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

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Research Article

Keywords: Ankyloglossia, Tongue Tie, Frenotomy, Release

DOI: https://doi.org/10.21203/rs.3.rs-820773/v1

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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 asymptomatic\textsuperscript{3}. The prevalence of ankyloglossia is estimated between 4–10\%\textsuperscript{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 children\textsuperscript{5}. 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 Tool\textsuperscript{6}. The Health Service Executive (HSE) have released a ‘tongue-tie’ assessment proforma\textsuperscript{7}, while also advocating for the use of Martinelli’s ‘Lingual Frenulum Protocol for Infants\textsuperscript{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 tongue\textsuperscript{7}. 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 tongue\textsuperscript{7}. 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 tongue\textsuperscript{7}.

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 scissors\textsuperscript{7}. In small infants this procedure is well tolerated in an outpatient setting without the need for general anaesthesia\textsuperscript{9}. 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 Kingdom\textsuperscript{10} and Canada recommend invasive intervention when there is a clear indication that a frenotomy would benefit the infant’s feeding\textsuperscript{11}. 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 procedure\textsuperscript{12}.

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-fold\textsuperscript{13}. 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 ankyloglossia\textsuperscript{5}. 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 were 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 ± 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 ± 0.61kg) while mean gestational age at time of birth was 38.5 weeks (SD ± 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**                        |                    |
| Male                           | 29 (61.7%)         |
| Female                         | 18 (38.3%)         |
| **Age at diagnosis (weeks)**   | 8.3 (± 6.6)        |
| **Feeding method**             |                    |
| Breastfeeding                  | 28 (59.6%)         |
| Bottle feeding                 | 19 (40.4%)         |
| **Birth weight (kg)**          | 3.4 (± 0.61)       |
| **Gestational Age At Birth**   | 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%)         |

*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**                  |                        |                           |          |
| Male                     | 18 (60%)               | 11 (64.7%)                | 0.750    |
| Female                   | 12 (40%)               | 6 (35.3%)                 |          |
| **Age at diagnosis (weeks)a** | 7.5 (± 6.3)          | 9.7 (± 6.9)               | 0.537c   |
| **Feeding method**       |                        |                           |          |
| Breastfeeding            | 20 (66.7%)             | 8 (47.1%)                 | 0.188    |
| Bottle feeding           | 10 (33.3%)             | 9 (52.9%)                 |          |
| **Birth weight (kg)a,b** | 3.4 (± 0.61)           | 3.3 (± 0.58)              | 0.579c   |
| **Gestational Age At Birthb (weeks)** | 38.8 (± 1.9)       | 38.1 (± 2.2)              | 0.704c   |
| **Sibling History Ankyloglossia** | 9 (30%)              | 2 (11.7%)                 | 0.156    |
| Yes                      | 21 (70%)               | 15 (88.3%)                |          |
| No                       |                        |                           |          |
| **Referral Method**      |                        |                           |          |
| General Practitioner     | 8 (66.7%)              | 4 (23.5%)                 | 0.239    |
| 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**       |                        |                           |          |
| None                     | 13 (43.3%)             | 11 (64.7%)                | 0.159    |
| Nipple pain/discomfort   | 17 (56.7%)             | 6 (35.3%)                 |          |

*Pearson’s Chi-Squared test unless otherwise specified

a Mean value with Standard Deviation (SD) in parentheses

b Incomplete data

Levene’s equality of variance test
| Infant Issue                  | Frenotomy (%) (n = 30) | No Frenotomy (%) (n = 17) | P value* |
|-------------------------------|------------------------|---------------------------|----------|
| None                          | 6 (20%)                | 6 (35.3%)                 | 0.524    |
| Poor Latch/Feeding issue      | 20 (66.7%)             | 10 (58.9%)                |          |
| Failure to thrive             | 2 (6.7%)               | 0 (0%)                    |          |
| Poor Tongue Mobility         |                        |                           |          |
| Ankyloglossia Location       |                        |                           |          |
| Anterior                     | 18 (60%)               | 0 (0%)                    | < 0.05   |
| Mid                          | 1 (3.3%)               | 1 (5.9%)                  |          |
| Posterior                    | 12 (70.6%)             | 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 recommendations. Evidence does support that anterior tongue-tie can impact on feeding and that frenotomy may be of benefit in these infants. At present there is no evidence within the literature to suggest that posterior tongue-ties negatively impact on an infant’s feeding. 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 procedure. These risks include: infection, bleeding, scarring, salivary duct damage and nerve damage. Previous studies have shown that ankyloglossia can be associated with poor feeding by reducing an infants’ ability to latch properly for feeding. 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. 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 studies\textsuperscript{18}. 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\textsuperscript{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\textsuperscript{14,18}. This appears to be largely due to two factors. Firstly, inappropriate screening and referral of infants with suspected ankyloglossia to a community clinic 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 months\textsuperscript{21}. Infants referred to our clinic with suspected ankyloglossia are seen and treated on an urgent basis in line with international best practice\textsuperscript{14}. 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 proforma\textsuperscript{7}. 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 mothers\textsuperscript{24}, 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 ankyloglossia\textsuperscript{22}. 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-ties. 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 practice.

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.

Funding:

No funding was obtained for this manuscript.
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

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