

A Narrative Review and Case Report: Frenotomy Procedure in Neonate with Tongue-Tie

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ABSTRACT

Objective: The objective of this report is to present the case of a neonate who presented to a chiropractor with a tongue-tie causing breastfeeding difficulties as well as evaluate the evidence for the frenotomy procedure.

Design: Case report

Clinical Features: A mother presented to the clinic for breastfeeding difficulties. Upon examination, the three-week-old neonate had clear signs of tongue-tie. The mother also had cracked and bleeding nipples associated with a poor latch caused by tongue-tie.

Intervention and outcome: After evaluation of the case, a frenotomy procedure was suggested to improve tongue function and reduce the breastfeeding difficulties and nipple injury. Following the frenotomy, the pain decreased substantially on latch-on and during the feed. The nipples began to heal as well.

Conclusions: It is becoming more common for neonates with biomechanical dysfunctions affecting breastfeeding to present to a chiropractor. This biomechanical dysfunction along with congenital limitations should be explored to solve breastfeeding issues. This case suggests that the frenotomy procedure can help breastfeeding difficulties linked to tongue-tie. Considering the evidence published in the literature on the effectiveness of frenotomy with very few complications, it is the author's view that patients experiencing breastfeeding difficulties caused by tongue-tie should consider the procedure.

Keywords: neonate, breastfeeding, tongue-tie, frenotomy, chiropractic

Introduction

Mothers around the globe are encouraged to breastfeed according to the recommendations of the World Health Organization that stipulate that infants should be exclusively breastfed for at least the first 6 months of their lives.¹ However, for some mothers this guideline may be difficult to follow when they experience challenges. Mothers with breastfeeding difficulties often present to a chiropractor for an assessment of biomechanical dysfunction interfering with breastfeeding.² During the assessment of the breastfeeding case, the mouth and tongue motion should be evaluated to determine the presence of tongue-tie.³ The International Affiliation of Tongue-Tie Professionals defines tongue-tie as "an embryological remnant of tissue in the midline between the undersurface of the tongue and the floor of the mouth that restricts normal tongue movement."⁴ The tongue-tie's effect on breastfeeding has been associated with sore nipples, poor infant weight gain, neonatal dehydration and shorter breastfeeding duration.⁵⁻⁷ Considering the controversy amongst professionals regarding the frenotomy procedure, a treatment option for tongue-tie (ankyloglossia), this case report and review of the literature

will help determine the necessity of the procedure.^{5,8,9}

Clinical Presentation

A 28-year-old primiparous mother with breastfeeding difficulties presented to the chiropractic clinic. The mother had a vaginal birth at 39 weeks with an epidural injection for pain control. Birth weight of the neonate was 7 pounds 1 ounce. From the first feed, the mother experienced pain and discomfort. The hospital nurse assisted to improve the latch without success. At initial latch-on, the pain was rated by the mother as a 7/10 and went down to 4/10 during the feed. The discharge weight of the baby was 6 pounds 3 ounces, which was approximately equal to a loss of 9% of the neonate's birth weight.

Once home, there was minimal improvement of the latch. Both nipples had started bleeding and cracking. The initial latch-on pain was rated at 7/10 but it reduced to 3/10 as the feed progressed. The mother was nursing 8-10 times per day with no supplementation. The neonate was voiding 6-8 times per day and had 4 stools of yellow color and mustardy texture. The mother contacted her chiropractor who was also an International Board Certified Lactation Consultant (IBCLC) for evaluation and lactation support.

Upon examination, the three-week-old neonate seemed alert and well. The current weight was 7 pounds 3 ounces. He had regained his birth weight and had been

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gaining on average 0.5 ounces per day since birth. Weight gain should be 15-30 gm/day or 0.5 ounces-1.05 ounces per/day. According to the WHO, a fully breastfed girl gains an average of 1,000 grams in the first month, 900 grams in the second month, 700 grams in the third month, and 600 grams in the fourth month of life. A fully breast-fed boy gains an average of 1,200 grams in the first month of life, 1,100 grams in the second month, 800 grams in the third month, and 600 grams in the fourth month.¹⁰

The musculoskeletal assessment revealed restrictions in the mandibular excursion, hypertonicity in the scalenes, sternocleidomastoid, temporalis and masseter muscles. Evaluation of the cranial and vertebral motion using motion palpation and craniosacral technique showed cervical dysfunction at C1 and dysfunction of the parietal, frontal and temporal bones. The mouth and tongue examination revealed the presence of a short, thin lingual frenulum that was attached to the lower ridge of the gum line. Tongue motion was restricted in three planes, elevation and lateral motion were reduced and extension could not reach past the lower gum line. When lifting the tongue, the heart shape associated with a tight anterior frenulum was visible.

The chiropractor, IBCLC assessed the sucking and could feel the lower gum hitting her finger with reduced cupping of the tongue. To evaluate the tongue-tie, the doctor used the Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF) and the Frenotomy Decision Rule for Breastfeeding Infant (FDRBI). The HATLFF function score was 6/14 and the appearance score was 3/10. Many components of the FDRBI tool were positive for the mother-baby dyad. An examination of the mother's nipples revealed bilateral cracks at the base. Following visual examination, the baby was put to breast to assess feeding. Even after latch adjustments, craniosacral treatment and soft tissue therapy, the pain did not decrease. The baby continued to receive craniosacral therapy and chiropractic treatment to the problematic area, but the mother was informed as well that a frenotomy procedure could potentially decrease the pain.^{11,12} To help the mother understand the procedure and make a decision, the question to be answered was does a frenotomy alleviate breastfeeding difficulties in a neonate with tongue-tie?

Literature Review

Method

The starting point for an evidence-based search was the Cochrane Library. The decision was made to start with the terms of the question. The database search was executed with "tongue-tie" and resulted in two randomized

control trials.^{13,14} Searching the database using "ankyloglossia" produced one methodological review,⁹ six trials and one technology assessment from the National Institute for Health and Clinical Excellence.¹⁵ Three of the trials were abstracts only and one was not retained because it used different treatments.^{5,16} Searching with "frenotomy" revealed nothing new.

Another database search using PubMed was done by combining the previous terms with "breastfeeding" using the Boolean operator "and". A first search with "ankyloglossia and breastfeeding" produced 42 papers. Limits of "randomized control trials (RCT), practice guidelines and review" were added which resulted in 9 papers. Most had already been found.¹⁷ A second search with the same limits was performed with "tongue-tie and breastfeeding" and resulted in 15 papers. A third search using the MeSH terms "lingual frenum/abnormalities and lingual frenum/surgery" produced 107 articles. The same limits as earlier resulted in 17 papers most of which were previously found. To assess the risk of complications of the procedure, the PubMed search was performed using the terms "frenotomy and complications". No new articles were retrieved except for a new case series.¹⁸

Results

The search revealed that only a small number of studies such as randomized controlled trials (RCT) and systematic reviews were available. However, even with a small number of studies, the National Institute for Clinical Excellence established the first guidelines in December 2005 and reviewed them in February 2011 without much change. The specialists that wrote these guidelines came from various health fields. The inclusion criteria for studies were well defined. When the guidelines were written only one RCT¹⁴ was available and a few case series.¹⁹⁻²¹ The revised guidelines state that "current evidence suggests that there are no major safety concerns about division of ankyloglossia (tongue-tie) and limited evidence suggests that this procedure can improve breastfeeding. This evidence is adequate to support the use of the procedure provided that normal arrangements are in place for consent, audit and clinical governance."²² The guidelines mention that frenotomy should be performed by registered healthcare professionals who are properly trained and that there is a need for further controlled trials on the long-term effect on breastfeeding. They also state "there were conflicting opinions among the Specialist Advisors and some stated that it is difficult to be certain whether any perceived improvement in breastfeeding is due to division of the tongue-tie." One would assume that this conflict was not strong enough to affect the guidelines recommendations.

Berry et al. (2012) produced a double-blind randomized controlled trial on tongue-tie division in Southampton, UK. The aim of this study was to see if maternally reported improvement in breastfeeding after the tongue tie division was due to the placebo effect. They had 66 babies from 5-115 days old. They were randomized to a division group and a non-division group. The mother and trained observer were blinded. The results showed an immediate improvement in feeding in 78% of mothers in the division group compared to 47% in the non-division group. This study was very well performed however one weakness to note is that they did not grade the tongue-tie. It was therefore difficult to know if these babies had anterior or posterior tongue-tie.

Buryk et al. (2011) produced another randomized single-blinded trial. Newborns with significant ankyloglossia and problems breastfeeding were followed over a 12-month period. Fifty-eight newborns were divided into two groups: 30 infants in the frenotomy group and 28 in the sham group. The diagnostic tool for ankyloglossia used was the HALTF. Other parameters such as the nipple pain scale and the Infant Breastfeeding Assessment Tool were used. They followed up at 2 weeks and regularly over a one year period. Both groups showed statistically significant decreases of pain scores after the intervention but the frenotomy group improved significantly more than the sham group ($p < 0.001$). Breastfeeding scores improved significantly in the frenotomy group ($p = .029$). This appears to be a well executed RCT however, with a small sample size. The authors decided to quantify the nipple pain and the breastfeeding capacity of the newborn which adds value to this study. One major issue with tongue-tie remains identifying which tongue-tie require frenotomy. This study used the HALTF tool since it is the only validated and reliable screening tool that considers function and appearance of the tongue in babies under 6 months.^{23,24} The HALTF tool can be used to determine tongue-tied neonate regardless of the feeding method. However, when working in a breastfeeding clinic where the focus is solely on breastfeeding considering the maternal signs and symptoms are very important factors when deciding to do the frenotomy. In the breastfeeding relationship, one needs to look at the dyad i.e. mother and baby. Since the HALTF tool does not include the mother's complaint or integrity of her nipple to determine if the frenotomy is needed, it would be helpful in a breastfeeding situation to add another tool when determining the need for the frenotomy. Such a tool (FDBR) has been developed to help identify anterior tongue-tie however it has not been validated.²⁵ This tool was also used in a prospective uncontrolled study and it incorporates both the appearance

and function of the tongue and the maternal signs and symptoms. Combining these two tools may help emphasize when the frenotomy is necessary for breastfeeding mothers if the HALTF score is borderline but the mother is experiencing much pain.

In the aforementioned study's discussion, the authors also reviewed the weaknesses, limitations and biases. An important bias was that mothers were motivated to breastfeed. One weakness involved the blinding process. The mothers were asked not to look in their baby's mouth after the procedure and this is very hard to control. Also, all the patients in the control group were offered the procedure at the two-week mark and all but one mother opted for the frenotomy. This eliminated any possibility for more comparison between the groups. This study did not help the practitioner determine what the best timing was to perform the frenotomy but, overall a well-designed study that showed that breastfeeding mothers whose baby were struggling could be helped by a simple frenotomy.

Dollberg et al. (2006) produced a randomized, prospective study that looked at 25 infants with ankyloglossia and mothers with nipple pain. It found "after frenotomy, there was an immediate and significant nipple pain relief as judged by a significant decrease in pain score after frenotomy then after sham"⁵ The trial was blinded and randomized in two sequences: 1) frenotomy followed by breastfeeding followed by sham followed by breastfeeding and 2) sham followed by breastfeeding followed by frenotomy followed by breastfeeding. Throughout the entire process, the authors ensured that the mother was blinded to the frenotomy procedure by having an investigator present during breastfeeding following the procedure. This made sure she did not look in the baby's mouth. The study did not use a validated tool for the identification of the tongue-tie but used a more subjective tool. This would make it difficult for a practitioner in the field to identify which tongue-tie would need frenotomy. Despite its small sample size, the frenotomy procedure had positive results.

The Hogan et al. (2005) study was used in the formulation of the National Institute for Health in Clinical Excellence (NICE) guidelines. The study included 57 babies with tongue-tie of which 40 were breastfed and 17 were bottle-fed. After 4 weeks of normal monitoring, if feeding problems emerged possibly due to a tongue-tie, they were sent for a thorough assessment. They were randomized into two groups: one that offered intensive support, advice and support from the IBCLC (control) and one that had an immediate frenotomy. There were 28 children (20 breastfed

and 8 bottle-fed) in the frenotomy group and 29 children in the control (20 breastfed, 9 bottle-fed). The improvement at 48 hours for the frenotomy group was statistically significant with 27/28 children improving in the division group versus 1/29 in the control group. In the breastfed baby group, 19/20 babies improved with frenotomy whereas only 1/20 improved without frenotomy, a statistically significant difference. All the remaining mothers in the control group, all requested tongue-tie division after the 48 hours of intensive lactation support and 27/28 babies improved. The overall improvement with feeding after frenotomy was 54/57 (95% of infants). This study showed that tongue-tie may also have an impact on either breastfeeding or bottle feeding. The randomization process was well explained. They found that there was no association between tongue-tie length and feeding difficulty meaning that by looking only at the tongue-tie, they could not predict which one was causing breastfeeding difficulties as some babies with 100% tongue-tie were breastfeeding asymptotically. One major weakness involves the identification process of tongue tie where clinical inspection was used. Even with a small number of subjects, it was still able to show statistical significance. It did not use however any quantification method to assess feeding pain. This being one of the first randomized trials, it was a good foundation for future researchers to improve upon.

Steehler et al. (2012) produced a retrospective review of frenotomy in infants with breastfeeding difficulties. This cohort study and retrospective review was done by gathering data and then following up with a telephone survey. The results showed that 367 neonatal and infant consultations were performed for feeding difficulties and 307 infants underwent a frenotomy and 91 mothers participated in the follow up telephone survey. It showed that 80.4% of the mothers believed that the procedure strongly benefited their child's ability to breastfeed, and 82.9% of the mothers were able to initiate or resume breastfeeding after the procedure. A very interesting result is that they showed more benefits when the procedure was done in the first week of life of the infant. This would be interesting to follow up upon to assess what is the best timing to perform the procedure. One weakness of retrospective study is always the recall bias that cannot be removed however an interesting study that looks at the timing of the procedure.

The Cochrane search resulted in a methodological review which was a clear and concise review of the literature on ankyloglossia/tongue-tie⁹ The aim of the study was clearly defined. The database sources were identified and synthesis of the studies well explained. All the articles were

analyzed by two of the authors independently and reached a consensus on the results. Their results showed that a diagnostic criteria is needed to allow better comparison between studies. It also showed that frenotomy is likely an effective treatment but more trials are necessary and that a reliable frenotomy decision rule is needed.

Another well executed literature review by Suter and Bornstein (2009) came to similar conclusions concerning the lack of uniform definitions and classifications. It also questioned which tongue-tie need intervention.

Regarding the risks and safety of the procedure, all the trials reported very few complications. The NICE guidelines list as potential risks: bleeding, infection, ulceration, pain, damage to the tongue and salivary glands. However, several advisors state these as very rare events. They have not been reported in any of the trials except for minor bleeding. One very small case series of only two patients though reported severe bleeding.¹⁸ The conclusion was that frenotomy should be performed by trained professionals to avoid complications. Considering that over 134 babies in the 3 previous trials showed no major complications, one could infer that if performed well, the frenotomy is a safe intervention. The parents should ensure that the professionals are well trained in the procedure before giving their consent to perform the procedure on their neonate/infant.

Interpretation of the Evidence

The evidence found on frenotomy and breastfeeding was discussed with the patient. The decision was made as follows:

- This patient had tried conservative care with an IBCLC with little improvement
- This patient was presently under care to resolve the musculoskeletal components
- Her HATLFF score indicated a need for frenotomy
- Many aspects of the FDRBI tool were present
- The NICE guidelines recommended frenotomy based on the available evidence
- Since these guidelines had come out in 2005, other RCTs had been produced with positive results
- Most of the studies except for a small case series were showing no serious complications associated with the frenotomy
- The NICE guidelines and all the studies recommended that the procedure be performed by trained professionals

Intervention and Outcomes

This mother was referred to a breastfeeding clinic where trained professionals perform the frenotomy procedure daily. The mother received her informed consent and agreed to the procedure. The baby experienced minimal bleeding and remained calm during the procedure. After the frenotomy, the baby went to the breast with a dramatic reduction in maternal pain. Over the next few weeks, the mother's pain continued to decrease and her nipple damage healed. Follow up care for the frenotomy procedure was provided by the breastfeeding clinic and the author continued to follow this neonate for the musculoskeletal problems.

Conclusion

This case showed that frenotomy, if performed by trained professionals, can improve breastfeeding and should be considered to help the mother-baby dyad. This case report demonstrates how the chiropractor can easily apply the appropriate assessment tools along with therapeutic techniques to assess, treat and recommend the appropriate consultations and or interventions for women who are experiencing breastfeeding difficulties secondary to tongue tie in their children.

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Appendix 1. Hazelbaker Assessment Tool for Lingual Frenulum Function

<p><i>Assessment Tool for Lingual Frenulum Function (ATLFF)</i></p> <p>© Alison K. Hazelbaker, PhD, IBCLC March 1, 2009</p>	<p>Mothers name: _____</p> <p>Baby's name: _____</p> <p>Baby's age: _____</p> <p>Date of assessment: _____</p>
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FUNCTION ITEMS

<p><input type="checkbox"/> Lateralization 2 – Complete 1 – Body of tongue but not tongue tip 0 – None</p> <p><input type="checkbox"/> Lift of tongue 2 – Tip to mid-mouth 1 – Only edges to mid mouth 0 – Tip stays at alveolar ridge OR tip rises only to mid-mouth with jaw AND/OR mid-tongue dimples</p> <p><input type="checkbox"/> Extension of tongue 2 – Tip over lower lip 1 – Tip over lower gum only 0 – Neither of the above OR anterior or mid-tongue humps and/or dimples</p>	<p><input type="checkbox"/> Cupping of tongue 2 – Entire edge, firm cup 1 – Side edges only, moderate cup 0 – Poor OR no cup</p> <p><input type="checkbox"/> Peristalsis (progressive contraction) 2 – Complete anterior to posterior (originates at tip) 1 – Partial: originating posterior to tip 0 – None OR Reverse peristalsis</p>	<p><input type="checkbox"/> Snap back 2 – None 1 – Periodic 0 – Frequent OR with each suck</p> <p><input type="checkbox"/> Spread of anterior tongue 2 – Complete 1 – Moderate OR partial 0 – Little OR none</p>
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APPEARANCE ITEMS

<p><input type="checkbox"/> Appearance of tongue when lifted 2 – Round OR square 1 – Slight cleft in tip apparent 0 – Heart shaped</p> <p><input type="checkbox"/> Length of lingual frenulum when tongue lifted 2 – More than 1 cm OR absent frenulum 1 – 1 cm 0 – Less than 1 cm</p>	<p><input type="checkbox"/> Attachment of lingual frenulum to inferior alveolar ridge 2 – Attached to floor of mouth OR well below Ridge 1 – Attached just below ridge 0 – Attached to ridge</p> <p><input type="checkbox"/> Elasticity of lingual frenulum 2 – Very elastic (excellent) 1 – Moderately elastic 0 – Little OR no elasticity</p>	<p><input type="checkbox"/> Attachment of lingual frenulum to tongue 2 – Posterior to tip 1 – At tip 0 – Notched OR under the mucosa at the tongue base</p>
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SCORING

Function Item score: _____	Combined Score: _____ / _____	Appearance Item score: _____
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TREATMENT RECOMMENDATIONS BASED ON SCORING

14 = Perfect Function score regardless of Appearance Item score. Surgical treatment not recommended.
 11 = Acceptable Function score only if Appearance Item score is 10.
 <11 = Function Score indicates function impaired. Frenotomy should be considered if management fails.
 Frenotomy necessary if Appearance Item score is < 8.

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Appendix 2. Frenotomy Decision Rule for Breastfeeding Infants

Mother with nipple pain or trauma while breastfeeding

AND/OR

Inability to maintain latch

AND/OR

Poor weight gain in the infant (<15 g/d)

AND

A visible membrane anterior to the base of the tongue, which restricts tongue movement, leading to:

Inability to touch the roof of the mouth

OR

Inability to cup and examining finger

OR

Inability to protrude the tongue past the gum line

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