

Friend or Frenotomy: A Single Institution's Experience With Ankyloglossia

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Abstract

Introduction/Aim

Ankyloglossia, or 'tongue-tie', is a common congenital anomaly in which a short lingual frenulum or genioglossus muscle restricts tongue movement. Ankyloglossia can be graded from 1 (most severe) to 4 (least severe). The effects of ankyloglossia can include breastfeeding and articulation issues; however, many infants will have no symptoms or developmental problems. The surgical intervention for ankyloglossia is frenotomy. This can be performed in the outpatient setting in small infants. Ankyloglossia referrals in neonates and small infants necessitate an urgent referral to the ear nose and throat (ENT) clinic in order to facilitate breast feeding and weight gain. We sought to analyse the ankyloglossia service in a district general hospital setting from referral to outpatient clinic.

Methods

We retrospectively analysed a consecutive cohort of babies referred to the Ear Nose and Throat service for consideration of frenotomy over an 18 month period, We analysed data from referral including demographics and clinical information, we recorded information from the clinical consultation and procedure details if frenotomy was performed.

Results

Between 1 January 2019 and 31 January 2021 referrals were made for consideration of frenotomy, all appointments were seen within 2 weeks. 55.3% of referrals were sent from public health nurses, 25.5% from primary care, 10.6% from lactation consultants and 8.5% from paediatric consultants. Of 47 referrals, a frenotomy was performed in 30 babies. All frenotomies were performed without complications.

Conclusion

Information on ankyloglossia is varied and available information is conflicting, without any clear standardised guideline or treatment algorithm. Referral indications can be unclear and result in unnecessary clinic appointments in an already heavily burdened service. Frenotomy can be performed safely by a trained clinician in an outpatient setting with minimal equipment.

Introduction

Ankyloglossia, or 'tongue-tie', is a congenital anomaly in which a short lingual frenulum restricts tongue movement. Restricted tongue movement in an infant can result in a number of complications for mother and infant. During breastfeeding, restricted tongue movement can impact an infant's ability to both properly latch to bottle or breast for feeding and to swallow feeds¹. As a result, these infants may be unable to adequately meet nutritional requirements for growth and development. Improper latch may also cause maternal nipple pain which may lead to early cessation of breastfeeding². However, many infants

with ankyloglossia remain asymptomatic³. The prevalence of ankyloglossia is estimated between 4–10%⁴, with a noted male preponderance. However, the true prevalence is difficult to ascertain due to the absence of practical diagnostic criteria and the number of asymptomatic patients who never present to healthcare services. The lack of a standardised diagnostic criterion means that diagnosis of ankyloglossia poses a diagnostic challenge for clinicians. This diagnostic 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 Breastfeeding 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. According to this proforma, an anterior tongue-tie is attached from the alveolar ridge or floor of the mouth to between 0 and 5mm from the tip of tongue⁷. A mid tongue-tie is attached from the base of the alveolar ridge or floor of the mouth to between 6-10mm from the tip of the tongue⁷. A posterior tongue-tie, the least restrictive, has the same inferior attachment as previously mentioned, while being attached greater than > 15mm from the tip of the tongue⁷.

The treatment for ankyloglossia includes conservative management and frenotomy. Conservative management includes education, breastfeeding support and reassurance. Where conservative measures fail or are not appropriate infants are referred for frenotomy. Frenotomy is defined as division of the lingual frenulum by laser or scissors⁷. 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 anaesthetic exposing these infants to increased risks associated with the procedure. There is no international consensus regarding the correct management strategy for ankyloglossia and no widely accepted indications for surgical management. The paediatric societies of the United Kingdom¹⁰ and Canada recommend invasive intervention 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 for the procedure 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 a remarkable 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 4-fold, while the number of frenotomies has increased 5-fold¹³. This increase in the rate of frenotomies performed is stark, particularly when coupled with the debate surrounding the indications and efficacy of the procedure within the literature. The recent Clinical Consensus Statement released by the American Academy of Otolaryngology regarding ankyloglossia in children has recognised that recently there has been overdiagnosis and overtreatment of infants with ankyloglossia⁵. With this in mind, 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.

Methods

We performed a retrospective study analysing a consecutive cohort of infants referred to our ENT service with ankyloglossia. Local institutional 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: (1) infants presenting to our ENT service between 1st January 2019 and 31st December 2020, (2) ankyloglossia confirmed by ENT clinician, (3) infants younger than 18 months of age. Infants were excluded on the basis of additional congenital abnormalities, chromosomal abnormalities, comorbidities impacting ability to feed or swallow, age and incomplete data.

Diagnosis of ankyloglossia was confirmed by presence of a sublingual frenulum impacting the infant's tongue's appearance, function or mobility. Data was collected including: (1) infant age, (2) sex, (3) primary method of feeding, (4) prior history of ankyloglossia in a sibling, (5) referral method (General Practitioner, lactation consultant, Public Health Nurse, Paediatric consultant) and (6) if frenotomy was performed. Where documented in the healthcare record, (7) gestational age at birth, (8) birth weight and (9) location of ankyloglossia was recorded. We also recorded (10) the primary reason(s) for referral to the ENT clinic. These included maternal issues (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

Clinical Characteristics

There were 47 infants included in this study. Twenty-nine infants (61.7%) were males while 18 infants (38.3%) were female. Mean age at time of diagnosis was 8.3 weeks (SD \pm 6.6 weeks). There were 28 breastfed infants (59.6%) while 19 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). There was a family history of ankyloglossia identified for 11 infants (23.4%) while 36 infants had no family history of ankyloglossia. Frenotomy was performed for 30 infants (63.8%) while 17 infants (36.2%) were managed conservatively. (Table 1).

Table 1
Clinical and Presenting Characteristics

	Total Patients (%) (n = 47)
Sex	29 (61.7%)
Male	18 (38.3%)
Female	
Age at diagnosis (weeks)^a	8.3 (± 6.6)
Feeding method	28 (59.6%)
Breastfeeding	19 (40.4%)
Bottle feeding	
Birth weight (kg)^{a,b}	3.4 (± 0.61)
Gestational Age At Birth^b (weeks)	38.5 (± 2.1)
Sibling History Ankyloglossia	11 (23.4%)
Yes	36 (76.2%)
No	
Referral Method	12 (25.5%)
General Practitioner	5 (10.6%)
Lactation Consultant	26 (55.3%)
Public Health Nurse	4 (8.5%)
Paediatric Consultant	
Maternal Issue	24 (51.1%)
None	23 (48.9%)
Nipple pain/discomfort	

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

^b Incomplete data

	Total Patients (%) (n = 47)
Infant Issue	12 (25.5%)
None	30 (63.8%)
Poor Latch/Feeding issue	3 (6.4%)
Failure to thrive	2 (4.3%)
Poor Tongue Mobility	
Ankyloglossia Location	18 (38.3%)
Anterior	2 (4.3%)
Mid	14 (29.8%)
Posterior	13 (27.7%)
Not Recorded	
^a Mean value with Standard Deviation (SD) in parentheses	
^b Incomplete data	

Presenting Features

There were 18 anterior (38.3%), 2 mid (4.3%), 14 posterior (29.8%) cases of ankyloglossia while this data was not documented in 13 cases (27.7%). The majority of referrals (n = 26) to our clinic were made by the PHN (55.3%). General practitioners accounted for 12 referrals (25.5%), lactation consultants accounted for 5 referrals (10.6%) while paediatric consultants accounted for 4 referrals (8.5%). There was 35 referrals citing infant issues with 30 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, 2 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 23 cases (48.9%). (Table 1).

Factors associated with surgeon performing frenotomy

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 reported maternal issue related to feeding underwent frenotomy. With regards to infantile issues at presentation, 20 infants with failure to thrive underwent frenotomy while 10 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	18 (60%)	11 (64.7%)	0.750
Male	12 (40%)	6 (35.3%)	
Female			
Age at diagnosis (weeks)^a	7.5 (± 6.3)	9.7 (± 6.9)	0.537 ^c
Feeding method	20 (66.7%)	8 (47.1%)	0.188
Breastfeeding	10 (33.3%)	9 (52.9%)	
Bottle feeding			
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	9 (30%)	2 (11.7%)	0.156
Yes	21 (70%)	15 (88.3%)	
No			
Referral Method	8 (66.7%)	4 (23.5%)	0.239
General Practitioner	5 (16.7%)	0 (0%)	
Lactation Consultant	14 (46.7%)	12 (70.6%)	
Public Health Nurse	3 (10%)	1 (5.9%)	
Paediatric Consultant			
Maternal Issue	13 (43.3%)	11 (64.7%)	0.159
None	17 (56.7%)	6 (35.3%)	
Nipple pain/discomfort			

*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

	Frenotomy (%) (n = 30)	No Frenotomy (%) (n = 17)	P value*
Infant Issue	6 (20%)	6 (35.3%)	0.524
None	2 (6.7%)	1 (5.9%)	
Poor Latch/Feeding issue	20 (66.7%)	10 (58.9%)	
Failure to thrive	2 (6.7%)	0 (0%)	
Poor Tongue Mobility			
Ankyloglossia Location	18 (60%)	0 (0%)	< 0.05
Anterior	1 (3.3%)	1 (5.9%)	
Mid	2 (6.7%)	12 (70.6%)	
Posterior	9 (30%)	4 (23.5%)	
Not Recorded			
*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 ($p < 0.005$). This is concordant with current international recommendations⁵ Evidence does support that anterior tongue-tie can impact on feeding and that frenotomy may be of benefit in these infants^{5,14}. At present there is no evidence within the literature to suggest that posterior tongue-ties negatively impact on an infant's feeding⁵. Further to this 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 it does expose them to the potential risks of the procedure¹⁵. These risks include: infection, bleeding, scarring, salivary duct damage and nerve damage⁵. Previous studies have shown that ankyloglossia can be associated with poor feeding by reducing an infants' ability to latch properly for feeding^{16,17}. Thus, they are unable to adequately meet their nutritional requirements leading to failure to thrive. As such, it is important that these infants undergo timely treatment of their tongue-tie. Within our study we found a higher proportion of infants presenting with ankyloglossia associated with failure to thrive underwent frenotomy. Previous studies have demonstrated that frenotomy is of more benefit in those infants that are breastfed^{5,14}. In our analysis, a higher proportion of breastfed infants

referred to our clinic with ankyloglossia underwent frenotomy. We observed that infants who underwent frenotomy tended to be younger than those who did not. Again, this is consistent with findings from previous studies¹⁸. In agreement with current evidence we would advocate that breastfed infants with anterior tongue-tie and failure to thrive undergo frenotomy at an early stage. Overall, it appears from our analysis that clinicians within our institution were appropriately screening and identifying the infants with suspected ankyloglossia in whom frenotomy was appropriate based on these recommendations.

Over one-third of infants referred to our ENT outpatient clinic for consideration of frenotomy did not require or undergo the procedure. This finding is encouraging given the desire for clinicians to avoid unnecessary invasive procedures in all patients and particularly in infants. Studies have demonstrated that the majority of infants with ankyloglossia are asymptomatic^{19,20}. As such the majority of infants with ankyloglossia 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}. This appears to be largely due to two factors. Firstly, inappropriate screening and referral of infants with suspected ankyloglossia in the community will lead to many infants being seen in a clinic who do not need surgical treatment. This will lead to a low rate of frenotomy being performed in a given clinic as many infants who do not require any treatment are seen by the clinic. Appropriately screening which infants require referral to an ENT clinic for assessment and consideration of frenotomy is critical. This is because in February 2021, there were 67,980 patients waiting to be seen as outpatients at various ENT clinics within Ireland with 31,853 of these patients expected to be waiting more than 18 months²¹. Infants referred to our clinic with suspected ankyloglossia are seen and treated on an urgent basis in line with international best practice¹⁴. If ankyloglossia is impacting on an infant's ability to feed and develop it is vital that this be corrected as soon as possible. Referral of infants to an urgent ENT outpatient appointment 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 cancer who also necessitate urgent outpatient ENT appointments. The HSE has attempted to aide clinicians in screening infants with suspected ankyloglossia within the community through dissemination of a HSE ankyloglossia assessment proforma⁷. 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 mothers²⁴, 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 to perform or not to perform frenotomy in infants with ankyloglossia²². In the last number of years performing frenotomy has become a lucrative private practice. Many private clinicians are advertising and performing the procedure while charging parents significant fees. We note that under-6's primary healthcare is free within Ireland. This is at odds with clinicians charging parents hundreds of euros for consultations and performing of frenotomy in infants privately. An area of particular concern to some authors is these private clinicians offering to divide

“posterior” tongue-ties²³. As discussed, the evidence to support dividing these posterior tongue ties is questionable at best with most evidence suggesting posterior tongue ties do not impact feeding. It would not be routine practice in HSE or National Health Service (NHS) clinics to divide these posterior tongue ties due to the lack of evidence to support the practice²³.

The present study has a number of limitations. First is the small study numbers. This may have led to underestimation of the effect various factors had on clinicians decision to perform or not to perform frenotomy. Secondly, the only post procedure information collected was procedural complications. No follow up outcome measures were obtained such as increase in infants’ weight or reported improvement in feeding to assess the efficacy of frenotomy in various circumstances. Additionally this study was performed retrospectively which may have led to bias within the analysis. Finally, 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 that infants with anterior tongue-ties, failure to thrive and breastfed infants were more likely to undergo frenotomy. This is concordant with the current available evidence within the literature. Additionally, 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 within Ireland it is critical that infants with suspected ankyloglossia are properly assessed in the community using appropriate tools such as the HSE assessment proforma to avoid unnecessary referrals.

Declarations

Ethics approval and consent to participate

Local institutional ethical approval was obtained.

Consent for Publication:

Consent was obtained for publication of retrospective anonymised data.

Availability of Data and Materials:

Full datasets are available on request from the principal investigator, Áine Kelly

Competing Interests:

The authors have no competing interests to declare.

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Authors Contributions:

Aine Kelly: Principal investigator and lead author

Mel Corbett: Concept creation and editor

Eoin Cleere: Literary Review and editing

Aishan Patil, Matthew G Davey: Data collection interpretation and analysis

Bridgene McGlynn: Nurse in charge of clinic patient identification and ethics

Marcus Choo: Senior author and final review

References

1. Messner AH, Lalakea ML, Aby J, Macmahon J, Bair E. Ankyloglossia: incidence and associated feeding difficulties. *Arch Otolaryngol Head Neck Surg*. 2000 Jan;126(1):36-9
2. Dennis C, Jackson K, Watson J. Interventions for treating painful nipples among breastfeeding women. *Cochrane Database of Systematic Reviews*. 2014 Dec 15
3. Rowan-Legg A. Ankyloglossia and breastfeeding. *Paediatr Child Health*. 2015 May;20(4):209-18.
4. Segal LM, Stephenson R, Dawes M, Feldman P. Prevalence, diagnosis, and treatment of ankyloglossia: methodologic review. *Can Fam Physician*. 2007 Jun;53(6):1027-33.
5. Messner AH, Walsh J, Rosenfeld RM, Schwartz SR, Ishman SL, Baldassari C, et al. Clinical Consensus Statement: Ankyloglossia in Children. *Otolaryngol Head Neck Surg*. 2020 May;162(5):597-611.
6. Ingram J, Johnson D, Copeland M, Churchill C, Taylor H, Emond A. The development of a tongue assessment tool to assist with tongue-tie identification. *Arch Dis Child Fetal Neonatal Ed*. 2015 Jul;100(4):F344-8.
7. HSE, 2016. *Tongue Tie: Fact sheet for Health Care Professionals*. HSE, Appendix 2.
8. Martinelli R. Lingual Frenulum Protocol for Infants. *Clin Lactation*. 2017;8(3):135-8.
9. National Institute for Clinical Excellence, 2005. *Interventional procedures overview of division of ankyloglossia (tongue-tie) in babies with difficulty breastfeeding*. p.2.
10. World Health Organization, UNICEF. What Mothers Need to Know about Tongue-Tie
<<http://www.unicef.org.uk/BabyFriendly/Parents/Problems/Tongue-Tie/Division-of-tongue-tie/>>
11. Rowan-Legg A. Ankyloglossia and breastfeeding. *Paediatr Child Health*. 2015 May;20(4):209-18.
12. National Institute for Health and Care Excellence (NICE). Division of ankyloglossia (tongue-tie) for breastfeeding. London: NICE; 2005.
13. Walsh J, Links A, Boss E, Tunkel D. Ankyloglossia and lingual frenotomy: national trends in inpatient diagnosis and management in the United States, 1997-2012. *Otolaryngol Head Neck Surg*. 2017;156(4):735-740

14. Bundogji N, Zamora S, Brigger M, Jiang W. Modest benefit of frenotomy for infants with ankyloglossia and breastfeeding difficulties. *Int J Pediatr Otorhinolaryngol*. 2020 Jun;133:109985
15. Fraser L, Benzie S, Montgomery J. Posterior tongue tie and lip tie: a lucrative private industry where the evidence is uncertain. *BMJ*. 2020;;m3928.1
16. Ballard JL, Auer CE, Khoury JC. Ankyloglossia: Assessment, Incidence, and Effect of Frenuloplasty on the Breastfeeding Dyad. *PEDIATRICS*. 2002 Nov 1;110(5):e63
17. Forlenza GP, Paradise Black NM, McNamara EG, Sullivan SE. Ankyloglossia, Exclusive Breastfeeding, and Failure to Thrive. *PEDIATRICS*. 2010 Jun 1;125(6):e1500-e1504.
18. Diercks GR, Hersh CJ, Baars R, Sally S, Caloway C, Hartnick CJ. Factors associated with frenotomy after a multidisciplinary assessment of infants with breastfeeding difficulties. *International Journal of Pediatric Otorhinolaryngology*. 2020 Nov;138:110212
19. Hogan, M., Westcott, C. and Griffiths, M., 2005. Randomized, controlled trial of division of tongue-tie in infants with feeding problems. *Journal of Paediatrics and Child Health*, 41(5-6), pp.246-250
20. Messner A, Lalakea M, Aby J, Macmahon J, Bair E. Ankyloglossia. *Archives of Otolaryngology–Head & Neck Surgery*. 2000;126(1):36.
21. The National Treatment Purchase Fund, 2021. *Outpatient Waiting Lists by Specialty*.
22. LeTran V, Osterbauer B, Buen F, Yalamanchili R, Gomez G. Ankyloglossia: Last three-years of outpatient care at a tertiary referral center. *International Journal of Pediatric Otorhinolaryngology*. 2019;126:109599.
23. Fraser L, Benzie S, Montgomery J. Posterior tongue tie and lip tie: a lucrative private industry where the evidence is uncertain. *BMJ*. 2020;;m3928.1