. If no improvement, were referred to treatment.	Period from treatment to data collection varied	NA
Dollberg, 2014[48]	Frenotomy with scissors 244	244	One of 2 authors (neonatologist or pediatric dentist)/ at bedside (presumably hospital) and as an office procedure	Not used/NR	NR	Most infants were referred by lactation consultants who evaluated them for other possible causes of breastfeeding difficulties.	6 months	NA
Ferrés-Amat, 2017[50]	Frenotomy with Metzenbaum	88	NR/unclear, Suction Pathology	None reported/NR	Infants went through 3 stages of	Lactation specialist	Unclear. Possibly	NA

Author, Year	Intervention N	Follow-up N	Provider/Setting	Anesthesia/ Procedure Duration	Other Therapy	Lactation Consult	Follow-up Time	Other
	dissecting scissors 88		Unit (CELERE) within a hospital		treatment; in group 1 (N=171) mothers received breastfeeding sessions (BFS) to correct position while breastfeeding (N=171, successful for N=33); in group 2 (N=50), if ineffective sucking continued after 3 sessions of breastfeeding, the infant was referred to the service of myofunctional therapy (MFT) involving stimulation of rooting and sucking reflexes through extra- and intraoral exercises in 30-minute sessions during one month; group 3 (N = 88) involved frenotomy	referred infants to the MFT service; possibly responsible for the BFS education.	immediately after	
Geddes, 2008[11]	Frenulotomy with scissors 24	NR; 6 mothers reported milk production at 6 to12 days after frenulotomy	Pediatric surgeon/ hospital	NR/NR	NR	Lactation consultants informed the mother about the study and assessed the feed using the LATCH system. The same consultant assessed the	6 to 12 days	NA

Author, Year	Intervention N	Follow-up N	Provider/Setting	Anesthesia/ Procedure Duration	Other Therapy	Lactation Consult	Follow-up Time	Other
						mother for pre- and post-frenulotomy breastfeeds.		
Griffiths, 2004[53]	Frenotomy with scissors 215	215	General neonatal and pediatric surgeon/unclear appears to be hospital	Not used/NR	All mothers had been given support by a midwife, health visitor, infant feeding advisor, or lactation consultant prior to procedure but continued to experience difficulties	All mothers had been given support by a midwife, health visitor, infant feeding advisor, or lactation consultant prior to procedure. Lactation consultant input was not usually available after frenotomy.	3 months	NA
Hong, 2010[58]	Frenotomy with scissors 341	341	NR (possibly clinician at clinic)/outpatient pediatric otolaryngology clinic	Topical 20% Benzocaine/NR	Conservative intervention included assistance with lactation consultants mainly involving trials with different positioning techniques; 80% of cases had conservative intervention with no improvement	In infants with posterior tongue-tie 15 (80)	NR	NA
Illing, 2019[59]	Frenotomy with scissors 197	175	General practitioner/ Tongue-tie clinic	Lignocaine gel. For Kotlow 1-2 cases, frenulum was injected with lignocaine and	NR	Ankyloglossia was confirmed via consultation with a GP and	Mean: 23 days (SD: 12 days); median 20 days	Parents were recommended to perform tongue elevation

Author, Year	Intervention N	Follow-up N	Provider/Setting	Anesthesia/ Procedure Duration	Other Therapy	Lactation Consult	Follow-up Time	Other
				adrenaline by dental syringe/NR		a lactation consultant.		stretch exercises prior to feeding at least 4 times daily for 7 days
Masaitis, 1996[61]	Frenotomy with scissors 36	36	Pediatrician/NR	None used/NR	NR	NR	3 months	NA
McKenna, 2024[62]	Frenotomy (clipping) 86	NA	NR/Hospital	NR/NR	NR	Clinical correlation of impact on feeding was obtained by observation of breastfeeding by certified lactation consultants.	NA; no follow-up	NA
Mettias, 2013[63]	Frenotomy with scissors 63	36	NR/outpatient hospital ENT clinic	Lignocaine gel 2%/NR	NR	NR	NR	NA
Miranda, 2010[64]	Frenulotomy with scissors 62	51	Surgeon/outpatient department	None used/30 seconds	NR	Unclear, all participants were resistant to initial lactation consultant management.	2 weeks	NA
Muldoon, 2017[65]	Frenotomy with scissors 98	89	Pediatrician or pediatric surgeon: 2 (2) General practitioner: 3 (3) Specialist oral ENT/surgeon: 1 (1)/ Health care clinic/GP practices	Varied; no analgesia or 5% lignocaine oral gel, topical local anesthetic gel (in infants >3 months), 24% sucrose solution in younger babies, or oral sucrose at >8 weeks prior to the procedure/NR	NR, probably varied	Lactation consultants recommended frenotomy for 30 (31%) of the participants. Possible help from lactation consultants, public health	1 month	NA

Author, Year	Intervention N	Follow-up N	Provider/Setting	Anesthesia/ Procedure Duration	Other Therapy	Lactation Consult	Follow-up Time	Other
						nurses, and midwives prior and post-frenotomy.		
Narsat, 2022[66]	Frenotomy with scissors 67	67	Pediatric surgeons/ Clinical conditions	None used/NR	All mothers were given proper breastfeeding education and educational materials	NR	90 days	NA
O'Callahan, 2013[67]	Frenotomy with scissors 299	157	Pediatrician/primary care practice	Topical 20% benzocaine/NR	NR	Some referrals were from lactation consultants. Breastfeeding consultation prior to the frenotomy consultation was sought by 98% of respondents.	Varied; as much as 4 years, 5 months	NR
Pransky, 2015[69]	Ankyloglossia and/or upper-lip-tie releases with scissors 491	491	Otolaryngology physician assistant/ ankyloglossia outpatient clinic	NR/NR	NR; after procedure encouraged to see lactation consultants or nurses that specialized in breastfeeding, if problems persisted	NR; after procedure encouraged to see lactation consultants or nurses that specialized in breastfeeding, if problems persisted.	Immediately after procedure	NA
Ramoser, 2019[70]	Frenotomy with scissors 295	126	NR/clinic at a medical university	Children younger than 1 year were not given anesthetic/NR	NR	NR	4 to 6 weeks	NA
Ridgers, 2009[71]	Frenotomy by snip (no other	220	Surgeon/hospital lactation and	None reported/NR	NR	Clinic was focused on lactation and	4 weeks	NA

Author, Year	Intervention N	Follow-up N	Provider/Setting	Anesthesia/ Procedure Duration	Other Therapy	Lactation Consult	Follow-up Time	Other
	details provided) 220		breastfeeding clinic			breastfeeding, counselors provided advice and assistance. If advice was unsuccessful and tongue-tie was identified as the problem the child was referred for surgical assessment. After the procedure, participants were contacted by the breastfeeding counselor or an administrative assistant and invited to participate in an interview.		
Sethi, 2013[72]	Frenotomy with iris scissors 85	52	"Senior authors" type NR/ENT outpatient clinic	None/NR	NR	20 referrals were from lactation consultants.	At least 5 months	NA
Siggard, 2022[73]	Frenotomy[a] 230	163	ENT specialist/ Private ENT clinic	NR/NR	NR	NR	1 year or less; all were contacted in April 2020 and procedures were done between April	NA

Author, Year	Intervention N	Follow-up N	Provider/Setting	Anesthesia/ Procedure Duration	Other Therapy	Lactation Consult	Follow-up Time	Other
							2019 and April 2020	
Srinivasan, 2019[75]	Frenotomy with scissors 30	30	Physician/hospital based breastfeeding clinic	NR/NR	Lactation counseling and education were given as part of the general care	Lactation counseling and education were given as part of the general care.	14 days	NA
Srinivasan, 2006[76]	Frenotomy with scissors 27	25	NR/breastfeeding clinic	NR/NR	Assistance from lactation consultant or physician in breastfeeding education and latch adjustment as part of the clinic's breastfeeding program	Mothers were seen by a lactation consultant or physician trained in lactation before the administration of the pain questionnaire, during, and after the procedure, and given latch adjustment and general breastfeeding education.	3 months	The 2 dyads lost to follow-up were known to have stopped breastfeeding
Todd, 2015[77]	Frenotomy after 7 days (2011)[a] 144 Frenotomy before 7 days (2008)[a] 115	Frenotomy after 7 days (2011)[a] 144 Frenotomy before 7 days (2008)[a] 115	NR/Hospital	None used	NR	NR	NR	NA
Toner, 2014[78]	Frenotomy[a] 55	25	Otolaryngologists/ main hospital campus and	Topical anesthesia with licocaine was used by some practitioners/NR	NR	NR	A few months to 6 years post-treatment	NA

Author, Year	Intervention N	Follow-up N	Provider/Setting	Anesthesia/ Procedure Duration	Other Therapy	Lactation Consult	Follow-up Time	Other
			suburban satellites					
Towfighti, 2022[79]	Frenotomy with scissors 224	224	Pediatric otolaryngologist/otolaryingology department	NR/NR	Mothers were counseled on breastfeeding techniques and observed by a certified lactation consultant during breastfeeding and given handouts on "suck training"; additional professionals (speech-language pathologists, occupational therapists, or chiropractors) were consulted for certain infants if needed	Before procedure, all infants received a breastfeeding evaluation from an in-house lactation consultant. After procedure, mothers were observed while breastfeeding by a lactation consultant.	1 week; those who could not attend 1 week attended at 2 weeks	NA
Wakhanrittee, 2016[80]	Frenulotomy with metzenbaum scissors 328	246	Pediatric surgeons/hospital	2% sterile lidocaine jelly was applied at the lingual frenulum/NR	NR	NR, were assessed for breastfeeding problems by the in-hospital postpartum nurses or the lactation clinic's nurses or pediatricians.	3 months	NA
Wallace, 2006[81]	Frenotomy with scissors 11	10	ENT consultant/outpatient clinic	None/NR	NR	Infants were identified for inclusion by lactation consultants.	3 to 20 months (median = 10 months)	NA

Author, Year	Intervention N	Follow-up N	Provider/Setting	Anesthesia/ Procedure Duration	Other Therapy	Lactation Consult	Follow-up Time	Other
Wen, 2022[82]	Frenotomy with scissors 41	41	NR/NR	50/50mix of 1% lidocaine and oxymetazoline was applied before procedure; a gauze sponge soaked in lidocaine with oxymetazoline solution was applied for 5 minutes after the procedure./~5 minutes	All dyads went through breastfeeding education with lactation consultants for at least 3 visits with documented ineffectiveness before frenotomy	Before evaluation by the otolaryngology service, the dyads underwent evaluation and breastfeeding education by a lactation consultant.	1 month	NA

Notes: [a] Method of frenotomy unclear; probably with scissors.

Abbreviations: ENT = ear, nose, and throat; GP = general practitioner; LATCH = Latch, Audible swallowing, Type of nipple, Comfort, Hold; N = number; NA = not applicable; NR = not reported; SD = standard deviation; SQ = safety question.

Table C-7. Single-arm studies examining frenotomy/frenectomy with scissors: SQ results

Author, Year	Study Reports No Harms Occurred	Reported Harms
Akbari, 2023[37]	NA	Adverse events: 21 (4.4) Readmissions to hospital (for ongoing feeding difficulties): 5 (1.1) Bleeding: 38% Ulcers: 33% Swelling: 5% Irritability: 19% Scar tissue:38% Later states "The current study revealed that frenotomy procedures led to adverse effects in an average of 6.5% of infants in the Central Australian population"
Amir, 2005[38]	All mothers were asked about any problems after the tongue-tie release; no problems were reported.	NA
Argiris, 2011[39]	NA	52% reported blood loss during procedure 3 (6.5%) required repeat procedure for further division No other significant adverse events occurred
Ballard, 2002[40]	There were no complications related to the procedure.	NA
Barberá-Pérez, 2021[41]	NA	No medium- or long-term complications of the procedure were recorded. Only 1 case (1/33; 3%) attended consultation for irritability 24 hours after frenotomy, which was self-limited after some days.
Benoiton, 2016[42]	NA	Two frenotomies were revised at 2 weeks and at several months. "No complications occurred."
Bhandarkar, 2022[43]	NA	Intraoperative complications: 0 Recurrence needing repeat frenotomy: 4 (0.66)
Blenkinsop, 2003[45]	No post-treatment complications were reported.	NA
Dollberg, 2014[48]	The procedure itself was regarded by both the mothers and the physician as essentially nontraumatic, with minimal bleeding or pain and without noticeable complications.	NA
Ferrés-Amat, 2017[50]	There were no surgical complications.	NA

Author, Year	Study Reports No Harms Occurred	Reported Harms
Geddes, 2008[11]	There were no complications of frenulotomy reported. Most infants cried briefly after the frenulotomy; however, they showed no signs of distress after a breastfeed immediately after the procedure.	NA
Griffiths, 2004[53]	NA	Increased cry after division: 128 (60) Of these: Cried for 5 seconds or less: 56 (44) Cried for 20 seconds or less: 183 (85) Cried for more than 1 minute: 2 (1) No bleeding: 84 (38) A few drops of blood: 113 (52) "Small amount" of blood: 18 (8) "Ulcer" under tongue for more than 48 hours: 4 (2); all 4 healed spontaneously "Sore" for more than 24 hours: 1 (0.5) Brown posset due to swallowed blood: 1 (0.5) There were no infections, bleeding problems, feeding problems, or serious complications such as damage to the tongue or submandibular ducts
Hong, 2010[58]	NA	"All patients tolerated the procedure well and there were no reported complications." Revision procedures Anterior: 12 (3.7) Posterior: 4 (21.1)
Illing, 2019[59]	NA	There were no life-threatening or persistent complications at time of follow-up reported. One previously breastfeeding infant required 2 weeks of syringe feeding. One parent reported worse pain and latch difficulties at follow-up. Infant unsettled or had swelling under the tongue for 1-3 days: 8 (4.1) Frenulum reattachment: 3 (1.5) Feeding deteriorated: 1 (0.5)
Masaitis, 1996[61]	There were no complications nor any excessive bleeding from any of the frenotomies.	NA
McKenna, 2024[62]	There were no complications reported for any patients in either group undergoing frenotomy.	NA
Mettias, 2013[63]	NA	No complications: 32 (88.9) Distressed or discomfort: 2 (5.6)

Author, Year	Study Reports No Harms Occurred	Reported Harms
		Mild bleeding on the day of surgery that stopped spontaneously: 1 (2.8) Ulceration: 1 (2.8)
Miranda, 2010[64]	Frenulotomy was performed in 62 neonates, without any complications.	NA
Muldoon, 2017[65]	NA	Tongue-tie reoccurred: 2 (2)
Narsat, 2022[66]	We had no complications in the frenotomy procedure.	NA
O'Callahan, 2013[67]	NA	Most respondents reported no complications or negative side effects resulting from the frenotomy (94%). Cauterization with silver nitrate for persistent oozing: 3 (1)
Pransky, 2015[69]	All patients who underwent tongue-tie and/or upper-lip-tie release procedures had no complications.	NA
Ramoser, 2019[70]	NA	99% (162/164) parents reported no complications. One mother reported infant refusal to drink from breast or bottle for 2 hours after the procedure. One mother reported infant child having fever for 1 day (probably out of review age range).
Ridgers, 2009[71]	"None reported adverse effects, most commenting that the recovery of the infant was uneventful."	NA
Sethi, 2013[72]	No complications were reported.	NA
Siggard, 2022[73]	No complications during the procedure were reported according to the patient files nor reported by the parents.	NA
Srinivasan, 2019[75]	NA	8 parents (22.2%) noted bleeding while doing the stretching exercises at home at Day 2 and Day 7. By day 14, 1 parent (2.78%) noted bleeding at home. No hospital visits or other medical complications related to bleeding episodes. All infants were stable during follow-up visits.
Srinivasan, 2006[76]	No complications were noted during or after frenotomy. There were no extended incidents of bleeding requiring active management, no infant fever, and no hospital admissions.	NA
Todd, 2015[77]	NA	There were no significant complications.

Author, Year	Study Reports No Harms Occurred	Reported Harms
		Minor bleeding occurred in all cases and a healing ulcer was noted at around 2–4 days post-division.
Toner, 2014[78]	There were no reported complications with any of the procedures.	NA
Towfighti, 2022[79]	NA	No infants in this cohort had any immediate post-procedural complications (i.e., infection, bleeding, frenula reattachment). Of 211/224 infants who received lingual frenulum release only, 4 underwent revision procedures.
Wakhanrittee, 2016[80]	NA	"During the procedure and in the 24 h after that, there were no complications including no significant bleeding requiring active or emergency management or a Wharton's duct injury." 1 week follow-up Minimal white patch at the frenulotomy site: 10.06 % No infection reported. No complications mentioned at 3 months in the paper. Unclear if none occurred.
Wallace, 2006[81]	There were no reported complications of the procedure.	NA
Wen, 2022[82]	NA	Among all 41 infants, no post-surgical complications were reported. Notable subjective postoperative pain response (mother reported): 10 (24.4) Duration 1 day: 8 infants (19.5) 2 days: 1 infants (2.4) 7 days: 1 infants (2.4)

Abbreviations: NA = not applicable; SQ = safety question.

Table C-8. Single-arm studies examining frenotomy/frenectomy with laser: SQ study characteristics

Author, Year Registration Funding ROB	Recruitment Setting	Inclusion/Exclusion Criteria	Age Mean (SD)	Gender, N (%)	Race/Ethnicity N (%)	Indications, N (%)	Tie Type, N (%) Ankyloglossia Type, N (%)	Feeding Method, N (%)	Other
Dell'Olio, 2022[47] NR No sources of funding	Aldo Moro University of Bari (Italy) Unit of Neonatology	Inclusion: Ankyloglossia according to Coryllos' criteria with indication for lingual frenotomy as determined by the ATLFF, corrected gestational age >40 weeks Exclusion: Infants aged >12 weeks, infants with craniofacial abnormalities, syndromes, or neurological abilities impairing sucking ability	47.2 days (20.2)	Male: 30 (53.6) Female: 26 (46.4)	NR	Coryllos' classification Anterior ankyloglossia Type 1: 10 (17.9) Type 2: 18 (32.1) Posterior ankyloglossia Type 3: 23 (46.4) Type 4: 2 (3.6) Mean ATLFF function score (SD): 7.8 (1.9)	Tongue-tie: 56 (100) Lip-tie: 0 (0) Anterior: 28 (50) Posterior: 28 (50)	Exclusively breastfed: 34 (60.7) Breastfeeding supplemented with formula: 22 (39.3)	NR
Freeman, 2022[51] NR NR	Identified retrospectively at the University of Texas Medical Branch Hospital	Inclusion: Aged <3 months with isolated lip-tie Exclusion: Aged > three months; concomitant tongue-tie	4.9 weeks (3.9) Range 1–12 weeks	NR	NR	Breastfeeding difficulties	Lip-tie: 7 (100) Tongue-tie: 0 NR	Assuming 100% were breastfeeding based on indication, but prevalence of feeding methods was not reported	NR
Ghaheri, 2017[52] NCT02642133 No funding	Community-based otolaryngology care center	Inclusion: Currently breastfeeding, younger than or equal to 12 weeks	Baseline, mean: 4.4 weeks (3.6)	Male: 56%	White/Caucasian: 86%	Infants were initially evaluated by lactation consultant before surgical referral. A head and neck examination was	Surgical treatment type, N (%) Tongue- and lip-tie: 178 (75)	Breastfed: 237 (100) per protocol	NR

Author, Year Registration Funding ROB	Recruitment Setting	Inclusion/Exclusion Criteria	Age Mean (SD)	Gender, N (%)	Race/Ethnicity N (%)	Indications, N (%)	Tie Type, N (%) Ankyloglossia Type, N (%)	Feeding Method, N (%)	Other
	(US, 2014-2015)	of age and older than or equal to 37 weeks gestational age Exclusion: Infants with life-threatening comorbid conditions, previous treatment for tongue-tie or lip-tie by another provider, multiple births, and mothers with previous breast surgery or insufficient glandular tissue				performed, examining anatomical features, and a sucking evaluation. The ATLFF staging system was used for formal scoring. Kotlow upper lip-tie classifications Class 1: 0 (0) Class 2: 2 (1) Class 3: 109 (46) Class 4: 126 (53) Coryllos tongue-tie classifications Type 1: 12 (5) Type 2: 40 (17) Type 3: 76 (32) Type 4: 109 (46) Maternal complaints, % Poor latching: 81% Falls asleep while attempting to nurse: 73% Creased, flattened, or blanched nipples after nursing: 68% Gumming or chewing of nipple when nursing: 67% Poor or incomplete breast drainage: 60% Slides off nipple when attempting to latch: 60% Severe pain when infant attempts to latch: 59%	Lip-tie only: 1 (0.4) Tongue-tie only: 58 (25) Isolated posterior tongue-tie: 78%		

Author, Year Registration Funding ROB	Recruitment Setting	Inclusion/Exclusion Criteria	Age Mean (SD)	Gender, N (%)	Race/Ethnicity N (%)	Indications, N (%)	Tie Type, N (%) Ankyloglossia Type, N (%)	Feeding Method, N (%)	Other
						Cracked, bruised, or blistered nipples: 49% Reflux symptoms: 45% Unable to hold a pacifier in mouth: 40% Poor weight gain: 32% Colic symptoms: 24% Bleeding nipples: 24% Plugged ducts: 21% Mastitis or nipple thrush: 14% Infected nipples or breasts: 6%			
Hand, 2020[55] NR NR	Referrals to private dental practice	Inclusion: Currently breastfeeding; infant <12 weeks of age and >37 weeks gestational age; functional restriction of movement of the tongue, upper lip, cheeks Exclusion: Patients who did not complete the outcome survey within the 1 month follow-up period; infants who had previously been treated for oral ties by another provider	Mean age: 43 days	Male: 67 (51) Female: 65 (49)	NR	Infants were referred by lactation consultants to be evaluated by surgeon at a private dental practice. Evaluation included examination of the mouth, frenulum, sucking, and tongue movement. The Bristol Tongue Assessment Tool and Tongue-tie and Breastfed Babies Assessment Tool was also used. Complaints, % Difficulty in achieving a good latch: 88 Reflux (clicking, swallowing air while nursing): 74 Painful latching of infant onto the breast: 68 Milk leaking out sides of	Surgery type received Lip and tongue: 99 (75) Tongue: 24 (18.2) Tongue, lip, and buccal: 7 (5.3) Lip only: 2 (1.5) NR	Breastfeed: 132 (100)	NR

Author, Year Registration Funding ROB	Recruitment Setting	Inclusion/Exclusion Criteria	Age Mean (SD)	Gender, N (%)	Race/Ethnicity N (%)	Indications, N (%)	Tie Type, N (%) Ankyloglossia Type, N (%)	Feeding Method, N (%)	Other
						mouth while feeding: 67 Falls asleep at the breast while attempting to nurse: 63 Slides off the breast while feeding: 49 Poor or incomplete drainage from breast: 46 Gumming or chewing of the nipples while feeding: 46 Creased, cracked or blanching of nipples: 46 Short sleep episodes (feeding every 1–2 hours): 38 Waking congested in morning: 38 Unable to keep pacifier in mouth: 36 Poor weight gain: 29 Apnea—snoring, heavy noisy breathing: 24 Undersupply of breast milk: 21 Waking congested after nap: 21 Oversupply of breast milk: 20 Only sleeping when in upright position or in car seat: 18 Blocked ducts: 14 Mastitis: 9 Nipple thrush: 8 Infected nipples: 5			

Author, Year Registration Funding ROB	Recruitment Setting	Inclusion/Exclusion Criteria	Age Mean (SD)	Gender, N (%)	Race/Ethnicity N (%)	Indications, N (%)	Tie Type, N (%) Ankyloglossia Type, N (%)	Feeding Method, N (%)	Other
						Depression: 5 Abraded nipples: 4 Coryllos classification, N (%) Type 1: 6 (4.5) Type 2: 32 (24.2) Type 3: 37 (28.0) Type 4: 57 (43.2) Kotlow lip-tie classification, N (%) Type 1: 0 (0) Type 2: 1 (0.8) Type 3: 51 (38.6) Type 4: 80 (60.6) VAS Pain Score (mean, SD; min-max) : 4.6 (2.8; 0–10) BSES-SF Total Score (mean, SD; min-max) : 48.7 (11.3; 20–70) I-GERQ-R total score (mean, SD; min-max): 16.3 (6.1; 5–30)			
Hill, 2022[56] NR MGH Institute of Health Professions School of Nursing.	Dental office in the northeast region of the United States between November 2020 and February 2021	Inclusion: Infants younger than 7 months old, diagnosed with tongue-tie, and undergoing frenotomy. Mothers 18 years of age or older	9.1 weeks (7.7), range 1 week to 28.6 weeks	Male: 14 (56) Female: 11 (44)	Race White: 23 (92) More than 1 Race (Asian and White): 2 (8) Ethnicity Hispanic or Latino: 1 (4)	Infants were assessed by a dentist using Kotlow diagnostic criteria and the HATLFF. Decision on need for frenotomy was based on results of the measures and concerns reported by the mother. Kotlow tongue-tie score	Diagnoses Tongue-tie: 25 (100) Lip-tie: 19 (76) Ties corrected Tongue-tie: 25 (100) Lip-tie: 18 (72)	Breastfeeding: Bottle feeding: Mixed feeding:	Treatments unrelated to the study, in the past month: Received therapy to help with feeding: 9 (36)

Author, Year Registration Funding ROB	Recruitment Setting	Inclusion/Exclusion Criteria	Age Mean (SD)	Gender, N (%)	Race/Ethnicity N (%)	Indications, N (%)	Tie Type, N (%) Ankyloglossia Type, N (%)	Feeding Method, N (%)	Other
		Exclusion: Infant prematurity (<37 weeks gestation at birth) and anomalies that impact feeding (e.g., cleft lip or palate)			Not Hispanic or Latino: 23 (92) Not Answered: 1 (4)	1: 1 (4) 2: 11 (44) 3: 12 (48) 4: 1 (4)	 Ankyloglossia type NR		Received health care related to feeding issues such as visits for growth or feeding concerns, or reflux: 16 (64) Previously received a medical diagnosis of GERD: 3 (12) Diagnosis of GERD requiring treatment with acid-reducing medication: 1 (4)
Patel, 2019[68] NR No funding	Using CPT code 40,806 (incision of labial frenum), infants were identified at an academic, referral-based, pediatric otolaryngology practice (New York,	Inclusion: Infants younger than 60 days old with lip-tie but without tongue-tie Exclusion: Infants with syndromes, craniofacial anomalies, or neuromuscular disorders	25 days; range 4 to 51 days	Female: 59%	NR	Restrictive upper lip frenum, inserting into the gingiva, without concomitant ankyloglossia. Restrictive lip frenum was defined as being difficult to manually evert the upper lip or if mother reported frequent "rolling under" of the upper lip during breastfeeding. All children had a grade 3	Lip-tie: 22 (100) Tongue-tie: 0 NR	Breastfeeding: 22 (100) Supplemental bottle feeding: 59%	NR

Author, Year Registration Funding ROB	Recruitment Setting	Inclusion/Exclusion Criteria	Age Mean (SD)	Gender, N (%)	Race/Ethnicity N (%)	Indications, N (%)	Tie Type, N (%) Ankyloglossia Type, N (%)	Feeding Method, N (%)	Other
	U.S., 2015–2017)					or grade 4 upper lip Kotlow grade.			
Slagter, 2021[74] ISRCTN64428423 No funding	Private practice (Netherlands, 2017–2018)	Inclusion: Healthy infants younger than 6 months with breastfeeding problems Exclusion: Premature infants, twins, or already had a revision for tongue-tie or lip-tie, exclusively received formula, or did not seem to have oral restrictions	Age, (n, %) 0 months: 90 (51.4) 1 month: 49 (28) 2 months: 24 (13.7) 3 months: 12 (6.9)	Male/female ratio: 93:82	Ethnicity Dutch: 171 (97.7) Non-Western Immigrant: 4 (2.3)	Infants were examined by a doctor in dental surgery and an International Board Certified Lactation Consultant for oral signs and usual causes of breast or nipple pain, dermatosis infection, and vasospasm. Other oral examinations included reporting sucking blisters, shape of the palate, retrognathia, location of attachment of the frenula, blanched frenula with elevation, anatomical restriction of elicited lateral lingual movement (impaired transverse tongue reflex), abnormal floor of mouth elevation of the tongue, and presence of thrush. A sucking evaluation was done, consisting out of the notification of abnormal gum/lip pressure, cupping of the tongue against the finger, seal on the finger, and the nature of the sucking tongue movements. A score form was used to	Lip-tie: 175 (100) Tongue-tie: 175 (100) NR	Only breastmilk: 93 (53.1) Breastmilk by bottle: 63 (36) Breastmilk and Formula: 19 (10.9) Formula only: 0 (0)	Child First: 78 (44.6) Second 64: (36.6) Third 27: (15.4) Fourth or more: 6 (3.4) Child with reflux: 49 (28)

Author, Year Registration Funding ROB	Recruitment Setting	Inclusion/Exclusion Criteria	Age Mean (SD)	Gender, N (%)	Race/Ethnicity N (%)	Indications, N (%)	Tie Type, N (%) Ankyloglossia Type, N (%)	Feeding Method, N (%)	Other
						record different anatomical differences. Lip-tie (n, %) Type 1: 1 (0.5) Type 2: 47 (26.9) Type 3: 123 (70.3) Type 4: 4 (2.3) Tongue-tie (n, %) Type 1: 11 (6.3) Type 2: 58 (33.1) Type 3: 100 (57.1) Type 4: 6 (3.4) Sucking blisters (n, %) Yes: 144 (82.3) No: 31 (17.7) Palate (n, %) High: 137 (78.3) Flat: 38 (21.7) Two colored tongue (n, %) Yes: 133 (76) No: 42 (24)			

Abbreviations: ATLFF = Assessment Tool for Lingual Frenulum Function; BSES-SF = Breastfeeding Self-Effocacy Scale; CPT = Current Procedural Terminology; GERD = gastroesophageal reflux disease; HATLFF = Hazelbaker Assessment Tool for Lingual Frenulum Function; I-GERQ-R = Infant Gastroesophageal Reflux Questionnaire Revised; N = number; NR = not reported; ROB = eisk of bias; SD = standard deviation; SQ = safety question; U.S. = United States; VAS = Visual Analog Scale; wks=weeks

Table C-9. Single-arm studies examining frenotomy/frenectomy with laser: SQ Intervention Characteristics

Author, Year	Intervention	Enrollment N	Follow-up N	Provider/Setting	Anesthesia/Procedure Duration	Other Therapy	Lactation Consult	Follow-up Time	Other
Dell'Olio, 2022[47]	Frenotomy with laser	56	56	Oral surgeon/neonatology unit	Topical, local anesthetic EMLA® on both sides of frenulum and its insertion on the tongue and floor of mouth/NR	NR	NR	30 days	
Freeman, 2022[51]	Frenotomy with laser (Colorado Needle monopolar electrocautery)	7	7	NR/hospital	Local anesthesia or general anesthesia under a face mask/NR	NR	NR	1 to 6 weeks	NA
Ghaheri, 2017[52]	Frenotomy with laser	237	NR	ENT surgeon (principal investigator)/community-based otolaryngology care center	Topical anesthetic EMLA/NR	NR	Infants were initially evaluated by a lactation consultant before surgical referral.	1 month	NA
Hand, 2020[55]	Laser frenulum release (tongue, lip, buccal)	132	132	Dental surgeon/private dental clinic	None/NR	NR	All infants were initially evaluated by a community lactation consultant before surgical referral.	1 month	NA Not clear if study was conducted in Australia or Italy; presumably Italy
Hill, 2022[56]	Frenotomy with CO2 laser	25	25	Probably dentist/dental office	None/NR	NR	The dentist required an evaluation with a lactation consultant with an indication of functional impairment with feeding before the visit. 16 (64%) of sample consulted	5 minutes after frenotomy	NA

Author, Year	Intervention	Enrollment N	Follow-up N	Provider/Setting	Anesthesia/Procedure Duration	Other Therapy	Lactation Consult	Follow-up Time	Other
							with lactation consultant about feeding issues.		
Patel, 2019[68]	Frenotomy with fine-tip electrocautery	34	22	NR/pediatric otolaryngologic practice	Injection with 1% lidocaine and 1:100,000 epinephrine solution/NR	NR	NR	4 weeks	
Slagter, 2021[74]	Frenotomy with electrosurgery 174 patients received both a tongue-tie release and a frenotomy; not clear what remaining patient received	175	146	Doctor in dental surgery/private practice	Topical: xylocaine 5%/NR	NR	All participants had previously been seen by an International Board Certified Lactation Consultant and referred to the practice.	6 months	NA

Abbreviations: ENT = ear, nose, and throat; NA = not applicable; NR = not reported; SQ = safety question.

Table C-10. Single-arm studies examining frenotomy/frenectomy with laser: SQ Results

Author, Year	Study Reported No Harms Occurred	Reported Harms
Dell'Olio, 2022[47]	NA	No infections at the site of frenotomy were found. No recurrence of ankyloglossia. AEs during procedure C.R.I.E.S Scale: Each domain is rated 0–2 with 2 indicating higher pain intensity. During surgery, mean score (SD): 5.7 (0.5) Pain intensity raised significantly during procedure, mean difference = 5 points; *p*<0.001 Individual domains, N (%) High pitched and inconsolable cry: 47 (83.9) Easily consolable: 9 (16.1) Oxygen desaturation <95%: 0 (0) Heart rate increase of more than 20% of baseline: 30 (53.6) Heart rate increase of 10 to 20% of baseline: 26 (46.44) Grimaced facial expression: 56 (100) AEs After procedure C.R.I.E.S. score, mean (SD): 4.4 (1.1) High pitched, easily consolable cry, n (%): 54 (96.4) Heart rate increase <20%, n (%): 47 (83.9) Heart rate return to baseline, n (%): 9 (16.1) AEs 30 minutes post-procedure C.R.I.E.S. score, mean (SD): 0.7 (0.8) Other AEs during procedure Punctiform bleeding: 17 (30.4%) Carbonization of the irradiated site: 11 (19.6) No injuries to Wharton salivary ducts AEs, 7-day follow-up, N (%) Punctiform bleeding due to accidental trauma: 1 (1.8) Refusal of pacifier: 39 (69.9) Frequently awake: 15 (26.8) AEs, 30-day follow-up, N (%) All mothers reported behavior was not different compared to before the operation

Freeman, 2022[51]	All patients tolerated the procedure well without any surgical or anesthesia complications.	NA
Ghaheri, 2017[52]	NA	Received second lingual frenotomy for lack of improvement or regression of symptoms: 8 (3) No complications were reported following any procedure.
Hand, 2020[55]	Within this study, no complications were reported following any procedure.	NA
Hill, 2022[56]	NA	Behavioral State Pre-Frenotomy Crying: 2 (8) Not crying: 23 (92) Behavioral State Post-Frenotomy Crying: 14 (56) Not crying: 11 (44) Of the 2 infants crying pre-frenotomy, 1 stopped crying before the post-frenotomy measurement and 1 continued to cry.
Patel, 2019[68]	NA	Recurrence of lip-tie: 9% Local pain, mild: 64% Local pain, moderate: 18% Any local pain: 82% No other complications or significant bleeding
Slagter, 2021[74]	NA	Second lingual frenotomy within 1 months: 8 (4.6) Temporary hypergranulated tissue of the wound: 1 (0.7) Complications in motor and cognitive growth after 6 months: 0

Abbreviations: AE = adverse event; C.R.I.E.S= C.R.I.E.S. = Crying, Requires increased oxygen administration, Increased vital signs, Expression, Sleeplessness; N = number; NA = not spplicable; SD = standard deviation; SQ = safety question.

Table C-11. Single-arm studies examining frenotomy/frenectomy, method unclear: SQ study characteristics

Author, Year Registration Funding	Recruitment Setting	Inclusion/Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N(%)	Feeding Method N (%)	Other
Billington, 2018[44] NR Received No Funding	Tongue-tie clinic in the greater London area between May and July 2016[a]	Inclusion: Mothers referred to the clinic by a community midwife specializing in breastfeeding after a minimum of 2 assessment confirming tongue-tie requiring frenotomy Exclusion: Ankyloglossia recurrence after previous frenotomy	Median: 17 days, range 2–88	NR	NR	Breastfed infants with confirmed tongue-tie and difficulty with breastfeeding	Tongue-tie: 87 in complete follow-up dataset (100); 100 confirmed overall (100) Posterior: 50 (57) Anterior: 28 (32) Combined: 9 (11)	Unclear; 87 (100%) based on included sample; Bottle fed NR	NR
Braccio, 2016[46] NR NR	Referrals to tongue-tie clinic at Evelina London's Children's Hospital from October 2013 to September 2014. Retrospective review of charts with telephone survey[b]	Inclusion: Confirmed diagnosis of tongue-tie related breastfeeding difficulties after examination by an experienced midwife Exclusion: NR	Median: 4 months (range 2 weeks to 9 months)	NR	NR	Problems for woman (e.g., breast pain, cracked nipples): 118 (43.4) Frequent/long feeds: 116 (42.6) Shallow latch: 118 (43.4) Fussiness/restlessness: 102 (37.5)	Tongue-tie: 272 (100) NR	Exclusive breastfeeding: 58 (21.3) Exclusive or partially breastfed: 149 (54.8) Formula use: 52 (19.1) Unknown: 162 (60)	NR
Donati-Bourne, 2015[49]	Tongue-tie clinic at Birmingham	Inclusion: Infants referred to clinic for tongue-tie	Median: 28.5 days	Male: 48 (62.5)	NR	58 (100)—tongue-tie identified and/or breastfeeding	Tongue-tie: 58 (100%)	43 (74) had persisted in attempts to	NR

Author, Year Registration Funding	Recruitment Setting	Inclusion/Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N(%)	Feeding Method N (%)	Other
NR NR	Heartlands Hospital in Birmingham, U.K.	assessment between April 2013 and July 2013. Exclusion: No exclusion criteria were applied	(range 1–126)	Female: 22 (31.4)		difficulty; referred by midwife, health visitor, breastfeeding counselor, neonatologist, or general practitioner	NR	breastfeed at time of clinic attendance	
Hale, 2020[54] NR The New Zealand Ministry of Health	Surveillance of complications from frenotomies by the New Zealand Pediatric Service, done by sending reporting cards to 88% of all registered pediatricians between August 2016 and July 2018	Inclusion: Children younger than 1 year of age with complications attributable to, or arising from, any form of frenotomy for ankyloglossia Exclusion: NR	31.7 days; range 1–79[c]	Male: 63% Female: 37%[c]	European: 14 (88) Maori: 1 (6) Asian: 1 (6)[c]	Breastfeeding difficulties: 13 (81) Breast/nipple pain: 3 (19) Weight loss: 3 (19) Other ("unsettledness and jaundice," "fussy and windy," and "slow weight gain"): 3 (19) Unknown: 2 (13)[c]	Lip-tie: 8 (50) Two or more types: 8 (50)[c] Posterior: 2 (13) Anterior: 8 (50)[c]	Exclusively breastfed: 6 (38) Breast and bottle fed: 6 (38) Exclusively bottle fed: 4 (25)[c]	Had undergone previous frenotomy: 4 (25)[c]
Hill, 2022[57] NR MGH Institute of Health Professions School of Nursing	Pediatric dentist in an urban setting (NY)	Inclusion: Infants <7 months old , diagnosed with tongue-tie, and undergoing frenotomy; mothers had to be proficient in English	Infant age at baseline (n=102) <2 months: 82 (79.4) 2 to <4 months: 13 (12.8) 4 to <7	Male 57 (55.9) Female 45 (44.1)	Race American Indian/Alaskan Native: 1 (1) Asian: 1 (1) White: 86 (84.3) More than 1 race: 11 (10.8) Other: 2 (2)	Tongue-tie severity (Kotlow class) 2: 1 (1) 3: 92 (90.2) 4: 9 (8.8) Maternal symptoms Painful latching: 61 (59.8) Difficulty achieving	Tongue-tie: 102 (100) Lip-tie: 102 (100) Buccal tie: 35 (34.3) NR	Breastfeeding: 85 (83.3) Exclusively breastfeeding: 35 (34.2) Feeding plan Exclusive breastfeeding: 78 (76.5)	Maternal perceived change in feeding plans due to tongue-tie Yes: 25 (24.5) No: 73 (71.6) Unsure: 4 (3.9)

Author, Year Registration Funding	Recruitment Setting	Inclusion/Exclusion Criteria	Age Mean (SD)	Gender N (%)	Race/Ethnicity N (%)	Indications N (%)	Tie Type N (%) Ankyloglossia Type N(%)	Feeding Method N (%)	Other
		Exclusion: Prematurity (<37 weeks), anomalies of the head, face, or neck that may impair feeding, or comorbid conditions associated with feeding difficulty	months: 8 (7.8)		Not answered: 1 (1) Ethnicity Hispanic/Latino: 4 (3.9) Not Hispanic/Latino: 88 (86.3) Other: 6 (5.9) Unknown: 3 (2.9) Not answered: 1 (1)	successful latch: 50 (49) Gumming/chewing of the nipple while eating: 49 (48)		Exclusive formula feeding: 3 (2.9) Breast and formula feeding: 4 (3.9) Exclusive pumping of breastmilk: 12 (11.8) Other: 2 (2) Not answered: 2 (2)	
Lyudin, 2018[60] NR NR	Primary care practice (New Zealand, January–December 2013)	Inclusion: NR Exclusion: Missing data or patient was declined frenulotomy	Age at frenulotomy, N (%) <7 days: 102 (24.6) 1–2 weeks: 87 (21.0) 2–3 weeks: 40 (9.7) 3–4 weeks: 46 (11.1) 1–2 months: 76 (18.4) >2 months: 63 (15.2)	NR	NR	NR	NR NR	NR	NR

Notes: [a] Reported sample demographics are of the 87 mother/infants who completed follow-up; 13 were lost to follow-up and their demographics were NR.
[b] Data are only available for survey respondents.
[c] Sample includes only those who had a complication from frenotomy. Data on all frenotomies NR.

Abbreviations: N = number; NR = not reported; SD = standard deviation; SQ = safety question; U.K. = United Kingdom.

Table C-12. Single-arm studies examining frenotomy/frenectomy, method unclear: intervention characteristics

Author, Year	Enrollment N	Follow-up N	Provider/Setting	Anesthesia/Procedure Duration	Other Therapy	Lactation Consult	Follow-up Time	Other
Billington, 2018[44]	100	87	Unclear, clinician in clinic/tongue-tie clinic	NR/NR	Patients met with a midwife specializing in breastfeeding at least 2 times before referral to the clinic.	Unclear, community midwife specializing in breastfeeding referred mothers of these infants to the TTC.	3 months	NA
Braccio, 2016[46]	272	158	NR/tongue-tie clinic	NR/NR	Upon referral, every infant's breastfeeding was assessed by a midwife to ensure the issues were not related to position or attaching technique. After the procedure, infants and mothers met with a midwife or neonatal nurse again for further support on breastfeeding technique and positioning.	NR, referrals were made by health professionals including community midwives, breastfeeding consultants, GPs, health visitors, general pediatric consultants, and others. Midwives from the breastfeeding team assessed breastfeeding problems and after the procedure midwives or neonatal nurse provided support and advice on breastfeeding technique and positioning.	Varied; infants ranged in age between 2 weeks and 9 months (median 4 months).	NA
Donati-Bourne, 2015[49]	58	58	NR/outpatient neonatal surgery department	NR/NR	NR	Referred by midwife, health visitor, breastfeeding counselor, neonatologist, or general practitioner.	NR	NA
Hale, 2020[54]	16	16	Dentist: 5 (31) General practitioner: 2 (13) Lactation consultant: 2 (13) Otolaryngology surgeon: 2 (13)	NR/NR	NR	Lactation consultants diagnosed some cases and performed some procedures.	Varied	NA

			Pediatrician: 1 (6) Pediatric surgeon: 1 (6) Unknown: 3 (19)/ Private clinic: 8 (50) Public hospital: 3 (19) Private operating theater: 1 (6) Location unknown: 4 (25)					
Hill, 2022[57]	102	84	Pediatric dentist/ Pediatric dental clinic	NR/NR	Therapy prior to frenotomy from lactation support provider: 4 from chiropractor: 4 Therapy after frenotomy from lactation support provider: 12 (11.8) from chiropractor: 10 (9.8) from craniosacral therapy: 4 (3.9)	Pediatric dentist recommended follow-up with lactation consultant after procedure. Participants were primarily referred to the pediatric dentist either by a lactation support provider (n=37) or a pediatric primary care provider (n=23).	2 weeks	NA
Lyudin, 2018[60]	445	414	NR/primary care practice	NR/NR	NR	NR	NR. Unclear if follow-up was standard.	NA

Abbreviations: GP = general practitioner; NA = not applicable; NR = not reported; TTC = tongue-tie clinic.

Table C-13. Single-arm studies examining frenotomy/frenectomy, method unclear: SQ results

Author, Year	Study Reported No Harms Occurred	Reported Harms
Billington, 2018[44]	There were no cases of postoperative bleeding or recurrence in this cohort. It is possible that the remaining 13 mothers who were unable to be contacted may have experienced complications, ongoing difficulties feeding and, possibly, recurrence.	NA
Braccio, 2016[46]	NA	No cases of major bleeding, infection or ulceration were reported. One infant attended the clinic four times with persistent breastfeeding problems and required a total of 3 procedures. Two infants reattended the clinic once and underwent a second procedure. One infant reattended the clinic with persistent breastfeeding problems twice and underwent a second procedure, but no tongue-tie was found on the third visit.
Donati-Bourne, 2015[49]	No complications were reported at follow-up telephone consultation.	NA
Hale, 2020[54]	NA	23 complications were reported. Detailed information was provided for 16. Poor feeding: 7 (44) Apnea, ALTE/BRUE, or other breathing difficulties: 4 (25) Pain: 3 (19) Bleeding: 3 (19) Weight loss: 3 (19) Pallor/anemia: 2 (13) Excess scarring: 2 (13) Unsettledness: 1 Peripheral cyanosis: 1 Grayish black stools: 1 Ulcer: 1 Severe hypernatremia, hypothermia and 20% weight loss: 1
Hill, 2022[57]	NA	Complications from frenotomy Yes: 0 (0) No: 101 (99) Unsure: 1 (1)
Lyudin, 2018[60]	NA	An additional 132 unplanned visits from 111 individuals were recorded post-frenulotomy Reasons for visit Infection concerns: 4 (1.0)

		Bleeding: 1 (0.2) Continued poor feeding: 2 (0.4) Continued nipple pain: 2 (0.4) Concern that tongue-tie persisted : 122 (29.5) Repeat frenulotomy performed: 96 (23) More than 2 frenulotomies performed: 13 (3.1)

Abbreviations: ALTE/BRUE = Apparently Life-Threatening Event/Brief Resolved Unexplained Event; NA = not applicable; SQ = safety question.

Appendix D. Individual Study Risk of Bias Assessments

Table D-1. Risk of bias for randomized, controlled trials

Study, Year	Risk of bias arising from the randomization process	Risk of bias due to deviations from the intended interventions	Missing outcome data	Risk of bias in measurement of the outcome	Risk of bias in selection of the reported result	Overall Risk of Bias	Comments
Knight, 2023[31]	Low	High	Low	Some concerns	Low	High	Lack of sense of equipoise likely led to a very high proportion of comparator arm participants receiving the intervention; potential for bias from lack of blinding after first 20 participants.
Ghaheri, 2022[30]	High	Low	Low	Some concerns	Low	High	Potential risk of bias from randomization and measurement of outcomes.
Emond, 2014[29]	High	High	Low	Some concerns	Low	High	Potential bias from differences at baseline for 5 day outcomes; although outcome assessors were blinded, mothers were aware of intervention; additional concerns arising from early crossovers s prior to planned date at 5 days.
Berry, 2012[26]	Low	Low	Low	Low High for breastfeeding improvement	Low	Low for other outcomes High for breastfeeding improvement	
Buryk, 2011[27]	Low	Low	Low	Low	Low	Low	Breastfeeding outcomes were measured after crossover leading to high ROB.
Dollberg, 2006[28]	Low	Low	Low	Low	Low	Low	
Hogan, 2005[8]	High	Low	Low	Some concerns	Low	High	No information on randomization or outcome assessment.

Table D-2. Risk of bias for cohort studies

Study, Year	Bias Due to Confounding	Bias in Selection of Participants into the Study	Bias in Classification of Interventions	Bias Due to Deviations from Intended Interventions	Bias Due to Missing Data	Bias in Measurement of Outcomes	Bias in Selection of Reported Result	Overall Risk of Bias Judgment	Comments
Dixon, 2018[32] (Study 1 and Study 2)	Serious	Moderate	Low	Low	Serious	Moderate	Low	Serious	No controlling for confounding; sample only included participates with breastfeeding
Guinot, 2022[33]	Serious	Low	Low	Low	Moderate	Moderate	Low	Serious	Confounding not addressed; potential risk of bias from outcome reporting and attrition
Schlatter, 2019[34]	Serious	Low	Low	Low	Moderate	Moderate	Low	Serious	No controls for confounding; potential bias from attrition and measurement of outcomes
Sharma, 2015[35]	Serious	Low	Low	Low	Moderate	Moderate	Low	Serious	No controls for confounding; potential for bias from attrition and measurement of outcomes
Steehler, 2012[36]	Serious	Low	Low	Low	Low	Moderate	Low	Serious	No controls for confounding; potential for bias in measurement of outcomes

Table D-3. Quality of single-arm studies of safety

Study, Year	Does the patient(s) represent(s) the whole experience of the investigator (center) or is the selection method unclear to the extent that other patients with similar presentation may not have been reported?	Was the exposure adequately ascertained?	Was the outcome adequately ascertained?	Were other alternative causes that may explain the observation ruled out?	Was there a challenge/rechallenge phenomenon?	Was follow-up long enough for outcomes to occur?	Is the case(s) described with sufficient details to allow other investigators to replicate the research or to allow practitioners make inferences related to their own practice?	Overall Comments
Akbari, 2023[37]	Yes	Yes	Yes	Unclear	NA or not specified	Unclear	No	None.
Amir, 2005[38]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Only reported no complications.
Argiris, 2011[39]	Yes	Yes	Yes	Unclear	NA or not specified	Yes	Yes	None.
Ballard, 2002[40]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Only reported no complications.
Barberá-Pérez, 2021[41]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Very little information provided. Only stated that there were complications and reported irritability for one case.
Benoiton, 2016[42]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Only reported no complications.
Bhandarkar, 2022[43]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Very little information provided on harms.
Billington, 2018[44]	Yes	Yes	Yes	Unclear	NA or not specified	Unclear	No	Stated that postoperative complications were defined as ongoing feeding difficulties, bleeding or recurrence of tongue-tie; no information on how they were collected or other potential harms. Measurement methods unclear.
Blenkinsop, 2003[45]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Only reported no post-treatment complications.
Braccio, 2016[46]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	None.

Study, Year	Does the patient(s) represent(s) the whole experience of the investigator (center) or is the selection method unclear to the extent that other patients with similar presentation may not have been reported?	Was the exposure adequately ascertained?	Was the outcome adequately ascertained?	Were other alternative causes that may explain the observation ruled out?	Was there a challenge/rechallenge phenomenon?	Was follow-up long enough for outcomes to occur?	Is the case(s) described with sufficient details to allow other investigators to replicate the research or to allow practitioners make inferences related to their own practice?	Overall Comments
Dell'Olio, 2022[47]	Yes	Yes	Yes	Unclear	NA or not specified	Yes	Yes	Asked parent to report any complications, adverse events and/or complains during the 30 day follow-up period. Included intraoperative complications. Used C.R.I.E.S. scale for infant pain and healing index.
Dollberg, 2014[48]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Not enough details provided on how outcomes were measured; alternative explanations could not be ruled out. Unclear who provided information or how questions were asked.
Donati-Bourne, 2015[49]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	None.
Ferrés-Amat, 2017[50]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Not enough information provided.
Freeman, 2022[51]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	None.
Geddes , 2008[11]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Only reported no complications.
Ghaheri, 2017[52]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Very little information provided. Only reported no complications.
Griffiths, 2004[53]	Yes	Yes	Yes	Unclear	NA or not specified	Yes	Unclear	None.
Hale, 2020[54]	Yes	Yes	Yes	Unclear	NA or not specified	Yes	Yes	Study provides details on harms and could be replicated.

Study, Year	Does the patient(s) represent(s) the whole experience of the investigator (center) or is the selection method unclear to the extent that other patients with similar presentation may not have been reported?	Was the exposure adequately ascertained?	Was the outcome adequately ascertained?	Were other alternative causes that may explain the observation ruled out?	Was there a challenge/rechallenge phenomenon?	Was follow-up long enough for outcomes to occur?	Is the case(s) described with sufficient details to allow other investigators to replicate the research or to allow practitioners make inferences related to their own practice?	Overall Comments
Hand, 222[55]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Very little information provided.
Hill, 2022[56]	Yes	Yes	Yes	Unclear	NA or not specified	Yes	Yes	None.
Hill, 2022[57]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	Unclear	Asked a yes or no question about complications. No further details on the type of complication.
Hong, 2010[58]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Reported "all patients tolerated treatment well" and reported no complications.
Illing, 2019[59]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	None.
Lyudin, 2018[60]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	No information on how harms were obtained. Only reported number of unplanned visits after the procedure and reasons.
Masaitis, 1996[61]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Reported no complications nor any excessive bleeding.
McKenna, 2024[62]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Only said no complications were reported in either group. One sample from NICU had a mean gestational age of 34.4 weeks.
Mettias, 2013[63]	Yes	Yes	Yes	Unclear	NA or not specified	Unclear	Yes	unsure if follow-up was long enough but helpful authors provided questionnaire.
Miranda, 2010[64]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Only reported no complications.

Study, Year	Does the patient(s) represent(s) the whole experience of the investigator (center) or is the selection method unclear to the extent that other patients with similar presentation may not have been reported?	Was the exposure adequately ascertained?	Was the outcome adequately ascertained?	Were other alternative causes that may explain the observation ruled out?	Was there a challenge/rechallenge phenomenon?	Was follow-up long enough for outcomes to occur?	Is the case(s) described with sufficient details to allow other investigators to replicate the research or to allow practitioners make inferences related to their own practice?	Overall Comments
Muldoon, 2017[65]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	Yes	Recurrence rate was based on parent reporting why they stopped breastfeeding.
Narsat, 2022[66]	Yes	Yes	Yes	Unclear	NA or not specified	Yes	No	None.
O'Callahan, 2013[67]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Few details provided.
Patel, 2019[68]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Very little information on measurement of outcomes. Complication reports limited to 4 weeks after procedure.
Pransky, 2015[69]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Only reported no complications.
Ramoser, 2019[70]	Yes	Yes	Yes	Yes	NA or not specified	Yes	No	Considered short- and long-term outcomes. Very little information provided on how complications were measured and very little information reported in results.
Ridgers, 2009[71]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Only reported no complications.
Sethi, 2013[72]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Few details provided.
Siggaard, 2022[73]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Does not provide specifics on how exactly complications were defined.

Study, Year	Does the patient(s) represent(s) the whole experience of the investigator (center) or is the selection method unclear to the extent that other patients with similar presentation may not have been reported?	Was the exposure adequately ascertained?	Was the outcome adequately ascertained?	Were other alternative causes that may explain the observation ruled out?	Was there a challenge/rechallenge phenomenon?	Was follow-up long enough for outcomes to occur?	Is the case(s) described with sufficient details to allow other investigators to replicate the research or to allow practitioners make inferences related to their own practice?	Overall Comments
Slagter, 2021[74]	Yes	Yes	Yes	Unclear	N/A or not specified	Yes	Yes	The study cited that they used the complication outcomes that were considered in the 2017 Cochrane review.
Srinivasan, 2006[76]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	None.
Srinivasan, 2019[75]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Reports no complications were reported and a small amount of data on bleeding.
Todd, 2015[77]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Study planned to collect information on complications immediately post-procedure, but did not provide information on what complications they were looking for.
Toner, 2014[78]	Yes	Yes	Unclear	Unclear	NA or not specified	Yes	No	Based on parent report, from 6 years to a few months after procedure.
Towfighi, 2022[79]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	None.
Wakhanrittee, 2016[80]	Yes	Yes	Unclear	Unclear	NA or not specified	Unclear	No	Few details provided. Only reported complications within 24 hours.
Wallace, 2006[81]	Yes	Yes	Unclear	Unclear	NA or not specified	Yes	No	No reported complications of the procedure based on the parents' report 4 months later.

Study, Year	Does the patient(s) represent(s) the whole experience of the investigator (center) or is the selection method unclear to the extent that other patients with similar presentation may not have been reported?	Was the exposure adequately ascertained?	Was the outcome adequately ascertained?	Were other alternative causes that may explain the observation ruled out?	Was there a challenge/rechallenge phenomenon?	Was follow-up long enough for outcomes to occur?	Is the case(s) described with sufficient details to allow other investigators to replicate the research or to allow practitioners make inferences related to their own practice?	Overall Comments
Wen, 2022[82]	Yes	Yes	Unclear	Unclear	N/A or not specified	Unclear	No	Reported no post-surgical complications and postoperative pain responses. No long-term harms outcomes.

Notes: Question 6 "Was there a dose–response effect?" was irrelevant for all studies and has been excluded from this table.

Abbreviations: C.R.I.E.S. = Crying, Requires increased oxygen administration, Increased vital signs, Expression, Sleeplessness. NA = not applicable; NICU = neonatal intensive care unit.