

Factors Affecting Frenotomy: A Single Institution's Experience with Ankyloglossia

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Abstract

Aim

Ankyloglossia, or 'tongue-tie', is a congenital anomaly in which a short lingual frenulum restricts tongue movement. The effects of ankyloglossia can include breastfeeding and articulation issues, with frenotomy being the surgical intervention of choice. Small infants necessitate an urgent referral to the ear nose and throat (ENT) clinic to facilitate breast feeding and weight gain. We sought to examine the ankyloglossia service in a secondary general hospital from referral to outpatient clinic and analyse factors affecting eventual outcome.

Methods

We retrospectively analysed a cohort of infants referred to the ENT service over a 24 month period. We analysed data from referral including demographics and clinical information and recorded information from the clinical consultation and procedure details if frenotomy was performed.

Results

Between 1st January 2019 and 31st January 2021 47 referrals were made for consideration of frenotomy. These referrals were from a variety of healthcare professionals. Of 47 referrals, a frenotomy was performed in 30 infants.

Conclusion

Information on ankyloglossia is varied without any standardised guideline or treatment algorithm. In our practice, frenotomy was required and performed more frequently in breastfed babies with failure to thrive. Referral indications can be unclear and result in overdiagnosis, overtreatment and unnecessary referrals.

Keywords: Tongue tie, Ankyloglossia, ENT

Introduction

Ankyloglossia is a congenital condition in which a short lingual frenulum restricts tongue movement. During breastfeeding, restricted tongue movement can impact an infant's ability to latch¹. As a result, these infants may be unable to adequately meet nutritional requirements. Improper latch may also cause maternal nipple pain which may lead to early cessation of breastfeeding². The majority of infants with ankyloglossia remain asymptomatic³. The prevalence of ankyloglossia is estimated at between 4-10%⁴, with a noted male preponderance. The lack of a single, standardised diagnostic assessment tool means that ankyloglossia poses a diagnostic challenge for clinicians. This challenge has been acknowledged by the recent Clinical Consensus Statement released by the American Academy of Otolaryngology regarding ankyloglossia in children⁵. When ankyloglossia is diagnosed, clinicians may use a number of tools to aide their assessment including: the Hazelbaker Assessment Tool for Lingual Frenulum Function and the Bristol Tongue-Tie Assessment Tool⁶. The Health Service Executive (HSE), have released a 'tongue-tie' assessment proforma⁷, while also advocating for the use of Martinelli's 'Lingual Frenulum Protocol for Infants'⁸. These tools may aid in clinical management and diagnosis including classification of ankyloglossia. The HSE proforma describes ankyloglossia as anterior or posterior.

The management options for ankyloglossia include conservative management or frenotomy. Where conservative management, such as education and breastfeeding support fails or is not appropriate, infants are referred for frenotomy. Frenotomy is defined as division of the lingual frenulum⁷. In small infants this procedure is well tolerated in an outpatient setting without the need for general anaesthesia⁹. Frenotomy in older infants typically requires general anaesthesia, increasing the risks associated with the procedure. As a result, all infants with ankyloglossia are given high priority in the outpatient setting. The paediatric societies of the United Kingdom¹⁰ and Canada recommend surgical intervention only when there is a clear indication that a frenotomy would benefit the infant's feeding¹¹. The National Institute of Health and Care Excellence (NICE), has stated that while the evidence to suggest that a frenotomy can improve breastfeeding is limited, the current evidence is *"adequate to support the use of the procedure provided that normal arrangements are in place for consent, audit and clinical governance"*, while also suggesting that breastfeeding issues may be alleviated following the procedure¹².

Despite the diagnostic challenge for clinicians, there has been an increase in the number of children diagnosed with, and treated for ankyloglossia in the last number of years. Between 2003 and 2012, the number of infants diagnosed with ankyloglossia increased fourfold, while the number of surgically managed cases has increased five fold¹³. The Clinical Consensus Statement released by the American Academy of Otolaryngology regarding ankyloglossia has recognised there has been overdiagnosis and overtreatment of infants in some communities⁵. The primary aim of this study was to assess the factors which may influence a clinician's decision to either perform or not to perform a frenotomy within our institution in the hope to better standardise practice.

Methods

We performed a retrospective study analysing a cohort of infants referred to our ENT service with ankyloglossia. Ethical approval was obtained prior to commencement of the study. Infants suitable for inclusion were identified through outpatient clinic records. Inclusion criteria were defined as infants presenting to our ENT service between 1st January 2019 and 31st January 2021, of age younger than eighteen months, with ankyloglossia confirmed by ENT clinician. Infants were excluded on the basis of additional congenital abnormalities, comorbidities impacting ability to feed or swallow, age and incomplete data.

Diagnosis of ankyloglossia was confirmed by consultant assessment. This included the presence of a sublingual frenulum inserting at or near the tip of the infant's tongue. It also included the presence of a shortened frenulum that impacted the tongue's appearance, function or mobility as revealed by impaired side to side movement or inability of the examiner to place a finger between tongue and mandibular alveolus. Data was collected including: infant age, sex, primary method of feeding, prior history of ankyloglossia in a sibling, referral method (healthcare professional referring) and if frenotomy was performed. Where documented in the healthcare record, gestational age at birth, birth weight and location of ankyloglossia was recorded. We recorded the primary reason for referral to the ENT clinic which included maternal (nipple pain/ discomfort) and infant issues. Infant issues were subcategorised as: failure to thrive, restriction of tongue mobility or latch/feeding issues.

Discrete data is represented in frequency tables with proportions in parentheses. For continuous variables mean value is presented with standard deviation in parentheses. For the purpose of this study $p < 0.05$ was considered statistically significant. Initial analysis assessed the clinical characteristics and presenting features of infants to our clinic. We subsequently performed an analysis looking at the factors associated with a frenotomy being performed. Pearson's Chi-squared test was used to compare categorical variables. Continuous variables were compared using Levene's equality of variance test. All statistical analysis was carried out using IBM SPSS (ver 27).

Results

There were forty seven infants included in this study. Twenty nine infants (61.7%) were males while eighteen (38.3%) were female. The mean age at time of diagnosis was 8.3 weeks (SD \pm 6.6 weeks). There were twenty eight breastfed infants (59.6%) while nineteen infants (40.4%) were bottle-fed. Mean birth-weight was 3.4kg (SD \pm 0.61kg) while mean gestational age at time of birth was 38.5 weeks (SD \pm 2.1 weeks). A family history was identified for eleven infants (23.4%) while thirty six infants had no family history of ankyloglossia. Frenotomy was performed for thirty infants (63.8%) while seventeen infants (36.2%) were managed conservatively. (Table 1).

Table 1. Clinical and Presenting Characteristics.

	Total Patients (%) (n=47)
Sex	
Male	29 (61.7%)
Female	18 (38.3%)
Age at diagnosis (weeks)^a	
	8.3 (±6.6)
Feeding method	
Breastfeeding	28 (59.6%)
Bottle feeding	19 (40.4%)
Birth weight (kg)^{a,b}	
	3.4 (±0.61)
Gestational Age At Birth^b (weeks)	
	38.5 (±2.1)
Sibling History Ankyloglossia	
Yes	11 (23.4%)
No	36 (76.2%)
Referral Method	
General Practitioner	12 (25.5%)
Lactation Consultant	5 (10.6%)
Public Health Nurse	26 (55.3%)
Paediatric Consultant	4 (8.5%)
Maternal Issue	
None	24 (51.1%)
Nipple pain/discomfort	23 (48.9%)
Infant Issue	
None	12 (25.5%)
Poor Latch/Feeding issue	30 (63.8%)
Failure to thrive	3 (6.4%)
Poor Tongue Mobility	2 (4.3%)
Ankyloglossia Location	
Anterior	20 (42.5%)
Posterior	14 (29.8%)
Not Recorded	13 (27.7%)

^a Mean value with Standard Deviation (SD) in parentheses

^b Incomplete data

There were twenty anterior (42.5%) and fourteen posterior (29.8%) cases of ankyloglossia while this data was not documented in thirteen cases (27.7%). The majority of referrals (n=26) to our clinic were made by a Public Health Nurse (55.3%). General practitioners accounted for twelve referrals (25.5%), lactation consultants for five referrals (10.6%) while paediatric consultants accounted for four referrals (8.5%). There were thirty-five referrals citing infant issues, with thirty infants (63.8%) referred due to reported issues with latch or other feeding issues such as slow feeding or aerophagia. Three infants (6.4%) were referred with failure to thrive and two infants (4.3%) were referred with poor tongue mobility. Twelve infants (25.5%) had no reported issues upon referral and no reported symptoms at clinic assessment. Maternal nipple pain or discomfort was present in twenty three cases (48.9%). (Table 1).

Patients with an anterior ankyloglossia were significantly more likely to undergo frenotomy ($p < 0.05$). Infants who underwent frenotomy were on average 2.2 weeks younger than those who did not. A higher proportion of infants undergoing frenotomy were breastfed (66.7% v 47.1%). A higher proportion of infants who underwent frenotomy were referred by a lactation consultant or a GP while infants referred by a PHN were most likely to not undergo the procedure. A higher proportion of infants referred with a maternal issue related to feeding underwent frenotomy. With regards to infantile issues at presentation, two infants with failure to thrive underwent frenotomy while one referred with failure to thrive did not undergo the procedure. No association was noted between gestational age at birth, birth weight, sex or sibling history of ankyloglossia and infants undergoing the procedure. (Table 2). There were no complications recorded for any infant undergoing frenotomy.

Table 2. Factors Associated with Frenotomy.

	Frenotomy (%) (n=30)	No Frenotomy (%) (n=17)	P value*
Sex			0.750
Male	18 (60%)	11 (64.7%)	
Female	12 (40%)	6 (35.3%)	
Age at diagnosis (weeks)^a	7.5 (± 6.3)	9.7 (± 6.9)	0.537 ^c
Feeding method			0.188
Breastfeeding	20 (66.7%)	8 (47.1%)	
Bottle feeding	10 (33.3%)	9 (52.9%)	
Birth weight (kg)^{a,b}	3.4 (± 0.61)	3.3 (± 0.58)	0.579 ^c
Gestational Age At Birth^b (weeks)	38.8 (± 1.9)	38.1 (± 2.2)	0.704 ^c
Sibling History Ankyloglossia			0.156
Yes	9 (30%)	2 (11.7%)	
No	21 (70%)	15 (88.3%)	

Referral Method			0.239
General Practitioner	8 (66.7%)	4 (23.5%)	
Lactation Consultant	5 (16.7%)	0 (0%)	
Public Health Nurse	14 (46.7%)	12 (70.6%)	
Paediatric Consultant	3 (10%)	1 (5.9%)	
Maternal Issue			0.159
None	13 (43.3%)	11 (64.7%)	
Nipple pain/discomfort	17 (56.7%)	6 (35.3%)	
Infant Issue			0.524
None	6 (20%)	6 (35.3%)	
Poor Latch/Feeding issue	20 (66.7%)	10 (58.9%)	
Failure to thrive	2 (6.7%)	1 (5.9%)	
Poor Tongue Mobility	2 (6.7%)	0 (0%)	
Ankyloglossia Location			<0.05
Anterior	18 (60%)	0 (0%)	
Mid	1 (3.3%)	1 (5.9%)	
Posterior	2 (6.7%)	12 (70.6%)	
Not Recorded	9 (30%)	4 (23.5%)	

*Pearson's Chi-Squared test unless otherwise specified

^a Mean value with Standard Deviation (SD) in parentheses

^b Incomplete data

^c Levene's equality of variance test

Discussion

Our analysis found that infants with an anterior tongue-tie were significantly more likely to undergo frenotomy versus those with a mid or posterior tongue-tie ($p < 0.05$). This is concordant with current international recommendations. Evidence supports that anterior tongue-tie can impact on feeding and that frenotomy may be of benefit in these infants¹⁴. At present, there is no evidence to support that performing a frenotomy in patients with a posterior tongue-tie is of any benefit to the infant, while concurrently exposing them to the potential risks of the procedure, including infection, bleeding, scarring and salivary duct damage¹⁵. Previous studies have shown that ankyloglossia can be associated with poor feeding by reducing an infants' ability to latch properly^{16,17}. Thus, they are unable to adequately meet their nutritional requirements leading to failure to thrive. Within our study we found a higher proportion of infants presenting with ankyloglossia associated with failure to thrive underwent frenotomy than those who had no signs of failure to thrive. Previous studies have demonstrated that frenotomy is of more benefit in those infants that are breastfed. In our analysis, a higher proportion of breastfed infants referred to our clinic with ankyloglossia underwent frenotomy than those who were bottle-fed although this did not reach statistical significance. We observed that infants who underwent frenotomy tended to be younger than those who were managed conservatively. Again, this is consistent with findings from previous studies¹⁸. In agreement with current evidence, we advocate that breastfed infants with anterior tongue-tie and failure to thrive undergo frenotomy at an early stage.

Over one-third of infants referred to our ENT outpatient clinic for consideration of frenotomy did not undergo the procedure. This finding is encouraging given the desire for clinicians to avoid unnecessary invasive procedures in all patients. Studies have demonstrated that many infants with ankyloglossia are asymptomatic¹⁹, and so do not require further assessment or treatment. The rate of frenotomy performed on infants referred with suspected ankyloglossia to a given clinic or institution varies within the literature^{14,18}. Inappropriate screening and referral of infants with suspected ankyloglossia in the community will lead to infants attending clinic who do not need surgical treatment. In Ireland in 2021, there were 67,980 patients waiting to be seen as outpatients at various ENT clinics around the country, with 31,853 of these patients expected to be waiting more than eighteen months²⁰. Infants referred with suspected ankyloglossia are seen and treated on an urgent basis in line with international best practice. If ankyloglossia is impacting on an infant's ability to feed and develop, it is vital that this be corrected as soon as possible. However, referral of infants with suspected ankyloglossia that can be managed conservatively has a significant opportunity cost. This includes potential delays in seeing patients with suspected head and neck cancers who also necessitate urgent appointments. The HSE has attempted to aid clinicians in screening infants with suspected ankyloglossia through dissemination of a HSE ankyloglossia assessment proforma. We would advocate the use of this proforma to both appropriately identify which infants require further assessment and treatment and to avoid referral of infants who do not need treatment of their ankyloglossia. As the rate of breastfeeding increases among Irish mothers²¹, we can expect to see the number of ankyloglossia referrals rise in tandem.

The other important determination on the rate of frenotomy within an institution is the clinicians themselves. With a lack of concrete diagnostic and therapeutic criteria, studies have demonstrated significant inter-clinician variability in deciding whether or not to perform frenotomy in infants with ankyloglossia²². This affects not only referral patterns, with some paediatricians or general practitioners not referring babies with ankyloglossia, but also the rate of conversion to a procedure by the treating clinician. This is particularly true of posterior tongue tie, where the evidence for release is even more controversial.

Our study has several limitations. This study has small numbers. The figures used in our study are small, due in part to the fact this study was conducted in a single centre where the numbers of babies referred for review of ankyloglossia are relatively small. This may have led to underestimation of the effect various factors had on clinicians' decision to perform or not to perform frenotomy. We would advise this study be repeated on a larger scale across numerous centres, to increase the number of patients included in the study. The resulting figures would be a better representation of the current rate of frenotomy in Ireland. Private referrals were not included in our study. This potentially had a significant impact on numbers as one to two frenotomies per week were being performed privately during the period we studied. The only post procedure information collected was procedural complications. No follow up outcome measures were obtained. Common practice in our institution is that these patients are discharged after a successful procedure and are given information to rebook an appointment should any problem arise.

Following up at four weeks and six months would provide additional follow up data and would add to our information but this is not the focus of our study. This study was performed retrospectively which may have led to bias within the analysis. As previously discussed there is no standardised diagnostic criterion for ankyloglossia. This may have led to some infants included in the analysis after being misclassified with ankyloglossia.

In conclusion, we demonstrated that within our institution, infants with anterior tongue-ties, failure to thrive and breastfed infants were more likely to undergo frenotomy. We demonstrated that over one-third of infants referred to our clinic with suspected ankyloglossia did not require frenotomy. With current long ENT clinic waiting lists in Ireland, it is important that infants with suspected ankyloglossia are properly assessed in the community using appropriate tools such as the HSE assessment proforma to avoid overdiagnosis and overtreatment.

Declaration of Conflicts of Interest:

The authors report no conflicts of interest in this work.

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