INTRODUCTION

Ankyloglossia, or tongue-tie, is a congenital anomaly that is characterized by a short or thickened lingual frenulum; this can be associated with impaired breastfeeding, speech, and dentofacial growth. The indications for performing frenotomy, frenuloplasty, or other operative interventions are unclear.

Background: Ankyloglossia, or tongue-tie, is characterized by a short or thickened lingual frenulum; this can be associated with impaired breastfeeding, speech, and dentofacial growth. The indications for performing frenotomy, frenuloplasty, or other operative interventions are unclear.

Methods: A meta-analysis was performed to identify the extent of the benefit from frenotomy in breastfeeding measures, degree of tongue-tie, and maternal pain during feeding in randomized controlled trials. A structured literature review analyzed the optimal type and timing of repair. An algorithm was developed to incorporate this evidence into a management pathway.

Results: Among 424 studies reviewed, 5 randomized controlled trials met inclusion criteria for meta-analysis. Frenotomy significantly improved the degree of tongue-tie, with a 4.5-point decrease in Hazelbaker Assessment Tool for Lingual Frenulum Function score compared with a decrease of 0 in those who did not undergo frenotomy ($P < 0.00001$). This was associated with improved self-reported breastfeeding (relative risk [RR] = 3.48, $P < 0.00001$) and decreased pain (Short-Form McGill Pain Questionnaire, $P < 0.00001$); however, Breastfeeding Self-Efficacy–Short Form and Latch, Audible Swallowing, Type of Nipple, Comfort, Hold scores did not significantly improve. Multiple studies demonstrated significant improvements following frenuloplasty when compared with frenotomy but demonstrated mixed results as to the effect of timing of tongue-tie division.

Conclusions: Frenotomy is associated with breastfeeding improvements that vary individually but trend toward significance collectively during a critical time in infant development. Among patients with a severe Hazelbaker Assessment Tool for Lingual Frenulum Function score or difficulty breastfeeding, we conclude that simple frenotomy without anesthetic is generally indicated in infancy and frenuloplasty under general anesthesia for older children. (Plast Reconstr Surg Glob Open 2021;9:e3336; doi: 10.1097/GOX.0000000000003336; Published online 25 January 2021.)
and dentofacial development. But the benefits of these procedures need to be weighed with the costs and risks associated with any surgery and anesthetic use in infants.10

The treatment that children currently receive is usually based on the personal experience of the provider, and there is a lack of evidence to guide management. A recent survey of providers from nursing, medical, dental, and allied health backgrounds showed varying sentiments about the association of ankyloglossia and breastfeeding difficulty, the diagnosis of ankyloglossia, the use of existing clinical practice guidelines, and the use of frenotomy for treatment across healthcare disciplines.10 There are a number of surgical techniques that can be employed to treat symptomatic ankyloglossia. Frenotomy can usually be performed in the office setting with the use of a grooved retractor and scissors, laser (neodymium-doped yttrium aluminum garnet [Nd:YAG], carbon dioxide, diode, erbium-YAG), or electrocautery, with or without topical anesthetic. Operative techniques include frenulectomy, horizontal-to-vertical frenuloplasty 2-flap z-frenuloplasty, and 4-flap z-frenuloplasty under general anesthesia.21

The lack of robust outcome data following these procedures creates an opportunity to develop standardized diagnostic and treatment protocols. The purpose of this study is to review the evidence for functional improvement derived from frenotomy, indications for repair, and optimal type and timing of repair.

METHODS

Outcome Meta-analysis

The population of interest included pediatric patients with symptomatic ankyloglossia (tongue-tie), with outcomes related to child or mother disease severity (breast or bottle-feeding effectiveness, maternal pain, degree of tongue-tie, dental, speech, or any combination therein) studied following surgical intervention. A comprehensive search of Ovid MEDLINE, EMBASE, and Cochrane Database of Systematic Reviews was performed in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines, using various combinations of the search terms “ankyloglossia” and “tongue-tie” and a wide range of potential measures of disease severity. Randomized controlled human trials were included, and studies including syndromic and/or other craniofacial conditions that could be potential causes of symptomatology were excluded (Table 1). Data from included studies were abstracted, and a level of evidence was determined for each article in accordance with the American Society of Plastic Surgeons Evidence Rating Scale for Therapeutic Studies.12

Structured Analysis of Repair Timing and Type

The population of interest included pediatric patients with symptomatic ankyloglossia (tongue-tie). Included studies directly studied the type of operative repair performed, timing of repair, or the use of anesthetic during repair. The same comprehensive search was done as for the meta-analysis of outcomes. Following the initial search, all of the studies were reviewed and analyzed. Data from the included studies were collected, and a level of evidence was determined for each article in accordance with the American Society of Plastic Surgeons Evidence Rating Scale for Therapeutic Studies.

Statistical Analysis

Statistical analysis was performed using Cochrane’s Review Manager 513 and Microsoft Excel (Redmond, Wash.).

RESULTS

Study Selection

The search was performed in January 2018, and yielded 567 articles, of which 424 remained following removal of duplicate results (Fig. 1). The abstracts of the resulting articles were reviewed and analyzed for inclusion and exclusion criteria. Six14-19 articles of interest resulted from the initial search for the meta-analysis of outcomes. One study was excluded following full-text review because the study compared methods of tongue-tie division and did not contain a control group with no intervention, leaving 5 studies for use in the meta-analysis. For the structured review of the optimal surgical repair, 10 articles of interest resulted from the initial search. Four studies addressed the timing of repair, 4 addressed the type of surgical repair, and 2 studies investigated the use of anesthetic in repair of ankyloglossia.

Outcome Meta-analysis

Study Characteristics

All of the included studies measured outcomes following division by frenotomy or nondivision of ankyloglossia in at least 1 of the 3 following domains: breastfeeding, degree of tongue-tie, and maternal pain. Validated measures of breastfeeding efficacy utilized by the authors of included studies were the Latch, Audible Swallowing, Type of Nipple, Comfort, Hold (LATCH) score,20,21 the Infant Breastfeeding Assessment Tool (IBFAT),23 and the Breastfeeding Self-Efficacy Score–Short Form (BSES-SF)23 (Table 2). The validated measure of degree of tongue-tie, utilized by Buryk et al,17 was the Hazelbaker Assessment Scale for Therapeutic Studies.12

Table 1. Criteria for Considering Studies for This Systematic Review and Meta-Analysis

| Study Criteria                                                                 | Description                                                                 |
|-------------------------------------------------------------------------------|-----------------------------------------------------------------------------|
| Population                                                                     | Symptomatic patients age 0–18 y with ankyloglossia (tongue-tie)             |
| Interventions                                                                 | Surgical intervention (frenotomy, frenectomy, frenuloplasty, frenulectomy) versus no intervention |
| Outcomes                                                                      | Feeding (breast or bottle), maternal pain, degree of tongue-tie, dental, speech |
| Inclusion criteria                                                             | Randomized control trials of human subjects with at least one of the above outcomes measured |
| Exclusion criteria                                                             | 1. Infant or maternal symptoms related to a labial frenulum                  |
|                                                                              | 2. Syndromic or other craniofacial conditions that are possible causes of the symptomatology |
a 1–10 scale, the Visual Analog Scale, and the validated Short-Form McGill Pain Questionnaire. An overview of the findings of each included study is outlined in Figure 2. The studies included for meta-analysis involved a consistent surgical procedure. No anesthetic is used, and the baby is wrapped securely in a towel. An assistant holds the infant’s shoulders back with their hands, and uses their wrist to fix the infants head in place. The tongue is then held out of the way by the provider performing the division by direct pressure on the tongue with the aid of a grooved director, and the lingual frenulum is divided using sharp, blunt-ended scissors.

Breastfeeding Outcomes
Dollberg et al and Emond et al found no difference in LATCH scores following frenotomy or nondivision. Pooling of the data also showed no statistically significant difference between groups (P = 1.0) (Table 3). Furthermore, there was no difference in the change from baseline of LATCH scores between study groups when study data were combined (P = 1.0).

Following frenotomy or nondivision, IBFAT scores following frenotomy or nondivision from Buryk et al and Emond et al favor frenotomy (P < 0.00001; 95% confidence interval [CI], 0.83–1.30) (Fig. 3A). In a similar fashion, there was a statistically significant benefit to frenotomy (P < 0.00001; 95% CI, 2.62–2.74) when examining the change from baseline of IBFAT scores following frenotomy or nondivision. Emond et al found no difference in BSES-SF scores following frenotomy or nondivision (P = 0.53), but the frenotomy group experienced a greater increase in BSES-SF scores when compared with the nondivision group (P = 0.002).

Meta-analysis showed that patients who underwent frenotomy had better absolute scores across validated breastfeeding efficacy scoring systems following the intervention than patients who did not undergo division (P = 0.04; 95% CI, 0.01–0.41), and patients who underwent frenotomy had a greater improvement across validated breastfeeding

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**Fig. 1.** Flow chart of systematic literature search.
efficacy scores when compared with patients who did not undergo division ($P = 0.003; 95\% \text{ CI}, 0.10–0.52$) (Fig. 4).

There was a subjective improvement in breastfeeding, as reported by the mother, in studies by Berry et al\textsuperscript{15} and Hogan et al\textsuperscript{18} following frenotomy when compared with nondivision ($P < 0.00001$; Relative Probability of Feeding Improvement 95\% CI, 2.18–5.56) (Table 4). Berry et al\textsuperscript{15} found no dichotomous change in breastfeeding efficacy as documented by an objective observer between patients who underwent frenotomy or nondivision. However, meta-analysis shows that frenotomy is favored when pooling dichotomous qualitative assessment of breastfeeding when measured by the mother and an objective observer ($P < 0.00001$, Relative Probability of Feeding Improvement 95\% CI, 1.75–3.58) (Fig. 3B).

**Degree of Tongue-tie**

Emond et al\textsuperscript{16} found that patients who underwent frenotomy had significant improvement in HATLFF score...
(mean = 4.5) compared with those who did not (mean = 0; \( P < 0.0001 \)) and higher HATLFF scores following frenotomy (mean = 13.5) compared with nondivision (mean = 8; \( P < 0.0001 \)).

**Maternal Pain during Breastfeeding**

The pooling of pain scores during breastfeeding by Berry et al,15 Dollberg et al,14 and Emond et al16 showed no significant difference following frenotomy or nondivision (\( P = 0.46; 95\% \text{ CI}, −0.98 \) to 0.44) (Table 5; Fig. 5). Buryk et al17 demonstrated lower mean Short-Form McGill Pain Questionnaire scores in mothers of infants who underwent frenotomy (mean = 4.9) when compared with those who underwent nondivision (mean = 13.5; \( P < 0.00001 \)) when imputed SDs were used for statistical analysis. In overall meta-analysis, frenotomy resulted in a significant decrease in maternal pain during breastfeeding across multiple validated scales (\( P = 0.004; 95\% \text{ CI}, −0.70 \) to −0.13) (Fig. 6).

**Structured Literature Review of Optimal Surgical Treatment**

All of the included studies measured various outcomes following tongue-tie division related to the timing of division, the technique used for division, and the use of anesthetic. Although there was heterogeneity in the specific outcomes measured, the optimal timing for division of tongue-tie was studied in relation to infant breastfeeding measures and weight gain. Tongue movement and speech measures were used to compare operative techniques in the included studies, and pain measures were used to evaluate the use of anesthetics in tongue-tie division.

**Timing**

Wakhanrittee et al28 found no difference in success of exclusive breastfeeding at 3 months between frenotomy done before and after 24 hours of life nor a difference in the median age of infants at the time of frenulotomy who successfully exclusively breastfed at 3 months of age compared with those who did not exclusively breastfeed at 3 months. Steehler et al29 concluded similarly in a retrospective study which utilized a prospective telephone survey. They found that there was no difference in the total length of breastfeeding between groups that underwent frenotomy within the first week of life or after the first week of life, but there was a subjective benefit, as reported by the mother, in the infant’s ability to feed if frenotomy was performed within the first week of life. In a small study by Praborini et al,30 it was found that infants who underwent frenotomy before 8 days of age gained significantly more weight than those infants who underwent frenotomy after 8 days. A retrospective study by Sharma and Jayaraj31 found improved breastfeeding unrelated to whether division was before 30 days of age or after.31

**Technique**

Tongue-tie, when identified in neonates, is typically divided as an in-office procedure, with the use of topical or no anesthetics. Most studies outline a methodology that includes holding the child’s shoulders back and using blunt-ended scissors to divide the lingual frenulum.

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### Table 3. Meta-Analysis of Quantitative Breastfeeding Outcomes by Validated Measures

| Outcome Measure | Control Group Mean (SD) | Frenotomy Group Mean (SD) | Mean Difference (95% CI) | Standardized Mean Difference (95% CI) | \( P \) |
|-----------------|-------------------------|---------------------------|--------------------------|----------------------------------------|------|
| LATCH following frenotomy or sham | 6.8 (1.48) | 9.0 (1.48) | 2.15 (1.82 to 2.58) | 1.78 (1.27 to 2.28) | <0.00001 |
| IBFAT following frenotomy or sham | 8.47 (0.89) | 12.0 (0.74) | 3.53 (3.09 to 3.96) | 4.17 (3.28 to 5.06) | <0.00001 |
| BSES-SF following frenotomy or sham | 53.0 (14.56) | 51.4 (14.56) | 1.60 (−0.56 to 3.76) | 0.12 (−0.44 to 0.68) | 1.00 |
| LATCH change from baseline following frenotomy or sham | 0.00 (1.00) | 0.00 (1.00) | 0.00 (−0.56 to 0.56) | 0.00 (−0.38 to 0.38) | 1.00 |
| IBFAT change from baseline following frenotomy or sham | 2.3 (0.57) | 2.7 (0.57) | 0.40 (−0.06 to 0.86) | 0.07 (−0.28 to 0.42) | 1.00 |
| BSES-SF change from baseline following frenotomy or sham | 9.60 (5.45) | 2.80 (5.45) | −6.80 (−12.24 to 0.44) | −0.80 (−1.94 to 0.34) | <0.00001 |

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*O'Shea et al obtained further data from Dollberg et al that is used in this meta-analysis.*
Fig. 3. Validated breastfeeding outcome measure score difference following frenotomy compared to sham (A, left side) and score difference compared to baseline (A, right side.) Validated feeding outcome measure change following frenotomy compared to sham (B).

Table 4. Meta-Analysis of Dichotomous Breastfeeding Outcomes by Validated Measures

| Outcome Measure                                      | Study                        | Frenotomy (No. Events/Total Subjects) | Control Group (No. Events/Total Subjects) | Relative Probability of Feeding Improvement (95% CI) | P      |
|------------------------------------------------------|------------------------------|---------------------------------------|-------------------------------------------|----------------------------------------------------|--------|
| Maternal qualitative improvement in feeding          | Hogan et al.                 | 27/28                                 | 1/29                                      | 27.96 (4.07 to 192.12)                              |        |
|                                                      | Berry et al.                 | 21/27                                 | 14/30                                     | 1.67 (1.08 to 2.57)                                |        |
|                                                      | Subtotal                     |                                       |                                           | 3.48 (2.18 to 5.56)                                | <0.0001|
| Objective observer improvement in feeding            | Berry et al.                 | 13/26                                 | 12/30                                     | 1.25 (0.70 to 2.24)                                | 0.45   |
|                                                      | Subtotal                     |                                       |                                           | 1.25 (0.70 to 2.24)                                |        |
| Combined dichotomous measures of feeding improvement | Total                        |                                       |                                           | 2.50 (1.75 to 3.58)                                | <0.0001|

Table 5. Meta-Analysis of Quantitative Maternal Breastfeeding Pain by Validated Measures

| Outcome Measure                                      | Study                        | Frenotomy (Mean (SD)) | Control Group (Mean (SD)) | Mean Difference (95% CI) | Standardized Mean Difference (95% CI) | P      |
|------------------------------------------------------|------------------------------|------------------------|---------------------------|--------------------------|--------------------------------------|--------|
| Visual Analog or 10-point Scale following frenotomy or sham | Berry et al.                 | 1.6 (1.22)             | 2.9 (3.35)                | -1.30 (-3.17 to 0.57)    | -0.41 (-0.70 to -0.13)               | 0.46   |
|                                                      | Dollberg et al.              | 5.3 (2.2)              | 5.5 (1.9)                 | -0.20 (-1.34 to 0.94)    | -0.41 (-0.70 to -0.13)               |        |
|                                                      | Emond et al.                 | 3 (2.44)               | 3 (2.96)                  | 0.00 (-1.04 to 1.04)     | -0.27 (-0.98 to 0.44)               |        |
|                                                      | Subtotal                     | 13.5 (1.5)             | 13.5 (1.5)                | -8.60 (-9.36 to -7.84)   | -0.41 (-0.70 to -0.13)               | <0.0001|
| SF-MPQ following frenotomy or sham                   | Buryk et al.                 | 4.9 (1.46)             | 13.5 (1.5)                | -8.60 (-9.36 to -7.84)   | -0.41 (-0.70 to -0.13)               | 0.004  |
|                                                      | Subtotal                     | 13.5 (1.5)             | 13.5 (1.5)                | -8.60 (-9.36 to -7.84)   | -0.41 (-0.70 to -0.13)               | <0.0001|
| Combined quantitative breastfeeding pain measures    | Total                        | 13.5 (1.5)             | 13.5 (1.5)                | -8.60 (-9.36 to -7.84)   | -0.41 (-0.70 to -0.13)               | <0.0001|
In a randomized trial of patients older than 3 years of age with constrained articulation, 4-flap z-frenuloplasty was compared with the traditional horizontal-to-vertical frenuloplasty and demonstrated greater absolute increases in frenulum length, tongue protrusion, and speech articulation measured by 2 speech pathologists when >10 months from the operative tongue-tie release. Z-frenuloplasty demonstrated better outcomes at 3 months following tongue-tie release than simple frenotomy in areas of parental satisfaction, tongue mobility and elongation, and speech articulation in a randomized trial. Choi et al found that z-frenuloplasty with partial myotomy of the genioglossus resulted in subjective speech improvement, and Klockars and Pitkäranta found that frenuloplasty required no reoperations, compared with the frequency of reoperation in patients following simple frenotomy. Frenulectomy was rarely described, and no data suggested its superiority over frenotomy or frenuloplasty.

Use of Anesthetic
A double-blinded randomized study by Shavit et al compared the use of topical 2% tetracaine or 20% benzocaine before frenotomy and found no difference between the 2 anesthetics in the first 30 days of life; furthermore, the study concluded that both anesthetics incompletely controlled infant pain. Another double-blinded study showed no difference between the use of topical benzocaine and no anesthesia during the procedure in infants 0–3 days of age. When the mean age of patients in all of the studies discussed in this review are plotted, the use of topical or no anesthetic is generally utilized in approximately the first month of life (Fig. 7), with tongue-tie division under no anesthetic being performed up to the 115th day of life (Table 6). Operative general anesthesia was utilized in patients from the 5th to the 3614th day of life, with the mean ages ranging from 960 to 2065 days (or 32 months to 5.65 years).

DISCUSSION
Several key findings emerged from this study. First, frenotomy is associated with breastfeeding improvements that vary individually but trend toward significance collectively during a critical time in infant development. Second, simple frenotomy with or without local anesthesia appears well tolerated until approximately 3 months of life, after which correction under general anesthesia better controls pain. Third, after 3 months of age, frenuloplasty with 2-flap z-plasty or 4-flap technique yields greater absolute improvements in tongue mobility and speech metrics when compared with the traditional horizontal-to-vertical frenuloplasty, and frenuloplasty is associated with decreased reoperation when compared with the traditional frenotomy. Finally, evidence supporting the superiority of earlier timing of frenotomy on infant breastfeeding duration and efficacy was not conclusive.

Treatment Algorithm

Intervention versus Nonintervention (Grade: B)
It is clear that the use of a scoring system in which functional considerations are taken into account is beneficial, as anatomic considerations alone do not correlate as strongly with breastfeeding difficulty. In the current literature, there is a lack of uniformity in the grading of ankyloglossia. For this reason, we propose the use of the HATLFF scoring system in assessment of ankyloglossia, which has been shown to correlate with breastfeeding difficulty and success of intervention, and more specifically,
the use of the “function impaired” HATLFF score as an indication for division.39–41 Some infants cannot be classified by the HATLFF scoring system,40,42 so infants who are experiencing breastfeeding difficulty should also undergo division, as our meta-analysis has shown that division is associated with breastfeeding improvement.

Age at Time of Intervention (Grade: D)

There is a lack of consistent empirical evidence to guide the timing of intervention in infants with tongue-tie, with available studies evaluating different metrics at varied time points. To facilitate breastfeeding, frenum release must necessarily be performed as early as possible in infancy. One study suggested infants over 4 months of age become more strong and aware, and division should be performed under general anesthesia.39 This is an area requiring further study, but it is clear that there is an improvement in breastfeeding following division; therefore, ankyloglossia should be addressed as early as possible in affected infants.

| Study                          | No. Patients | Average Age at Time of Tongue-tie Division (d) |
|-------------------------------|--------------|-----------------------------------------------|
| No anesthetic                  |              |                                               |
| Berry et al15                 | 27           | 33                                            |
| Hogan et al16                 | 28           | 20                                            |
| Topical anesthetic            |              |                                               |
| Buryk et al17                 | 30           | 6.2                                           |
| Wakhanrittee et al28          | 328          | 2.08*                                         |
| Stechler et al29              | 302          | 18                                            |
| General OR anesthesia         |              |                                               |
| Yousefi et al20               | 50           | 960                                           |
| Heller et al21                | 16           | 2064.5                                         |

*Median age.

Procedure for Division (Grade: B or C)

Randomized controlled trials have shown that z-frenuloplasty and 4-flap frenuloplasty are superior to simple frenotomy across multiple metrics but must be performed under general anesthesia. The benefit of this procedure must be weighed against the potential dangers that general anesthesia can pose to infants. No identified evidence supports the superiority of frenulectomy over either frenotomy or frenuloplasty. Because of these considerations, we only recommend performing z-frenuloplasty or 4-flap frenuloplasty under general anesthesia, which is more safely performed in childhood rather than in infancy.

Synthesizing this evidence about type and timing of procedure, we propose a treatment algorithm that identifies 2 ideal opportunities for addressing symptomatic ankyloglossia (Fig. 8). The first is in infancy, when frenotomy of a thin band of tissue can be performed with local anesthesia in the clinic setting and, in many cases, facilitate improved breastfeeding. The second is in childhood, when more extensive frenuloplasty can be performed more safely under general anesthesia. The evidence suggests either of these options is likely superior to a procedure under general anesthesia in infancy or a simple frenotomy without general anesthesia in childhood. However, we advocate for this algorithm being but one factor in a conversation between a physician and a patient’s family, with treatment tailored to the clinical situation.

This study has several notable limitations. There were few articles that met inclusion criteria, mainly due to the lack of randomized control trials on this topic. Further, there was significant variability among outcome measures used across studies, leading to incomparability. Future studies may benefit from using standardized outcome measures in measuring the degree of tongue-tie and outcome measures, so that strong conclusions can be drawn.
as to the indications and effectiveness of tongue-tie division on pediatric patients.

CONCLUSIONS

The first months of a child’s life can be incredibly stressful for parents/caregivers, and this can be compounded by the uncertainty of the need or potential benefit of tongue-tie release. This article seeks to bring clarity to these issues by assembling evidence regarding the indications for and potential benefit of this procedure. Key findings include that frenotomy is associated with breastfeeding improvements that trend toward significance collectively, the HATLFF scoring system has been shown to correlate with breastfeeding difficulty and success of intervention, ankyloglossia should be addressed as early as possible, and those with severe HATLFF score or difficulty breastfeeding should undergo simple frenotomy without anesthetic if under 4 months of age and z-frenuloplasty or 4-flap frenuloplasty under general anesthesia if over 4 months of age. We advocate for standardization to the existing body of literature for continued exploration of this topic.

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