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Revision Lingual Frenotomy Improves Patient-Reported Breastfeeding Outcomes: A Prospective Cohort Study

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Abstract

Background: Lingual frenotomy improves patient-reported outcome measures, including infant reflux and maternal nipple pain, and prolongs the nursing relationship; however, many mother–infant dyads continue to experience breastfeeding difficulty despite having had a frenotomy.

Research aim: The aim of this study was to determine how incomplete release of the tethered lingual frenulum may result in persistent breastfeeding difficulties.

Methods: A one-group, observational, prospective cohort study was conducted. The sample consisted of breastfeeding mother–infant (0-9 months of age) dyads (N = 54) after the mothers self-elected completion lingual frenotomy and/or maxillary labial frenectomy following prior lingual frenotomy performed elsewhere. Participants completed surveys preoperatively, 1-week postoperatively, and 1-month postoperatively consisting of the Breastfeeding Self-Efficacy Scale–Short-Form (BSES-SF), Visual Analog Scale (VAS) for nipple pain severity, and the Revised Infant Gastroesophageal Reflux Questionnaire (I-GERQ-R).

Results: Significant postoperative improvements were reported between mean preoperative scores compared with 1-week and 1-month scores of the BSES-SF, $F_{(2)} = 41.2$, $p < .001$; the I-GERQ-R, $F_{(2)} = 22.7$, $p < .001$; and VAS pain scale, $F_{(2)} = 46.1$, $p < .001$.

Conclusion: We demonstrated that besides nipple pain, measures of infant reflux symptoms and maternal breastfeeding self-confidence can improve following full release of the lingual frenulum. Additionally, a patient population was identified that could benefit from increased scrutiny of infant tongue function when initial frenotomy fails to improve breastfeeding symptoms.

Keywords

ankyloglossia, breastfeeding, breastfeeding assessment, health services research, tongue-tie

Background

Breastfeeding initiation rates in the United States have been increasing, with renewed emphasis on the importance of breastfeeding exclusivity and duration (Office of Disease Prevention and Healthy Promotion, 2017). Previous authors have demonstrated the health risks and financial detriment resulting from premature breastfeeding cessation (Bartick et al., 2017), in addition to how breastfeeding provides a protective effect against dental malocclusion (Peres, Cascaes, Nascimento, & Victora, 2015). It is of vital importance to identify and treat the causes of dysfunctional breastfeeding and associated symptoms that may result in premature cessation (Newby & Davies, 2016). Neonatal ankyloglossia has been identified as an established cause of breastfeeding problems and a risk factor for premature cessation (Ricke, Baker, Madlon-Kay, & DeFor, 2005; Todd & Hogan, 2015). Prior to ultrasound studies of the infant mouth during breastfeeding, it was thought that the tongue was responsible for milk withdrawal via a peristaltic wave that compresses lactiferous sinuses in the breast (Woolridge,

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1986). Two additional studies have reported mid-tongue mobility toward the palate as the critical motion in achieving the seal and suction necessary for successful breastfeeding (Elad et al., 2014; Geddes et al., 2008).

A lingual frenotomy may alleviate ankyloglossia and improve lingual mobility, subsequently improving infant latch and decreasing maternal nipple pain (Dollberg, Botzer, Grunis, & Mimouni, 2006; O'Callahan, Macary, & Clemente, 2013). Unfortunately, previous studies examining benefits of lingual frenotomy have been limited to either children with obvious anterior ("classic") tongue-tie or mothers who report only limited breastfeeding treatment outcomes like nipple pain (Dollberg et al., 2006; O'Callahan et al., 2013). A recent, comprehensive outcomes study included infants with both anterior and posterior tongue-tie (Ghaheri, Cole, Fausel, Chuop, & Mace, 2017). These researchers demonstrated significant average improvements in patient-reported outcome measures (PROMs), including infant gastroesophageal reflux scores, breastfeeding efficiency, nipple pain, and poor maternal breastfeeding self-confidence, following surgical intervention for ankyloglossia. In that study, infant–mother dyads were excluded if previous frenotomy had been performed by an alternate provider.

Classic lingual frenotomy typically involves division of an anterior lingual band but not the release of tissue directly overlying and lateral to the genioglossus muscle. To date, no investigation has focused on infant–mother dyads who fail to experience symptom improvement following lingual frenotomy. Because previous researchers demonstrated the importance of releasing the posterior/submucosal fibers of ankyloglossia, we hypothesized that infant–mother dyads who do not report symptom improvement from previous frenotomy have persistent ankyloglossia that needs further intervention. The study aim was to evaluate improvement of patient-reported infant reflux, maternal nipple pain, and breastfeeding self-efficacy following completion lingual frenotomy.

Methods

Design

Mother–infant dyads were enrolled in an observational, prospective, consecutively enrolled single-cohort study to evaluate the efficacy of revision frenotomy with or without maxillary labial frenectomy for improving breastfeeding symptoms. Because identification of a comparable control group in such specific circumstances was difficult to achieve, the use of a cohort study provides some internal consistency and reasonably allows for analyzing treatment effect. The study was authorized by the institutional review board at Providence Health & Services (IRB No. 16-218B; Portland, OR, USA).

Key Messages

- This study was designed to determine how complete release of the tethered lingual frenulum can result in a durable reduction in persistent breastfeeding difficulties, as most practitioners' conception of lingual frenotomy is to release the thin anterior band under the tongue.
- Significant and discernible postoperative improvements in breastfeeding self-efficacy, maternal nipple pain, and symptoms associated with infant gastroesophageal reflux in children who had already undergone lingual frenotomy were realized. Our findings introduce the notion that children who have not improved following a previous frenotomy may have further restriction under the tongue that still needs attention.
- We have provided preliminary evidence to support the use of revision frenotomy to improve a range of breastfeeding outcomes following incomplete initial frenotomy.

Setting

The study was undertaken in an urban setting in two clinics in the Portland, Oregon, area; Portland is the largest city in Oregon, and its metropolitan area includes approximately two million people. The clinics serve a population that is mostly White (71%), followed by Hispanic (10%), Asian (8%), and African American (6%). Data were collected between June 2014 and April 2015.

Sample

The target population was mothers and infants who continue to exhibit abnormal breastfeeding symptoms despite previous lingual frenotomy. The sample inclusion criteria were infants ≤ 9 months of age, ≥ 37 weeks of gestational age at birth, who had previously undergone anterior lingual frenotomy. Potential participants were excluded from the study if (a) they were unable to complete outcome measure evaluations during all study time points. Additional exclusions were (b) infants with preoperative life-threatening comorbidity, (c) infants born from mothers of multiple births, and (d) mothers with previous breast surgery or insufficient glandular tissue, which affected breastfeeding efficacy. Study participants consisted of breastfeeding dyads referred for evaluation and consult surrounding revision lingual frenectomy because of persistent symptoms associated with impaired breastfeeding efficacy. A total of 54 mother–infant dyads were prospectively enrolled between June 2014 and April 2015.

Study sample size was calculated using previously reported within-subjects differences in average total scores on the Breastfeeding Self-Efficacy Scale–Short Form (BSES-SF) between preoperative and 1-month postoperative evaluations for a comparable cohort (Ghaheri et al., 2017). Assuming unequal standard deviations, a 5% alpha level, 80% power (1 – β), and a within-subjects Pearson correlation of .50, two-tailed testing estimated that a total of 46 participants was required to detect an average postoperative difference of at least 5.0 points (~7%) on BSES-SF total scores.

Measurement

Participants were asked to provide demographic and medical history before completing preoperative study-related outcome measures. Outcome measures were provided again at 1 week and 1 month postoperatively during either physician-directed clinical appointments or electronic correspondence using an Internet-based survey compliant with the Health Insurance Portability and Accountability Act of 1996 (SurveyMonkey, Palo Alto, CA, USA). Infants were followed through the standard of care to confirm adequate recovery and symptom improvement. Participants were asked to return to the clinic 1 week postoperatively or earlier if symptoms persisted or worsened, and they were followed for 6 months to determine reattachment rates.

Study participants self-administered complementary survey instruments designed to measure variables of interest (PROM) that may affect breastfeeding outcomes (e.g., self-efficacy, infant reflux, and maternal pain during feedings). Research staff were available to answer any questions about the instruments. The variable self-efficacy was measured using the BSES-SF (Dennis, 2003), a validated, 14-item survey designed to assess breastfeeding efficacy and maternal confidence. Total scores (range = 0-70) were summarized from response items (range = 0-5), and higher scores reflect lower breastfeeding impairment and higher maternal confidence.

Infant complications related to gastroesophageal reflux were measured by the Revised Infant Gastroesophageal Reflux Questionnaire (I-GERQ-R), a validated, 13-item survey designed for caregivers and medical practitioners to quantify gastroesophageal reflux severity in infants (Kleinman et al., 2006). Total I-GERQ-R scores (range = 0-42) were the summation of items that evaluate symptoms of reflux disease. Higher scores reflect more severe symptoms. Last, participants completed a single 10.0-cm Visual Analog Scale (VAS) to determine breastfeeding pain severity; higher scores reflect greater pain.

Data Collection

Voluntary written consent was obtained by trained research staff. Participants were assured that continued study involvement was voluntary and did not change the standard of care surrounding lingual frenotomy. Data were safeguarded using unique study identification numbers and elimination of protected health information from report forms. A closed, relational database (Access; Microsoft Corporation, Redmond, WA, USA) was used to secure data entry.

Data collection procedures were completed within a community-based otolaryngology clinic (The Oregon Clinic, Portland, OR, USA) by the principal investigator (B.A.G.) and trained research staff. Mother–infant dyads were prospectively enrolled and observed through the standard of care surrounding surgical intervention for symptoms of ankyloglossia. Surgical consultation prerequisites included oral and feeding evaluations conducted by a community International Board Certified Lactation Consultant (IBCLC). At the discretion of the IBCLC, objective measures of latch quality were utilized and incorporated into the reports that were reviewed prior to the procedure. Latch assessments were a major factor in determining surgical candidacy.

Targeted head and neck evaluations were completed by the principal investigator to identify anatomic restrictions, including lingual movement (transverse tongue reflex impairment), maxillary bony alveolar notching, blanched and restrictive frenula, abnormal mouth floor elevation with tongue elevation, and abnormal attachment of the frenula. In children who still reflexively suck, sucking evaluations were completed to identify abnormal gum/lip grip pressure, tongue cupping, seal quality, and manner of tongue movements. Infant anatomy was categorized using both the Kotlow (1999) upper lip-tie and Coryllos et al. (2004) tongue-tie classification systems to describe preoperative frenula anatomy (see Table 1). Baseline maternal symptoms were evaluated simultaneously using a symptom/complaint instrument during infant evaluations (see Table 2).

Surgical candidates were offered and voluntarily selected revision lingual frenotomy. Surgical consent was obtained from parents and the patient was relocated to a laser-safe operatory suite. Topical anesthetic (EMLA; Actavis Pharma, Parsippany, NJ, USA) was applied to the surgical site(s) with a cotton swab. Surgical procedures were conducted using a 1,064-nm InGaAsP semiconductor diode laser (Xlase; Technology4Medicine, San Clemente, CA, USA) with variable pulsed wave and wattage settings (0.7-0.8 W pulsed for 200 μs on and 100 μs off; actual wattage = 0.47-0.53 W), incorporating a 300-μm-diameter laser fiber.

The tongue was elevated using a grooved director while the laser tip was applied to the residual frenulum. Beginning with the remaining anterior edge of the frenulum, any remaining midline tissue was incised. Further incision of a central window, approximately 1.0-mm deep in the mucosa overlying the genioglossus muscle, was then performed. That central window was then extended bilaterally to release the mucosa, with particular attention toward the preservation of the fascia of the underlying genioglossus muscle. A full release was determined only after completion of the diamond-shape incision that is flush with the adjacent floor of mouth tissue. If needed, maxillary labial release involved elevation

Table 1. Participants' Anatomic Classification Types and Preoperative Frequency (N = 54).

Classification	Description	*n* (%)
Kotlow upper lip-tie classification[a]		
Class II	Attachment mostly into the gingival tissue	7 (13.0)
Class III	Attachment in front of the anterior papilla	25 (46.3)
Class IV	Attachment into the papilla or extending into hard palate	22 (40.7)
Coryllos tongue-tie classification[b]		
Type I	Attachment of the frenulum to tip of tongue	1 (1.8)
Type II	Attachment is 2-4 mm behind tip of tongue/on or behind alveolar ridge	8 (14.8)
Type III	Attachment to mid-tongue/middle of the floor of the mouth	23 (42.6)
Type IV	Attachment against base of tongue, thick and inelastic	22 (40.7)

[a]Kotlow (1999). [b]Coryllos, Genna, and Salloum (2004).

Table 2. Maternal Complaints at the Time of Study Enrollment (N = 54).

Symptom/complaint	*n* (%)
Poor latching	49 (90.7)
Creased, flattened, or blanched nipples after nursing	41 (75.9)
Falls asleep while attempting to nurse	36 (66.7)
Gumming or chewing of nipple when nursing	36 (66.7)
Cracked, bruised, or blistered nipples	35 (64.8)
Poor or incomplete breast drainage	34 (63.0)
Slides off nipple when attempting to latch	34 (63.0)
Severe pain when infant attempts to latch	34 (63.0)
Reflux symptoms	30 (55.6)
Short sleep episodes requiring feeding every 2-3 hr	29 (53.7)
Unable to hold a pacifier in mouth	20 (37.0)
Plugged ducts	19 (35.2)
Poor weight gain	17 (31.5)
Colic symptoms	15 (27.8)
Bleeding nipples	14 (25.9)
Mastitis or nipple thrush	8 (14.8)
Infected nipples or breasts	8 (14.8)

of the lip with gauze. Release of the frenulum from the alveolar ridge to the mucogingival junction was then completed.

After completion of the procedure, infants were returned to the parents to offer immediate breastfeeding. Postoperatively, parents completed stretching exercises to avoid tissue reattachment. This was accomplished by gently elevating the tongue for manual wound massage with a sanitized fingertip 4 to 6 times/day for several weeks postoperatively. Postoperative acetaminophen was available for analgesia as needed.

Data Analysis

Statistical analyses were completed using commercial software (SPSS Version 24; IBM Corporation, Armonk, NY, USA). Study data were reviewed descriptively and survey score distributions were evaluated for normality. Mean (standard deviation) values are reported where appropriate. To evaluate within-subjects changes in average breastfeeding survey response over time, level III repeated measures analyses of variance (ANOVAs) were used with Greenhouse-Geisser corrections, as necessary. Covariates were further screened at the .050 alpha level for significant between-subjects effects with each outcome, including age (weeks), gender, race (White/Caucasian vs. non-White/Caucasian), birth type, birth location, Kotlow (1999) classification, Coryllos et al. (2004) classification, and revision surgery type. Comparisons between matched pairs of PROM scores were completed using paired-samples *t* testing or Wilcoxon signed ranking. Relative mean improvement percentages were calculated between preoperative and 1-month postoperative scores to account for variability in preoperative outcome measures. Individual relative mean improvement percentages were determined using the following formula and then averaged: [(1-month score – preoperative score) / preoperative score] × 100. All statistical comparisons assumed a standard .050 Type I error probability.

Results

Sample Characteristics

Enrolled infants ranged in age from 7 days to 37 weeks, with a mean (*SD*) age of 8.3 (8.9) weeks. Most infant participants were found to be of male gender ($n = 28$; 52%) and reported as White/Caucasian ($n = 43$; 80%). Additionally, the majority of infants were delivered vaginally ($n = 44$; 82%) compared with cesarean section ($n = 10$; 18%) and were delivered in a hospital setting ($n = 37$; 69%) compared with at home ($n = 12$; 22%) or birthing centers ($n = 5$; 9%). Revision surgical procedures consisted of only lingual frenotomy ($n = 20$; 37%) or concurrent labial and lingual frenotomy ($n = 34$; 63%). A third frenotomy procedure was not needed for any participant after revision was completed and no operative complications were reported after any revision procedure.

Overall Postoperative Improvement

Significant within-subjects improvement was reported across all PROMs between preoperative and 1-month postoperative

Table 3. Overall Preoperative and all Postoperative Average Outcome Measure Scores ($N = 54$).

	Preoperative	1-week postoperative	1-month postoperative		
Outcome measure	*M* (*SD*)	*M* (*SD*)	*M* (*SD*)	*F*	*p*[a]
BSES-SF total score	45.1 (10.9)	52.1 (12.0)	56.9 (12.2)	41.2	.00*
I-GERQ-R total score	15.7 (6.1)	11.9 (5.2)	10.4 (5.0)	22.7	.00*
VAS breastfeeding pain score	4.8 (2.8)	2.2 (2.1)	1.6 (2.0)	46.1	.00*

Note. BSES-SF = Breastfeeding Self-Efficacy Scale–Short-Form; I-GERQ-R = Revised Infant Gastroesophageal Reflux Questionnaire; VAS = 10-cm Visual Analog Scale.
[a]*p* values reflect omnibus significance between all within-subjects time points using repeated measures analysis of variance *F* test with 2 degrees of freedom.
*$p \leq .01$.

scores (see Table 3). Surgical procedure type was significantly associated ($p = .048$) with differences in I-GERQ-R scores; concurrent labial and lingual frenotomy were associated with lower mean I-GERQ-R scores at all time points overall. Immediate relative improvement was identified 1-week following revision frenotomy on average for BSES-SF total scores (18%), I-GERQ-R total scores (18%), and VAS pain scores (50%).

Subgroup Postoperative Improvements

Postoperative changes were stratified across subgroups to evaluate consistency of surgical effect across gender, anatomic classification, and revision procedures. Significant mean postoperative improvements in BSES-SF total scores were reported 1 month after revision frenotomy by all subgroup classes (see Table 4) with adequate sample size. Significant mean postoperative improvements in I-GERQ-R total scores were reported up to 1 month following revision frenotomy by all subgroups (see Table 5). Likewise, significant average improvements in VAS pain scores were reported up to 1 month following revision procedures by all subgroups (see Table 6). On average, every subgroup reported some degree of relative mean improvement up to 1 month following revision frenotomy, regardless of statistical significance.

Discussion

In the majority of the existing studies, researchers have analyzed the efficacy of frenotomy by using the prevalence of maternal nipple pain as a primary PROM. In this study, we have demonstrated that adding qualitative PROMs about breastfeeding, including reflux disease, maternal breastfeeding self-confidence, and efficiency of milk transfer, is essential.

Classically, examination of the tongue focuses on adequate extension of the tongue over the mandibular gumline, as a marker for normal lingual mobility. Before completing this study, we hypothesized that infants with a prior frenotomy who continue to exhibit pathologic breastfeeding do so because the frenotomy was incomplete (upward motion of the mid-tongue is still limited). In fact, the symptom severity exhibited preoperatively in this cohort (after their initial frenotomy elsewhere) is almost identical to the preoperative symptom set in infants from our previous article who had never received frenotomy (Ghaheri et al., 2017). We have demonstrated symptom improvement with completion frenotomy, allowing for improved upward movement of the mid-tongue. O'Callahan and colleagues (2013) used a similar surgical technique to completely release ankyloglossia in a mixed patient population of infants undergoing initial and revision frenotomy, but the initial and revision groups were not separated for subgroup analysis. Further investigation is needed to determine whether dyads can experience improvement of qualitative PROMs with conservative anterior frenotomy and which infants are at risk for needing more complete lingual release.

Whereas our previous study analyzed infants 0 to 12 weeks of age, this investigation analyzed infants up to 9 months of age who continued to exhibit significant breastfeeding pathology. The persistence of symptoms in older age infants runs counter to the idea that time alone might alleviate symptoms of poor breastfeeding. Furthermore, the older age range of presentation is indicative of a clinical situation in which the family felt that the initial frenotomy done elsewhere had completely eliminated ankyloglossia as a factor in their breastfeeding difficulties. It demonstrated a delay in treatment because the dyad, IBCLC, and breastfeeding specialists have to reevaluate the latch mechanics before discovering the persistence of ankyloglossia. Although we acknowledge the clinical differences in suckling patterns between younger infants and older infants in the cohort, we feel that the postsurgical improvement in the cohort demonstrates a commonality of tongue motion regardless of infant age. Although infants adapt as they get older, it is important to understand that the dyads presented to the clinic with specific and persistent breastfeeding difficulties regardless of infant age and only after involvement of lactation support failed to alleviate symptoms conservatively. In effect, any adaptations made by the dyad were insufficient to avoid presentation to the clinic for additional help with the identified oral restrictions.

Table 4. Comparisons of Mean Preoperative, 1-Week Postoperative, and 1-Month Postoperative BSES-SF Total Scores Stratified by Participant Subgroups.

	Preoperative	1-week postoperative	1-month postoperative		
Subgroup	*M* (*SD*)	*M* (*SD*)	*M* (*SD*)	*F* (*p*)[a]	RMI
Male (*n* = 28)	45.8 (10.5)	52.1 (11.2)	56.0 (12.8)	16.7 (.00*)	26%
Female (*n* = 26)	44.5 (11.7)	52.2 (12.9)	57.8 (11.7)	24.7 (.00*)	36%
Kotlow upper lip-tie classification					
Class III (*n* = 25)	43.6 (10.5)	51.1 (12.1)	57.6 (10.5)	27.8 (.00*)	38%
Class IV (*n* = 22)	46.5 (10.3)	53.8 (10.5)	58.9 (9.4)	22.9 (.00*)	32%
Coryllos tongue-tie classification					
Type II (*n* = 8)	50.9 (10.3)	55.0 (8.2)	60.4 (7.9)	5.9 (.02)	21%
Type III (*n* = 23)	44.7 (11.6)	52.3 (12.1)	55.7 (13.3)	20.8 (.00*)	29%
Type IV (*n* = 22)	43.5 (10.6)	50.4 (13.0)	56.3 (12.4)	14.0 (.00*)	35%
Revision procedure type					
Lingual frenotomy (*n* = 20)	44.5 (11.1)	52.0 (12.2)	55.3 (14.8)	11.9 (.00*)	27%
Lingual and labial frenotomy (*n* = 34)	45.5 (11.1)	52.2 (12.0)	57.9 (10.5)	29.8 (.00*)	33%

Note. BSES-SF = Breastfeeding Self-Efficacy Scale–Short-Form (higher scores on BSES-SF indicate improvement); RMI = relative mean improvement between preoperative and 30-day follow-up scores.
[a]*p* values reflect omnibus significance between all within-subjects time points using repeated measures analysis of variance *F* test with 2 degrees of freedom.
*$p \leq .01$.

Table 5. Comparisons of Mean Preoperative, 1-Week Postoperative, and 1-Month Postoperative I-GERQ-R Total Scores Stratified by Participant Subgroups.

	Preoperative	1-week postoperative	1-month postoperative		
Subgroup	*M* (*SD*)	*M* (*SD*)	*M* (*SD*)	*F* (*p*)[a]	RMI
Male (*n* = 28)	16.4 (5.8)	12.1 (4.4)	10.6 (3.9)	17.1 (.00*)	27%
Female (*n* = 26)	15.0 (6.4)	11.7 (5.9)	10.2 (6.0)	7.2 (.00*)	25%
Kotlow upper lip-tie classification					
Class III (*n* = 25)	14.4 (5.5)	11.0 (4.9)	10.8 (5.1)	5.9 (.00*)	16%
Class IV (*n* = 22)	16.1 (6.4)	12.6 (5.1)	9.2 (3.6)	14.2 (.00*)	36%
Coryllos tongue-tie classification					
Type II (*n* = 8)	17.4 (6.7)	13.0 (4.6)	9.5 (3.9)	5.4 (.02)	37%
Type III (*n* = 23)	14.7 (5.0)	11.4 (4.8)	9.2 (4.9)	10.8 (.00*)	31%
Type IV (*n* = 22)	16.5 (6.8)	12.2 (5.9)	12.0 (5.3)	7.6 (.00*)	19%
Revision procedure type					
Lingual frenotomy (*n* = 20)	17.3 (6.1)	13.3 (5.1)	11.8 (5.8)	9.0 (.00*)	25%
Lingual and labial frenotomy (*n* = 34)	14.8 (5.9)	11.1 (5.1)	9.6 (4.3)	13.3 (.00*)	27%

Note. I-GERQ-R = Revised Infant Gastroesophageal Reflux Questionnaire (lower scores on I-GERQ-R indicate improvement); RMI = relative mean improvement between preoperative and 30-day follow-up scores.
[a]*p* values reflect omnibus significance between all within-subjects time points using repeated measures analysis of variance *F* test with 2 degrees of freedom.
*$p \leq .01$.

Nanishi, Green, Taguri, and Jimba (2015) found that BSES-SF cutoff scores of ≥ 50 were predictive of future breastfeeding success. In our participants, the average postoperative BSES-SF scores improved above this threshold in both our total cohort and across all subgroups, indicating associations with improved maternal breastfeeding self-confidence at the 1-week and 1-month time points. Although we do not have data from dyads prior to the original frenotomy, we feel that it is safe to assume that breastfeeding was problematic enough that an initial surgical procedure was attempted. The preprocedural low BSES-SF scores in this study indicate persistent poor self-confidence about breastfeeding that significantly improved once the tongue was fully released. Future studies correlating postfrenotomy BSES-SF scores with the duration of breastfeeding would help further clarify the importance of frenotomy; we feel that demonstrating the improvement in BSES-SF scores following revision frenotomy is an important first step.

Table 6. Comparisons of Mean Preoperative, 1-Week Postoperative, and 1-Month Postoperative VAS Breastfeeding Pain Scores Stratified by Participant Subgroups.

	Preoperative	1-week postoperative	1-month postoperative		
Subgroup	*M* (*SD*)	*M* (*SD*)	*M* (*SD*)	*F* (*p*)[a]	RMI
Male (*n* = 28)	5.1 (2.9)	2.3 (2.4)	1.9 (2.5)	20.6 (.00*)	38%
Female (*n* = 26)	4.4 (2.8)	2.0 (1.7)	1.2 (1.1)	26.5 (.00*)	62%
Kotlow upper lip-tie classification					
Class III (*n* = 25)	3.9 (2.7)	1.5 (1.8)	1.3 (1.7)	14.5 (.00*)	32%
Class IV (*n* = 22)	6.1 (2.5)	2.6 (2.2)	1.6 (1.8)	44.3 (.00*)	74%
Coryllos tongue-tie classification					
Type II (*n* = 8)	5.9 (2.5)	1.5 (0.9)	0.4 (0.7)	35.1 (.00*)	93%
Type III (*n* = 23)	4.4 (2.9)	2.3 (2.1)	1.9 (2.3)	19.8 (.00*)	52%
Type IV (*n* = 22)	4.8 (2.9)	2.3 (2.5)	1.8 (1.8)	12.2 (.00*)	26%
Revision procedure type					
Lingual frenotomy (*n* = 20)	4.3 (2.8)	2.1 (1.8)	1.8 (2.1)	15.6 (.00*)	53%
Lingual and labial frenotomy (*n* = 34)	5.1 (2.9)	2.2 (2.3)	1.5 (1.9)	31.0 (.00*)	47%

Note. VAS = 10-cm Visual Analog Scale (lower scores on VAS indicate improvement); RMI = relative mean improvement between preoperative and 30-day follow-up scores.

[a]*p* values reflect omnibus significance between all within-subjects time points using repeated measures analysis of variance *F* test with 2 degrees of freedom.

*$p \leq .01$.

Similarly, average I-GERQ-R reflux scores significantly improved following completion frenotomy. A recent review of reflux management guidelines indicated that 82% of physicians overuse proton pump inhibitors in infants (Puntis, 2015). Our previous cohort of infants with ankyloglossia and high reflux scores displayed significant average improvement following frenotomy (Ghaheri et al., 2017). The current cohort had average I-GERQ-R scores that were similar to infants without prior frenotomy, likely the result of incomplete seal formation and aerophagia. Investigators have previously proposed the presence of aerophagia as a mechanism for reflux in infants with ankyloglossia (Siegel, 2016). In these infants, completion frenotomy allowed the mid-tongue to elevate appropriately to the palate, forming a proper seal. Once that seal is obtained and maintained, an intraoral vacuum can be created without allowance of aerophagia. We feel that future studies could be instrumental in demonstrating the importance of full lingual release on latch quality. Although there are technical limitations in visualizing small amounts of intraoral air in the presence of an imperfect seal, further ultrasound studies could be done to show the difference in lingual movement between conservative and complete frenotomy.

One of the primary determinants of successful breastfeeding is the absence of nipple pain. Although a more common symptom early in infant life (Santos et al., 2016), persistent pain is considered abnormal at any age. Maternal nipple pain with shallow latch symptoms is among the most common indications for infant frenotomy (Ghaheri et al., 2017), so improvement in latch quality should improve long-term breastfeeding duration. In this cohort, who had previously undergone frenotomy, the persistence of nipple pain served as the indication that a normal latch was not present. It is important to note that a prerequisite for surgical intervention was evaluation and treatment by an IBCLC to ensure that symptoms were not due to poor latch techniques or inadequate positioning. Improvement in VAS pain scores potentially demonstrated a tangible change in latch quality over time. Following a proper lingual release, the latch should improve, as the seal is formed by the tongue instead of the lips and gums, which in turn should improve breastfeeding pain. Further investigation should be done to help quantify latch depth following completion frenotomy. Researchers have previously demonstrated the capacity to ultrasonographically measure the distance between the nipple and the hard palate–soft palate junction (Geddes et al., 2008). Quantifying the change in this measurement following completion frenotomy could be correlated with latch depth and comfort.

This investigation analyzed mother–infant dyads who presented for evaluation and treatment because of persistent symptoms. Therefore, we could demonstrate that those infants requiring revision frenotomy make up only a small portion of infants who have undergone the procedure. Conversely, the authors cannot determine what percentage of infants who have undergone failed frenotomy subsequently wean without further intervention. The authors also concede that the impact of a maxillary lip-tie release on infants with persistent breastfeeding difficulty following previous frenotomy is unknown. Given the lack of evidence about the impact of lip-tie release, however, the authors attribute breastfeeding improvement primarily to the improvement in lingual mobility. Furthermore, a subgroup analysis of the infants who only received completion lingual frenotomy

without lip release demonstrated significant improvement, isolating the improvement to the tongue release. Although we acknowledge that other nonsurgical options often used in treatment of latch difficulties (e.g., chiropractic, craniosacral therapy, or osteopathic manipulation) exist, the aim of this study was specifically to evaluate postoperative improvements in PROMs following surgical intervention. Furthermore, there is a paucity of published evidence for these nonsurgical modalities. To address this lack of data, future studies could be performed comparing surgical and nonsurgical treatment for qualitative PROMs.

Although we cannot assume that 1 month of follow-up after the procedure is predictive for long-term success, the statistical significance at 1 month, the magnitude of average improvement seen statistically between the 1-week and 1-month time points, and the lack of repeat procedures in any of the infants in the 6 months following the procedure indicate that the revision procedure was helpful in improving PROMs and breastfeeding quality overall. Additionally, the older relative mean ages of this cohort, including some infants up to 9 months old, are evidence against the notion that the improvement was from time alone. Keeping in mind that these infants already underwent frenotomy prior to inclusion in this study, the authors feel that the examination of the cohort before and after intervention reasonably demonstrates the importance of a full frenotomy. Although an age-matched control group would be optimal, this type of study represents the next step in the continued progression of evidence into outcomes of surgical alleviation of ankyloglossia. Future analysis of this patient population would include analysis of rates of breastfeeding, exclusive breastfeeding, and long-term sequelae in infants who undergo frenotomy compared with infants with ankyloglossia who undergo anterior frenectomy alone.

Limitations

There are several caveats to consider when interpreting our study findings. The most obvious is the potential for provider/referral bias. The authors acknowledge difficulty in identifying how many infants underwent anterior frenotomy but went on to experience symptom improvement; those infants were never referred to our clinic for evaluation. This single-arm, longitudinal cohort study is limited in terms of causal association because of the lack of a control population for comparison and determination of relative risk; however, this investigation provides evidence of a consistent treatment effect across subgroups, biological plausibility, and statistical association and supports strong temporal associations between revision frenotomy and improvements in breastfeeding outcomes.

Conclusion

Incomplete lingual frenotomy may cause persistent breastfeeding pathology in infants with previously diagnosed ankyloglossia. Completion lingual frenotomy with or without maxillary labial frenectomy has been associated with significant improvement in measurements of infant reflux, maternal breastfeeding self-confidence, and maternal nipple pain. Demonstrating these associations in children who exhibit less obvious frenulum attachment to the tongue represents a paradigm shift in our understanding of tongue restriction, as it pertains specifically to lingual movement during breastfeeding.

Authors' Note

This study was registered (NCT02642133) at www.clinicaltrials.gov in December 2015.

Declaration of Conflicting Interests

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