

A retrospective review of frenotomy in neonates and infants with feeding difficulties[☆]

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ABSTRACT

Objectives: To measure maternal breast feeding benefit after infant frenotomy. To investigate if timing of neonatal/infant frenotomy affects outcome.

Study design: Cohort survey and retrospective review.

Methods: Medical records of neonates and infants suspected to have ankyloglossia between April 2006 and February 2011 were reviewed. Patient demographic data was compiled. A telephone survey was conducted to gather data on this cohort of patients.

Results: Neonatal and infant consultations ($N = 367$) were performed for feeding difficulties due to suspected ankyloglossia, 302 of these infants underwent frenotomy for ankyloglossia. A total of 91 mothers agreed to participate in a follow-up telephone survey regarding the intervention. Results showed that 80.4% of mothers strongly believed the procedure benefited their child's ability to breastfeed, and 82.9% of mothers were able to initiate/resume breastfeeding after the procedure was performed. The belief that frenotomy significantly benefitted an infant's ability to feed significantly differed in patients that had the procedure performed in the first week of life (86%) as compared to infants that had the procedure performed after the first week of life (74%) ($p < 0.003$).

Conclusions: Based on maternal observations, when frenotomy is performed on neonates with ankyloglossia and feeding difficulties in the first week of life, there is more benefit than when it is performed after the first week of life. The population of patients with ankyloglossia is predominantly male with a high familial/genetic correlation associated with the phenotypic trait. Frenotomy for ankyloglossia demonstrates a high degree of maternal satisfaction, is well tolerated and has been shown to improve breastfeeding and decrease pain and difficulty associated with breastfeeding.

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1. Introduction

Ankyloglossia is a congenital problem characterized by an abnormally short and/or anterior lingual frenulum that can limit the tongue's mobility and limit a neonate's ability to breastfeed. While there is some controversy regarding the clinical significance of ankyloglossia, many lactation consultants and physicians believe that tongue-tie can lead to adverse consequences for both infant and mother including sore nipples, mastitis, poor infant weight gain, and early weaning from maternal breast feeding [1–3]. Although the exact incidence of this condition is unknown, studies

have suggested that between 1.8 and 4.8% of the infant population are affected with a male predominance with a male:female ratio of 2.6:1.0 [4–8].

A paper by Ruben entitled, "Development of Otorhinological Care of the Child" cites that the first documented frenotomy for ankyloglossia with feeding difficulties stems from the infant Dauphin of France, who later became Louis XIII. This child's physician kept a daily diary. In 1601, when the child was 1-day old, his doctor wrote: "September 28th. His nurse was demoiselle Marguerite Hotman and as he seemed to have some difficulty in sucking his mouth was examined and it was found that he was tongue-tie; so at five o'clock in the evening M. Guillemeau, the King's surgeon, cut the tendon three times." Two weeks later, an outcome was documented in the physician's diary: "...when he sucks it is in great gulps so that he swallows as much in one gulp as other babies in three. His nurse never has enough for him [9,10]."

In the 19th century, ankyloglossia was commonly thought to significantly impact breastfeeding, and frenotomies were commonly performed by midwives at the time of delivery [11–13]. As

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formula milk gained popularity, and the popularity of breast feeding declined in the 1940s and 1950s, the procedure fell out of favor since the vast majority of infants with tongue-tie are able to bottle-feed successfully [14]. In the 1980s and 1990s, a correlation between breast feeding and maternal and infantile health was discovered. Further research in developing and developed countries now indicates that breastfeeding (human milk) decreases the incidence of a variety of infectious diseases in infants, including bacterial meningitis, bacteremia, diarrhea, respiratory tract infections, necrotizing enterocolitis, otitis media, and urinary tract infections [15]. Postneonatal infant mortality rates in the United States are also reduced by as much as 21% in breastfed infants [16]. Mothers who breastfeed their infants experience decreased postpartum bleeding and more rapid uterine involution due to increased concentrations of oxytocin, decreased menstrual blood loss, an earlier return to pre-pregnancy weight, decreased risk of breast cancer, ovarian cancer, and possibly decreased risk of hip fractures and osteoporosis in the postmenopausal period [17–24]. The benefits of breast feeding on both infant and maternal health are strongly advocated by the American Academy of Pediatrics, which consider exclusive breastfeeding as the benchmark model against which all other alternative feeding methods are measured with regard to growth, health, and development [25].

Given the infant and maternal benefits of breastfeeding, the population of infants and mothers who are unable to breastfeed have become a population of interest. In the infant population born with ankyloglossia (1.8–4.8% of infants), the literature supports that there is a positive correlation with between their ankyloglossia and breastfeeding difficulties [10,26–28]. Successful resolution of breastfeeding problems have been reported with frenotomy procedures [28–30].

Currently, there are no established criteria or gradation for diagnosing ankyloglossia and tongue tie. Several studies have suggested various guidelines including the length of the lingual frenulum, the amount of tongue movement (difficulty lifting the tongue to the upper dental alveolus, inability to protrude the tongue more than 1–2 mm past the lower central incisors, and impaired side-to-side movement of the tongue) the appearance of a “heart-shaped tongue,” and a thick fibrous cord palpated on physical exam [1,14,31,32,7,33–35]. Lactation literature and clinical scenarios define the need for intervention based on symptomatic complaints of the mother such as discomfort and poor infant weight gain in correlation with the aforementioned infant physical examination findings [35]. In lieu of the compelling evidence in support of neonatal and maternal breast-feeding, this study provides a retrospective review of a large case series of neonates and infants with difficulty latching and breast-feeding. These infants were evaluated for frenotomy based on the degree of neonate and infant ankyloglossia, and if frenotomy was performed, results of the intervention for mother and infant are quantified and compared.

2. Materials and methods

Medical records of neonates and infants suspected to have ankyloglossia between April 2006 and February 2011 were reviewed. A total of 367 neonatal and infant consultations were performed for feeding difficulties due to suspected ankyloglossia. In this population, infant demographic data was compiled; including: age, sex, ethnicity, breastfeeding history (maternal and infant), family history of ankyloglossia, the degree of lingual frenulum restriction (based on ankyloglossia grading scale), and whether or not frenotomy was performed. Using previous hospitalization and clinic records, an IRB approved telephone survey of this patient cohort was performed. The survey consisted

of up to 14 questions involving infant feeding ability, length of breast feeding, and whether or not an intervention was performed based on clinical findings and physical examination. The data was compiled and reviewed for analysis.

2.1. Frenotomy grading system

Infants were first evaluated as either having ankyloglossia or not having ankyloglossia on evaluation from a pediatric otolaryngologist. The patients with ankyloglossia were then classified into one of four types based on the location of tongue-tie using a modified grading system developed by Coryllos et al. [36]. Type 1 was considered to be the most extreme form of ankyloglossia where the lingual frenulum was tethered to the floor of the mouth anteriorly (100% of the tongue is attached to the lingual frenulum) to the extent that the tongue appears as a “heart shape.” Type 2 ankyloglossia consists of tongues that are tethered proximally by the lingual frenulum (75% of the tongue), with restricted elevation and extension of the tongue (Fig. 1). Type 3 ankyloglossia may appear normal, but tongue mobility is limited by a short, bunched frenulum (typically frenulum is halfway to the tip of the tongue). Lastly, type 4 ankyloglossia was classified as limited tongue mobility in the setting of a posterior, fibrous limitation of the most distal portion of the lingual frenulum.

2.2. Description of frenotomy procedure

Informed consent is obtained prior to intervention. The infant is swaddled and held in an upright position by parent or assistant making sure to secure the head in order to prevent lateral motion. Topical viscous lidocaine is applied to the lingual frenulum, which is easily exposed with the aid of two cotton-tip applicators or a Tongue Tie groove director (Pilling and Co. – Philadelphia, PA) (Fig. 2). Sterile iris scissors are used to release the lingual frenulum. The posterior limit of frenulum incision/excision is the anterior genioglossus muscle. At the conclusion of the procedure, the infant is immediately breast fed. Fig. 2 shows an example of pre and post procedure results. When performing the procedure, caution must be taken because orifices of submandibular and lingual salivary glands open under the tongue on the floor of the mouth. The frenotomy must be performed close to the ventral surface of the tongue to avoid injury to the duct lumens.

3. Results

A total of 367 infants diagnosed with ankyloglossia were included in the study, 302 (82.3%) of which underwent frenotomy and 65 (17.7%) of which received no intervention. There were 216 (58.9%) males and 151 (41.1%) females. The mean age was of the



Fig. 1. An example of type 2 ankyloglossia, the most common type of ankyloglossia found in this study.

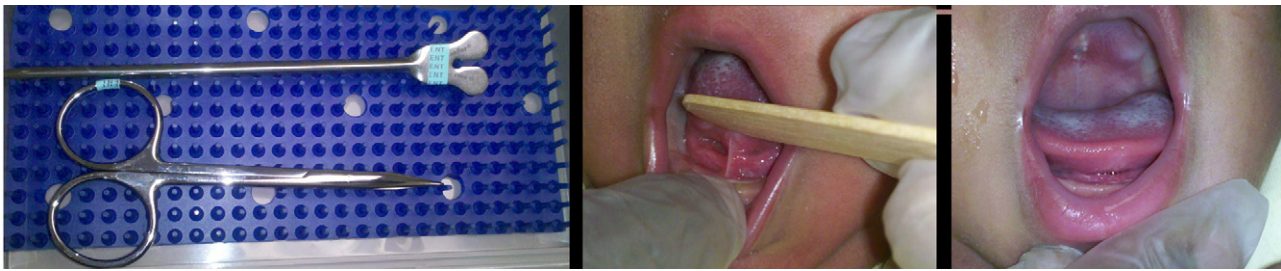


Fig. 2. The frenotomy procedure. (A) Sterile tongue-tie grooved director (Pilling and Co. – Philadelphia, PA) and Iris Scissors. (B) Patient with type 2 ankyloglossia pre-intervention. (C) Post-intervention photograph of same patient, the procedure can be accomplished with minimal bleeding.

infants in this study was 18 days. A family history of ankyloglossia was reported in 127 (34.6%) of the patients, 194 (52.9%) with no family history and 46 (12.5%) that were unsure. Each infant was classified by ankyloglossia grade, of which 64 (17.4%) were type 1, 167 (45.5%) were type 2, 93 (25.3%) were type 3, 18 (4.9%) were type 4 and 25 (6.8%) were indeterminate. A revision procedure was performed on 8 (2.6%) of the frenotomy patients for incomplete clipping or scarring (Table 1).

In the cohort of the 367 infants diagnosed with ankyloglossia, 91 (24.9%) mothers agreed to participate in a telephone survey. Of the 91 respondents, 82 (90.1%) of the infants underwent frenotomy and 9 (9.9%) infants received no intervention. Of the 82 infants that received frenotomy for treatment of ankyloglossia, 66 (80.4%) respondents felt the procedure benefited their child's ability to feed. The percentage of mothers that continued to breastfeed following frenotomy was 82.9% at an average of 7.09 months total, while the percentage of mothers that continued to breastfeed following diagnosis of ankyloglossia without intervention was 66.7% at an average of 6.28 months. When comparing mothers who discontinued breastfeeding due to difficulty or pain due to ankyloglossia, 17.1% stopped in the frenotomy group while 33.3% stopped in the group with no intervention. The average

age for starting solid foods was 5.8 months for the frenotomy group and 6 months for the group with no intervention (Table 2).

The belief that frenotomy significantly benefitted an infant's ability to feed was 86% in patients that had the procedure performed in the first week of life and 74.3% in infants that had the procedure performed after the first week of life. On Student *t*-test, the aforementioned values showed a significant difference ($p < 0.003$), however there was no difference in number of total months breast feeding between the two groups (Table 3). Comparing frenotomy outcomes by ankyloglossia classification, a success rate of 81.3% was seen for type 1, 83.8% for type 2, 85.7% for type 3 and 33.3% for type 4 (Table 4).

4. Discussion

The benefits of breastfeeding are well described in medical literature. This has led to more neonates being brought up exclusively on maternal breast milk, particularly in the first months of life. However, ankyloglossia can adversely affect an infant's ability to breastfeed. Ballard et al. found there to be a 3.2% and 12.8% incidence of ankyloglossia in both inpatient and outpatient infants with breastfeeding problems respectively [30].

Frenotomy appears to effectively treat breastfeeding difficulties secondary to ankyloglossia by decreasing compression of the nipple from the tongue. This leads to better attachment, increased milk transfer, alleviation of nipple pain and improved breastfeeding [37]. Since ankyloglossia appears to play a significant role in early breastfeeding difficulties and frenotomy is a simple yet effective treatment, ankyloglossia should always be considered as a potential cause in the initial assessment of difficulty with breastfeeding [30,38].

Though frenotomy in neonates has been performed for generations, there is limited research and data on the subject. The purpose of this study is to analyze the demographic characteristics of the neonatal and infant population that undergoes frenotomy for ankyloglossia and breast feeding difficulties as well as to analyze the frenotomy procedure and its effectiveness as a therapy in treating infants with difficulty breastfeeding secondary to ankyloglossia. We retrospectively examined the outcomes of each case through chart review and telephone surveying. The primary complaint among this population were difficulties with feeding and latching, along with pain when breastfeeding. Of the 367 diagnosed with ankyloglossia, 302 (82.3%) underwent a frenotomy. The only complication of the procedure was recurrent ankyloglossia secondary to scarring. In 2.6% of cases (8 patients), the lingual frenulum scarred to the floor of the mouth, recreating the patient's ankyloglossia. Repeat intervention successfully treated all 8 of these patients.

Our patient population showed findings consistent with previous studies on ankyloglossia demographics. Klockars reported a significant family history component to ankyloglossia, in particular, an inherited autosomal dominant trait with incomplete penetrance [39]. Our findings support a strong genetic

Table 1
Demographic data and ankyloglossia type distribution of patient population.

Demographics		
Patients with ankyloglossia	367	(100%)
Outcome		
Frenotomy	302	(82.3%)
No intervention	65	(17.7%)
Gender		
Male	216	(58.9%)
Female	151	(41.1%)
Mean age of patient in days	18	
Race		
Caucasian	258	(70.3%)
African-American	57	(15.5%)
Hispanic	16	(4.4%)
Multiethnic	12	(3.3%)
Indian	10	(2.7%)
Asian	6	(1.6%)
Arabic	4	(1.1%)
Persian	2	(0.5%)
Filipino	1	(0.3%)
Unknown	1	(0.3%)
Family history		
Yes	127	(34.6%)
No	194	(52.9%)
Unknown	46	(12.5%)
Type of ankyloglossia		
1	64	(17.4%)
2	167	(45.5%)
3	93	(25.3%)
4	18	(4.9%)
Insufficient data	25	(6.8%)

Table 2

Pertinent survey results of the intervention and no-intervention groups.

Respondents (n=91)	Frenotomy (n=82)	No procedure (n=9)
Frenotomy benefitted ability to feed	66/82 (80.4%)	N/A
Continued to breastfeed after frenotomy	68/82 (82.9%)	N/A
Continued to breastfeed after diagnosis of “tongue tie”	N/A	6/9 (66.7%)
Total months breastfeeding after frenotomy	7.09	N/A
Total months breastfeeding after diagnosis of “tongue tie”	N/A	6.28
Breastfeeding discontinued due to difficulty or pain from “tongue tie”	14/82 (17.1%)	3/9 (33.3%)
Age (in months) when beginning solid foods	5.8	6

Table 3

Frenotomy survey outcome based on age.

	Within first week of life (≤ 7 days)	After first week of life (> 7 days)	Student <i>t</i> -test
Frenotomy benefitted ability to feed	37/43 (86.0%)	29/39(74.3%)	$p < 0.003$
Total months breastfeeding after frenotomy	7.11 months	7.06 months	$p < 0.9$

correlation with ankyloglossia in our population, with at least 34.6% reporting a family history of ankyloglossia [30,39]. Regarding timing of intervention (average), our average age of intervention was very similar to Hong et al., who reported a median age of intervention of 19 days as compared to 18 days with our patient population [40]. It is certainly reasonable to consider frenotomy in the first week of life if there is an appropriate clinical picture. Our study shows a significant difference in the telephone survey in the patients/mothers believing their infant benefited from the procedure if it was performed in the first week of life (Table 3). Although patients did not demonstrate a significant difference the number of total months breast feeding, the early benefit of frenotomy in breast feeding in the first few months is not measured with this variable. Future iterations of this study could monitor infant weight gain or infant per os intake pre and post procedure for objective measurements. Buryk et al., also used the ‘Infant Breast Feeding Assessment Tool’ (IBFAT) to obtain maternal subjective opinion in the setting of sham surgery and frenotomy: showing there was a significant improvement in the IBFAT score after frenotomy [28]. There is also said to be a higher predominance of ankyloglossia in males (2:6 M:1 F) [4–8,38,39]. In our patient population, we found a similar predominance in males, with 58.9% being male and 41.1% being female.

Ankyloglossia can be categorized into four types; type 1 which is classical heart-shaped; type 2 which is classical; type 3 which is at the middle of the floor of the mouth, tighter and less elastic; and type 4 which is at the base of the tongue. Types 1 and 2 are reported to be more common and readily managed, while types 3 and 4 are less common and usually require digital exam for diagnosis [35,40]. Types 1 and 2 were diagnosed in 62.9% of our patient population. Types 3 and 4 were diagnosed in 30.2% of our population (Table 4). It was noted that most of our patients were referrals and a recent article by Hong et al. confirms that posterior ankyloglossia (types 3 and 4) is poorly recognized due to its subtle findings and the need for digital physical exam [40]. The abundance of anterior (types 1 and 2) ankyloglossia in this study could be a result of the challenge in diagnosing and referring posterior ankyloglossia. In comparing the outcome of frenotomy by ankyloglossia types, results in types 1, 2 and 3 yielded similar results, with approximately 83% of mothers reporting an improvement in their infant’s ability to feed. Type 4 yielded a

lower result of 33%, however, the sample size of respondents with type 4 ankyloglossia was low, which may account for the low success rate in type 4 patients. It has also been shown that infants with posterior ankyloglossia do not fare as well with the frenotomy procedure as well as those with anterior ankyloglossia [40].

In analyzing the effectiveness of frenotomy as a therapy for treating infants with breastfeeding secondary to ankyloglossia, the mothers surveyed expressed great degree of satisfaction in follow-up. Of those surveyed, 80.4% strongly believed the procedure benefitted their child’s ability to breastfeed. When comparing mothers who continued to breastfeed, 82.9% continued in the frenotomy group as opposed to 66.7% in the no intervention groups. It should be noted that some of the patients did not continue to breastfeed following the frenotomy procedure due to reasons not associated with ankyloglossia. A few infants in the study suffered from problems such as a retracted jaw, high palate and cleft lip while a few mothers in the study coincidentally suffered from problems such as multiple sclerosis, Reynaud’s of the nipple, thrush, insufficient milk production and appendicitis.

The average age at which breastfeeding was discontinued improved in the frenotomy group (7.09 months) when compared to the control group (6.28 months). Also, the number of mothers who stopped breastfeeding due to difficulty or pain from ankyloglossia decreased by nearly 50% when comparing the frenotomy group (33.3%) to the control group (17.1%). The benefit frenotomy that can be maximized if frenotomy is performed in the first week of life (Table 3). Other studies confirm the importance that frenotomy be performed early. Ricke et al. found that affected infants were significantly more likely to be exclusively bottle-fed by 1 week of age with more challenging rehabilitation back to breast feeding [27]. While the procedure was still largely successful for those having the frenotomy procedure done after 1 week of age, the ‘benefit rate’ dropped more than 10%. When comparing the age for starting solid foods, the frenotomy group began at an average of 5.8 months while the control group began at an age of 6 months; an improvement of approximately 1 week. Post-frenotomy improvements in breastfeeding were more significant than that of solid foods, most likely because of the importance of the tongue in latching onto the nipple in breastfeeding.

Limitations of this study include a lack of sample size in the ‘no intervention’ group. This was based on a lack of cooperation with

Table 4

Frenotomy outcome based on type of ankyloglossia.

Type	1	2	3	4
Frenotomy benefitted ability to feed	13/16 (81.3%)	31/38 (83.8%)	18/21 (85.7%)	1/3 (33.3%)

the survey and incomplete patient follow up data. The hypothesized way to achieve more meaningful numbers in the 'no intervention' group would be to carry out the study over a longer period of time to accrue a larger sample size and to provide incentive for this population to complete the telephone survey (however, this study had no financial support and therefore could not offer incentive for completion of the telephone survey). Authors of this paper believe that the population that refuses medical intervention is usually less compliant with telephone surveys. In discussing the frenotomy in consultation with the 'no intervention' group and, later, the telephone survey, it was clear that this group was more likely to say 'no' to medical personnel. Another limitation to consider in this study is the duration from the time of frenotomy to the time of the telephone survey (telephone survey conducted in 2011). This ranged from 3 months to 5 years. Those who were surveyed further out from intervention may have had a harder time recalling their experience with breast feeding following frenotomy. When reviewing the data, responses among our sample population were consistent over time, with no clear trends in response directly related to the duration between intervention and telephone survey.

Future studies are warranted to further evaluate improvements in breastfeeding from frenotomy in infants with ankyloglossia. Additional research on this topic, including a prospective controlled study would be of benefit to investigate the findings discovered in this retrospective study.

5. Conclusions

Based on maternal observations, when frenotomy is performed on neonates with ankyloglossia and feeding difficulties in the first week of life, there is more benefit than when it is performed after the first week of life. Regardless of timing of intervention, frenotomy for ankyloglossia helps to improve breast feeding for infant and mother. The population of patients with ankyloglossia is predominantly male with a high familial/genetic correlation associated with the phenotypic trait. Frenotomy for ankyloglossia demonstrates a high degree of maternal satisfaction, is well tolerated and has been shown to improve breastfeeding and decrease pain and difficulty associated with breastfeeding. Further evaluation into the benefits and outcomes of frenotomy on this patient cohort may include determining its effects on speech and language development.

Conflict of interest

There was no financial support or funding for this study. The authors have no financial obligations to disclose and there are no conflicts of interest.

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