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Frenotomy in Infants with Tongue-Tie and Breastfeeding Problems

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Abstract

Background: Infant tongue-tie can cause breastfeeding problems, which may be improved by frenotomy. However, controversy exists among the medical community.

Research aim: To examine the influence of frenotomy on infants with posterior ankyloglossia, by quantifying the changes in breastfeeding and maternal nipple pain using standardized tools.

Methods: Newborns (N = 30) below 12 weeks of age were recruited from the Herzl–Goldfarb Breastfeeding Clinic between April 2014 and April 2015. Diagnosis of posterior ankyloglossia was made clinically. Frenotomy was performed. Mothers received breastfeeding counseling before and after the procedure. Pain and breastfeeding were assessed before the procedure, immediately after the procedure, and after 2 days, 7 days and 14 days. Breastfeeding was assessed using the LATCH Tool and by subjective questioning. Maternal nipple pain was assessed using the Numeric Rating System.

Results: No complications were reported with frenotomy. There was a significant improvement in LATCH score immediately post-frenotomy, with an increase in median scores from 7.5 to 8.5 ($p < .0001$, Wilcoxon signed rank test). There was a significant decrease in median pain score immediately post-frenotomy, from 3.0 on the left nipple and 3.25 on the right nipple, to 0 bilaterally ($p < .0001$, Wilcoxon signed rank test). Subjective improvement in breastfeeding was reported by 90% of mothers immediately after frenotomy and 83% of mothers at Day 14.

Conclusion: Frenotomy for posterior ankyloglossia may improve breastfeeding and nipple pain.

Keywords

ankyloglossia, breastfeeding difficulties, breast pain, LATCH assessment tool, tongue-tie

Background

Ankyloglossia or tongue-tie is a condition where the sublingual frenulum is short, inelastic, thickened, or attached too close to the tongue tip, thus changing the appearance or function of the infant's tongue (Lawrence et al., 2016). This may increase the incidence of breastfeeding difficulties by interfering with how the infant attaches to the breast. Controversy exists as to when and if surgical treatment, or frenotomy, is indicated in these infants.

Breastfeeding difficulties reported with tongue-tie include poor latch, poor milk transfer, maternal nipple pain, milk supply issues, and poor infant weight gain. These may improve with frenotomy, which is a safe and effective procedure according to several studies (O'Callahan et al., 2013; Mettias et al., 2013; Sethi et al., 2013; Garbin et al., 2013; Ito, 2014; Emond et al., 2014; Ghaheri et al., 2017), including a review by the Canadian Agency for Drugs and Technologies in Health (2016). However, O'Shea and colleagues (2017) report that there are no consistent positive breastfeeding effects with frenotomy, although it reduces short-term nipple pain. Since the maternal breastfeeding experience can be difficult with a tongue-tied baby, it is important to accurately identify and treat this condition (Edmunds et al., 2013).

Coryllos and colleagues (2004) described anterior (Types 1 and 2) and posterior tongue-ties (Types 3 and 4), but research on the posterior type is lacking. Haham et al. (2014) examined the incidence of these types but did not find a difference in breastfeeding problems based on type. O'Callahan

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et al. (2013) retrospectively examined anterior and posterior tongue-ties; they found many infants with posterior ankyloglossia had persistent breastfeeding difficulties after initial anterior frenotomy. The breastfeeding issues improved only after the posterior frenotomy was performed. Ghaheri et al. (2017) also found an improvement in breastfeeding after laser frenotomy for anterior and posterior tongue-ties. However, Douglas (2013) questions the widespread diagnosis of posterior tongue-ties and discusses the role of other factors in impaired tongue function (e.g., infant musculoskeletal tensions and breastfeeding positioning).

There is no universally accepted tongue-tie assessment tool. The Hazelbaker tool (Amir et al., 2006) and the Neonatal Tongue Screening Test (Martinelli et al., 2016) are two validated tools. However, they do not include breastfeeding parameters which, in our opinion, is essential. The aim of this study was to determine if frenotomy is a safe and effective procedure that improves breastfeeding and decreases maternal nipple pain.

Key Messages

- There is controversy as to when and whether frenotomy improves breastfeeding for mother–infant dyads with posterior tongue-tie.
- The safety and effectiveness of frenotomy in improving LATCH score, subjective breastfeeding and decreasing nipple pain has been demonstrated.
- We introduce a tool to help diagnose clinically significant tongue-tie necessitating treatment with frenotomy.
- The results of our study add to the body of evidence supporting the use of frenotomy in cases of posterior tongue-tie causing breastfeeding problems.

Methods

Design

A longitudinal prospective one-group pre/post intervention study design was used. This design allowed us to examine the direct influence of the intervention on our outcome variables by comparing data before and after the intervention. The Jewish General Hospital Research Ethics Board approved this study.

Setting

The study took place at the hospital-based Herzl–Goldfarb Breastfeeding Clinic in Montreal, a multicultural Canadian city. Mother–infant dyads are referred by health professionals for breastfeeding problems not resolved with initial support. Physicians and International Board-Certified Lactation Consultants (IBCLCs) work together to see more than 3000 mother–infant dyads a year. The waiting list is 2 to 3 weeks for new consultations. Breastfeeding rates in Montreal are 94% during the first 6 days of life (for any breastfeeding), and 77% by 6 months (Régie de l'assurance maladie du Québec, 2018).

Sample

The target population was breastfeeding mother–infant dyads with posterior tongue-tie and breastfeeding problems. The sample population was restricted to mother–infant dyads referred to the breastfeeding clinic. Inclusion criteria were infant age below 12 weeks, diagnosed with posterior tongue-tie and having persistent breastfeeding problems (e.g., poor latch, nipple pain, poor infant gain and/or poor milk supply) despite lactation support. We chose a cut-off age of 12 weeks because older infants may have more difficulty correcting sucking technique post-frenotomy, possibly making the procedure less useful (Mettias et al., 2013). Posterior tongue-tie was diagnosed by one of the clinic physicians as a restrictive lingual frenulum either submucosal or thickened and posterior, restricting movement at the base of the tongue, corresponding to Coryllos (2004) Types 3 and 4 tongue-ties. Exclusion criteria were diagnosis of anterior tongue-tie (lingual frenulum near or at the tongue tip), gestational age below 37 weeks, neurodevelopmental anomalies (e.g., Down Syndrome, cleft lip/palate), and previous frenotomy.

Recruitment of a convenience sample occurred between April 2014 and April 2015. Mother–infant dyads who fit the study's inclusion criteria were approached to participate. The recruitment was halted when we reached a sample size above 34. A power calculation for sample size was performed before study initiation; a sample size of 34 was necessary to obtain a minimum effect size of 0.5 with the level of statistical significance set to 0.05 and a power of 0.80.

A total of 36 mother–infant dyads were enrolled in the study. All mothers who were approached agreed to take part in the study. The final number of mother–infant dyads who attended all visits was 30 because of loss to follow-up. The power calculation was repeated for a sample size of 30, keeping the effect size at 0.5 and a level of statistical significance of .05. The calculated power was .75.

Measurement

In this study, we used the Frenotomy Decision Tool for Breastfeeding Dyads-version 2012 (FDTBD) (see Supplementary Materials), which is based on an earlier tool, the Frenotomy Decision Rule for Breastfeeding Infants (FDRBI; Srinivasan et al., 2006). The tool was refined using clinical feedback from within the clinic and internationally, to strengthen the tool's content validity. A test-retest procedure was implemented to determine inter-rater reliability. The 2017 version of the FDTBD is in the

process of being validated. Scores range from zero to 10, the higher the score, the more indicative of ankyloglossia needing frenotomy. The first five points (first category) include breastfeeding symptoms, while the second five points (second category) include tongue anatomy and function. The FDTBD was administered to all mother–infant dyads pre-frenotomy only.

Baseline information was recorded on mother–infant dyads at the visit when frenotomy was performed, including maternal age, obstetrical, breastfeeding and medical history, infant age and gender, and timing of frenotomy (initial or subsequent clinic visit).

Breastfeeding was assessed using the LATCH Tool (Jensen et al., 1994) and maternal nipple pain was assessed using the Numeric Rating Scale (NRS) (McCaffery & Beebe, 1993). Both tools have been validated (Riordan et al., 2001; Ferreira-Valente et al., 2011; Altuntas et al., 2014) and used in other studies (Berry et al., 2012). The LATCH tool was chosen because it is simple and fast to administer and has been frequently used in research.

Cronbach's alpha values were calculated for the tools, using the number of complete cases for each variable and excluding cases with missing data. A value of Cronbach's alpha = 0.71 was obtained for the LATCH tool ($n = 29$), and 0.91 for the pain scale ($n = 23$). Cronbach's alpha exceeding 0.90 suggests the items have a relatively high internal consistency with 0.70 considered to be acceptable.

The LATCH tool and pain scale were administered pre-frenotomy, immediately post-frenotomy, and at 2, 7 and 14 days post-frenotomy. Subjective breastfeeding improvement was assessed immediately post-frenotomy and at follow-up visits. Mothers were asked whether breastfeeding felt "better", "the same" or "worse", compared to pre-frenotomy. This follow-up schedule was chosen so we could assess immediate and longer-term effects. Based on our clinical experience, we have observed some mother–infant dyads improve immediately or 2 days post-frenotomy, whereas others take up to 2 weeks.

Data Collection

Mothers signed a consent form written in English and French. All tools were administered by one of three physicians or two IBCLCs at the clinic, all of whom attended an information session by AS (the main physician who conducted the study) to ensure consistency. The tools were in English because they were administered by the clinic staff, who were all fluent in English. All data were stored in a locked cabinet at the breastfeeding clinic and had no identifying names.

The Intervention: Frenotomy

The infant was swaddled in a sheet while an IBCLC held the neck and head in extension with a rolled towel under the neck. The physician used a sterile tongue retractor to lift the tongue and isolate the frenulum. With the other hand, sterile blunt-ended scissors were used to sever the entire membranous part of the frenulum. One main incision was made, followed by one or two smaller incisions on either side of the frenulum if there was any tissue remaining. The baby was then given to the mother to breastfeed.

Parents were taught stretching exercises to be performed three times a day (beginning with the evening of the frenotomy). Post-frenotomy exercises may reduce rates of frenulum regrowth (Demyati et al., 2014). Parents were instructed to lift and stretch the baby's tongue using fingers on either side of the incision, and to gently apply pressure at the site of the incision, for approximately 5 seconds each. The physician examined the infant and demonstrated these exercises at follow-up visits. Parents were also questioned on occurrence of bleeding at home post-frenotomy.

Mothers were seen by an IBCLC or physician pre-frenotomy, immediately post-frenotomy, and at follow-up visits. Lactation counseling and education were given as part of the general care received by all patients of the clinic.

Data Analysis

Descriptive statistics were used to characterize the study population. LATCH score and nipple pain score were measured. Maternal age, infant age at frenotomy, LATCH score, and nipple pain score were analyzed as continuous variables. Other characteristics of the participants were categorical dichotomous (yes/no) variables. Medians and corresponding interquartile ranges (*IQR*) for continuous variables and frequency distributions for categorical variables were calculated. Box-and-whisker plots were built to show medians and *IQR* of LATCH and nipple pain scores pre-frenotomy, immediately post-frenotomy, and at Days 2, 7 and 14 post-frenotomy. To evaluate differences between LATCH scores and nipple pain scores pre- and post-frenotomy, non-parametric Wilcoxon signed rank test was used, given the non-normal distribution of these variables. The correlation between continuous variables was analyzed by Pearson correlation test. Significance level was set at $\alpha = .05$.

Results

Characteristics of the Sample

The mean ($SD = 5.8$) maternal age among participants was 33.7 years (median of 34.1 years, *IQR* of 8.7 and range of 24.3–46.0 years). The mean ($SD = 21.1$) infant age at frenotomy was 37.9 days of life (median of 34.5 days, *IQR* of 34 and range of 9–80 days). Additional baseline demographic data and infant and maternal medical history can be found in Table 1.

FDTBD scores measured pre-frenotomy ranged from 2.5 to 9.0, the mean score was 5.5 ($SD = 1.82$) and the median was 5.0 ($IQR = 3.0$). The median LATCH score for

Table 1. Baseline Data and Medical History of Mother–Infant Dyads, *N* = 30.

Variable	*n* (%)
Male infant	20 (66.7)
Female infant	10 (33.3)
Frenotomy performed at initial clinic visit	15 (50)
Vaginal delivery	21 (70)
Cesarean section delivery	9 (30)
Complications during delivery	6 (20)
Previous breastfeeding experience	9 (30)
Previous breastfeeding problems (for mothers who previously breastfed)	7 (77.8)
Maternal history of breast reduction	2 (6.7)
Maternal diabetes or gestational diabetes	3 (10)
Maternal polycystic ovarian syndrome	2 (6.7)
Maternal history of infertility	4 (13.3)
Maternal history of thyroid issues	3 (10)
Maternal history of mood issues	6 (20)
Maternal high blood pressure	2 (6.7)
Maternal cardiac issues	1 (3.3)

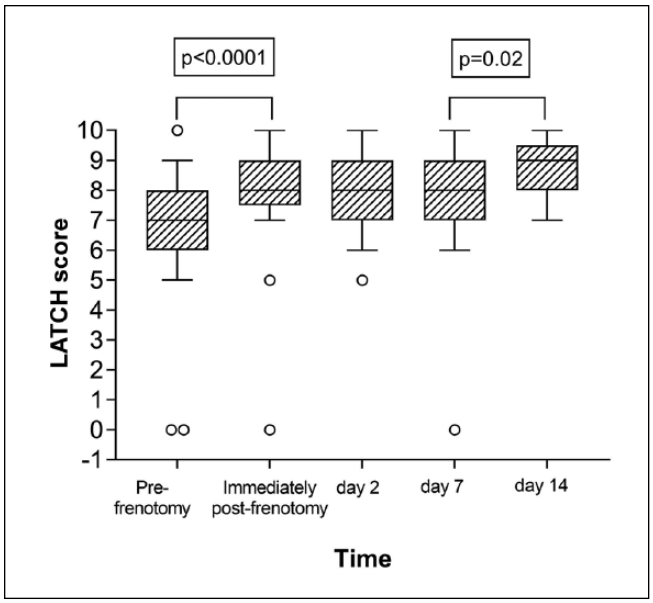

Figure 1. Box-and-whisker plots of the LATCH scores pre- and post-frenotomy.

participants ($n = 30$) at baseline was 7.5 (*IQR* = 2.0) and the mean score was 7.0 (*SD* = 2.2). The median pain score at baseline was 3.0 (*IQR* = 5.0) for the left nipple (measured in 26 patients) and 3.25 (*IQR* = 5.0) for the right nipple (measured in 24 patients).

Post-Frenotomy Outcomes

No complications were noted during or post-frenotomy. Eight parents (22.2%) noted bleeding from the frenotomy site while doing the stretching exercises at home, which they reported at Day 2 and Day 7. By Day 14, only one parent (2.78%) noted bleeding at home. There were no hospital visits or other medical complications related to these episodes of bleeding. All infants were stable when examined by the clinic physician during follow-up visits.

There was a statistically significant and clinically relevant improvement in LATCH score immediately post-frenotomy ($p < .0001$, Wilcoxon signed rank test), as seen on the box-and-whisker plots (Figure 1). There was also a clinically substantial and statistically significant ($p = .02$, Wilcoxon signed rank test) improvement in LATCH score from Day 7 to Day 14. The number of observations for the LATCH score was 29 because some participants were lost to follow-up. Five mother–infant dyads missed a visit, two of whom had stopped breastfeeding. Two other mother–infant dyads attended all visits, but one baby was too sick to latch and the other refused to latch during one of the visits.

There was significant and clinically relevant decrease in median pain scores immediately post-frenotomy ($p < .0001$; Wilcoxon signed rank test), as seen on the box-and-whisker plots (Figures 2A and B). From immediately post-frenotomy to Day 2, there was a slight increase in the upper quartile of *IQR* for nipple pain bilaterally. We found no change in pain scores between Days 2 and 7. By Day 14, there was a slight decrease in the upper quartile of *IQR* compared with Day 7. The number of observations for the pain score was 23 for the right nipple and 28 for the left nipple because of loss to follow-up and missing data. There were some mother–infant dyads who missed appointments or whose baby did not latch as described above. There were two mother–infant dyads whose babies did not latch pre-frenotomy, so pain could not be assessed. However, one of these babies latched well post-frenotomy with no pain. Four mother–infant dyads had babies who latched onto one side but not the other, so pain could only be assessed on one of the sides.

Our analyses showed a strong negative correlation between LATCH scores and right nipple pain scores on Day 2 post-frenotomy ($r = –0.54$, $p = .009$) indicating that mothers who had less pain had higher LATCH score, and vice versa.

Subjective improvement in breastfeeding was noted in 27 mothers immediately post-frenotomy (90%), 23 mothers at Day 2 (77%), 24 mothers at Day 7 (80%), and 25 mothers at Day 14 (83%).

Discussion

The aim of this study was to determine if posterior frenotomy was safe and effective in improving breastfeeding. Our aim was met to various degrees. In line with the findings of other researchers (Mettias et al., 2013; Sethi et al., 2013; Ito, 2014; Emond et al., 2014), there were no

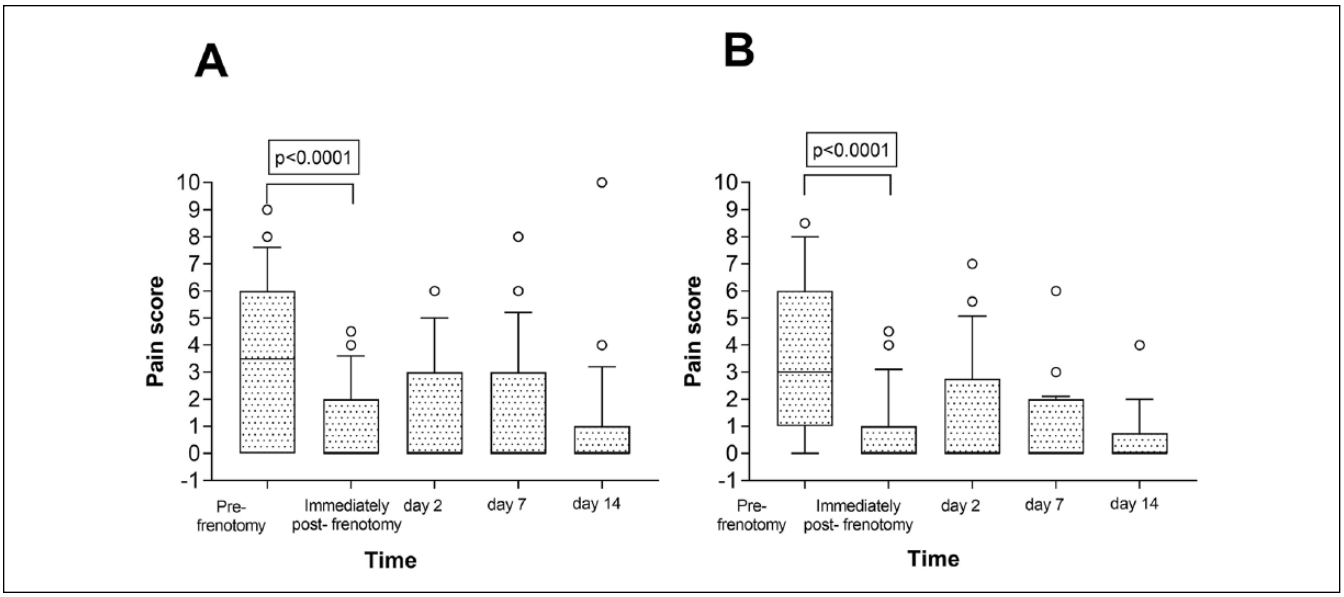

Figure 2. Box-and-Whisker Plots of the Pain Scores Pre- and Post-Frenotomy for Left Nipple (A) and Right Nipple (B).

complications from frenotomy. We studied only posterior tongue-ties and attempted to differentiate between anterior and posterior types, which has only been done in a few other studies (O'Callahan et al., 2013; Ghaheri et al., 2017). The challenge is that the definition of posterior tongue-tie can vary among clinicians. We based ourselves on the Coryllos (2004) definitions of tongue-ties as described above. However, this definition may not adequately select the posterior tongue-ties in which frenotomy is indicated. We chose cases where there were persistent breastfeeding problems, but these problems could be caused by other factors and managed partially with lactation counseling alone. It would be useful to find a way to control for the role of lactation counseling in future studies.

LATCH score

LATCH scores improved post-frenotomy. Improvements in breastfeeding after frenotomy have been observed in previous studies (O'Callahan et al., 2013; Mettias et al., 2013; Sethi et al., 2013; Ito, 2014; Emond et al., 2014; Ghaheri 2017). Although LATCH score was most improved immediately post-frenotomy, it continued to further improve until Day 14. Most participants had an improvement in LATCH score over the course of the study. This could suggest that frenotomy improved the LATCH score, and not other factors (e.g., time, technique, age of baby, or lactation counseling). The LATCH tool includes various non-modifiable elements (e.g., nipple shape). It also includes elements which can be influenced by lactation counseling alone. Therefore, an analysis of the separate items of the LATCH tool would have been useful in isolating the specific influences of frenotomy.

Nipple Pain

Nipple pain also improved substantially immediately post-frenotomy, as seen in previous studies (O'Callahan et al., 2013; Mettias et al., 2013; Sethi et al., 2013; Ito, 2014; Emond et al., 2014; Ghaheri 2017). There is occasionally an increase in nipple pain or sensitivity immediately post-frenotomy, as seen in this study, especially if a baby is not breastfeeding often because of a tongue-tie pre-frenotomy. Once the frenotomy is done, a baby may latch onto the breast more effectively and for longer duration, causing a temporary increase in nipple stimulation and possibly nipple sensitivity or pain. This usually improves with time and lactation counseling. Most mothers had decreased nipple pain over the course of the study. The differences in pain between the right and left nipples may be due to factors including variations in breast or nipple shape and infant musculoskeletal tensions.

The correlation seen between lower pain scores and higher LATCH scores is consistent if we consider the main causes of maternal nipple pain in breastfeeding mothers. One of these is poor or improper latch and breastfeeding. Optimization of the latch and breastfeeding technique can decrease maternal nipple pain by allowing the infant to grasp a larger portion of the nipple and areola and use fuller movements of the tongue (Berens et al., 2016). This would increase the LATCH score.

Subjective Breastfeeding

Most mothers noted a subjective improvement in breastfeeding immediately after frenotomy, and at all follow-up visits until Day 14. The highest percentage of mothers noted the

improvement immediately post-frenotomy, compared to follow-up visits. Since LATCH score and pain had improved at this time, the fact that mothers would subjectively feel an improvement in breastfeeding is consistent. Also, mothers may be more likely to state they are "better" post intervention, especially if they believe this intervention to be useful.

Stretching Exercises

Stretching exercises post-frenotomy, also known as post-frenotomy care, have been described in other studies involving posterior ankyloglossia but have not been shown to be safe and effective in preventing frenulum reattachment (O'Callahan et al, 2013), except for a small study done at our clinic (Demyati et al., 2014). Bleeding from the frenotomy site during home stretching exercises was seen in a minority of patients (eight out of 36) within the first week post-frenotomy, but all were self-limited and did not require hospital or extra clinic visits. Additional studies are needed to determine whether there is an association between regrowth of the frenulum and nipple pain and whether home stretching exercises prevent regrowth.

The Frenotomy Decision Tool for Breastfeeding Dyads (FDTBD)

This tool was introduced in this study. It includes both infant anatomic factors and clinical breastfeeding issues, which is helpful to evaluate the global picture. Since only one criteria from each section is needed for it to be positive, it may be too inclusive as a tool. However, the higher the score, the more likely a frenotomy would be indicated and clinically useful. Having various levels of positivity based on an increasing score may be useful in the future for selecting mother–infant dyads who would benefit the most from a frenotomy, as opposed to those who may benefit less.

Clinical Implications

This study adds to the body of literature on the safety of frenotomy. The fact that an improvement in breastfeeding and a decrease in nipple pain was most apparent immediately post-frenotomy was interesting in view of O'Shea's (2017) findings that only short-term nipple pain is improved with this procedure. Long-term studies of the influence of frenotomy would be very useful.

Limitations

Shortcomings of this study include the lack of a randomized control group and the lack of blinding. All participants received both counseling and frenotomy. Therefore, the separate influences of counseling and frenotomy could not be studied. The improved LATCH score and decreased pain could have been a result of either or both. Changes in specific items of the LATCH tool were not assessed separately.

Infants with structural issues that could affect tongue movement (e.g., plagiocephaly and torticollis), were not excluded from the study. These have been described in the literature as possibly having a detrimental influence on breastfeeding (Kotlow, 2013; Ghaheri et al., 2017). We did not include the evaluation and treatment of labial frenulums, but it is an important research subject for the future. Therefore, these potential confounding factors were not controlled for, and could have affected the results.

An unvalidated tool, the FDTBD, was used in this study. However, study participants were selected using clinical criteria based on the Coryllos (2004) types of tongue-ties. The tool was used as a guide.

The sample size was relatively low. One reason for this was that there were many mother–infant dyads previously treated for ankyloglossia (thus excluding them from the study). Since our breastfeeding clinic acts as a referral centre, all cases are referred by other healthcare professionals who have already seen these patients. There were also several participants lost to follow-up, potentially affecting the results.

Future studies would benefit from a larger sample size, recruiting mother–infant dyads earlier (before having a frenotomy elsewhere), as well as controlling for certain confounders including torticollis.

Conclusion

Frenotomy for posterior ankyloglossic infants with breastfeeding difficulties in this sample was found to be a safe and effective procedure, which improved breastfeeding and decreased maternal nipple pain in the short-term. Further studies are warranted to examine posterior tongue-ties in more detail, and related subjects including post-frenotomy exercises, upper labial ties, and the validation of the FDTBD.

Access to Underlying Research Materials Related to this Paper

The data entry sheets are stored in a locked cabinet at the authors' institution. The patient charts from which these data were obtained are in archives at the authors' institution.

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Declaration of Conflicting Interests

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Investigating Team

The investigating team consisted of AA (medical resident), AS, MS and HM (physicians at the Breastfeeding Clinic), CD and LM (IBCLCs at the Breastfeeding Clinic). Recruitment of participants was carried out by members of the team, and was dependant on their availability at the clinic each day. Data gathering, administration of the questionnaires, and breastfeeding counselling was done by one of the physicians or IBCLCs working at the clinic on a particular day (please see list above). Frenotomies were performed by either AS, MS or HM.

Supplemental Material

Supplemental material for this article is available online.

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