ABSTRACT

Ankyloglossia is thought to impact important early infant milestones associated with development such as latching and breastfeeding as well as potentially affecting palate formation and airway patency. Not only is this issue important for infants and neonates, who are in a vulnerable and malleable state where function dictates structure, but its effect on breastfeeding can be devastating for both mother and infant. The clinical question then is, ‘what are the best treatment options for infants with ankyloglossia?’ Although there are positive links between frenotomy procedures and outcomes, follow-up of frenotomy is vague, incomplete and inconclusive. Likewise, evidence is inconclusive for management options such as manual therapy to treat surrounding structures and areas of dysfunction in order to relax tethered oral tissues. Lack of conclusive evidence to determine a clear way forward in ankyloglossia treatment requires a deeper look and search for the best evidence available to assist parents and clinicians to make the best decision for their infant’s treatment. The goal of this clinical research-based viewpoint is to shed light on the treatment of frenotomy to help determine the best treatment at the right time. This treatment plan may be unique to each infant.

Key Words: Ankyloglossia, tongue-tie, lingual frenulum, manual therapy, diagnosis tool, management pathway.

Background

The diagnosis of ankyloglossia in infants is not a new concept and has been considered throughout history as early as the 18th century.1 The most common treatment is a frenotomy.2 It is difficult to ascertain the number of infants born with ankyloglossia, as prevalence ranges in literature from 0.1% to 12%.2-4 In the United Kingdom the number of infants undergoing frenotomy procedures was most recently collected in 2011. These figures suggested that 11.8 frenotomies were carried out per 1,000 children per annum.5 It is possible that over-diagnosis may lead to unnecessary surgeries.

This clinically focused article aims to evaluate research and potential consequences of the frenotomy procedures on neonates, as well as to provide an opinion in the consideration of other types of treatments for ankyloglossia.

Introduction

The lingual frenulum (LF) is considered an embryological tissue which, during the fetal period attaches to the underside of the tongue and the floor of the mouth.2 The initial function of this tissue is thought to maintain the lips and tongue in union with the cranial bones during fetal growth.2 In 2019 Mills et al6 published an infant cadaver study assessing the components of the LF. Their paper hypothesised that the function of the LF was likely to change throughout growth to an ultimate role of stabilization.6 Mills et al6 highlight key inconsistencies in anatomy with the current understanding of the topic claiming that the LF was composed of a fold of fascial tissue.6 This is an important discovery when considering the present understanding and management regarding ankyloglossia (AG) in pediatric patients. Ankyloglossia is currently defined as a congenital anomaly characterised by a short LF.7

An abnormally short frenulum (concurrent with certain theories) has been linked to restrictions in tongue mobility.8 Typically, AG has been associated with problems for both mother and infant. Studies investigating AG draw comparisons between a functionally restrictive frenulum and difficulties latching, breastfeeding (BF), teething, obstructive sleep apnea and even speech.2,7,9

There is no doubt of the importance of a mother’s breast milk when feeding the infant for the first six months and up to two years of life and beyond.10 Nutritional value gained from the mother not only provides the infant with necessary proteins, fats and sugars to support life, but also significantly contributes to the maturation of the gut.11 It is considered a vital time in the maturation of the infant where the healthy bacterium of the microbiome is cultivated. This contributes to both the infant’s ability to digest and to the emergence of their immune system, enabling the infant to combat disease effectively.12,13

Krol and Grossmann demonstrated the impact of BF on infant psychology.14 This could impact further issues such as mother and infant bonding, which can also affect aspects of emotional regulation, with the emotional deficit in some
infants that fail to attach to the breast even described as crippling.\(^2\) Recent studies, although unable to identify the exact mechanisms that link brain myelination and cognitive outcomes in BF individuals, highlight the importance of known nutrients including long-chain polyunsaturated fatty acids and other important myelin components to assist early neurodevelopment.\(^15\)

In 2021, the Academy of Breastfeeding Medicine produced a position statement on AG in breastfeeding dyads.\(^16\) This position statement considered that AG may significantly impede infant latching, suckling and breast milk transfer, while also impacting maternal nipple discomfort and trauma during BF.\(^16\) This results in an increased risk of discontinuing BF.\(^17\) An infant struggling to latch due to AG could suffer impaired development, especially at these key stages in early growth.\(^18,19\) As a result of this, it is important to ensure that the infant has minimal obstacles regarding BF. This evidence supports the hypothesis that AG is a significant problem that merits treatment to enable normal function.

**Prevalence**

Hazelbaker\(^3\) in 2017 suggested the prevalence of infants with AG can be up to 10%. However, other studies have reported varied prevalence ranging from 0.1% to 12%.\(^2-4\) Males have a greater prevalence than females (0.62).\(^20\) The figures for current corrective frenotomy procedures are illusive. In England the Department of Health produced an Infant and feeding profile from 2003-2011.\(^3\) In this literature the number of corrective frenotomy procedures in infants less than one years of age was around 11.8 per 1000 with a total of 8,088 procedures altogether in the year 2010 to 2011 (see tables 1 and 2).\(^3\) However, this is not strictly an accurate representation of the number of these procedures carried out today as this data only represented procedures carried out in Strategic Health Authorities. The figures, although the most recent, are also outdated, having been collected over ten years prior to the writing of this paper.

From 2007 to 2011 there was a significant rise of these procedures each year (see table 1).\(^5\) After plotting a forecast for future values, it suggests that if numbers for procedures continue to increase at the same rate, by 2022, the predicted number of frenotomies carried out in England would have been around 30,000 (see table 3). This signifies nearly 50 in 1000 babies having the procedure completed in these healthcare institutions (see table 4). Current diagnosis errors are thought to affect prevalence as there are numerous diagnostic tests which have been poorly validated.\(^21\)

### Diagnosis

Signs, symptoms, observation and a functional assessment can lead to a diagnosis of a functionally restrictive frenulum. The presence of one or more of the manifestations in table 5 should lead to the exploration of the infant’s mouth and assessment of the BF technique and nipples as well as a clinical breastfeeding assessment.\(^2\)

Current diagnosis is determined with a combination of both identification of a ‘short’ lingual frenulum and diminished oral function; this is because the limit of function is not always impacted by the length of the LF. Geddes et al,\(^7\) suggest this is because the function of infant sucking is affected by more components than just the tongue.

Assessment tools vary depending on location, institution and practitioner. The Hazelbaker assessment tool is

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**Table 1.** Department of Health Infant Feeding Profile. Trend in number of inpatient & outpatient tongue tie (Ankyloglossia) corrective surgeries in infants aged less than 1 year old per 1,000 live births at Strategic Health Authorities in England.\(^5\)
commonly known. This assesses five appearance and seven functional items of the tongue reviewing aesthetics and properties of the tongue, and how the tongue resides and is operational within the mouth of the infant. The five appearance items consist of appearance when lifted, elasticity, length when tongue is lifted, attachment to tongue and attachment to inferior alveolar ridge (see table 6).

The Coryllos Assessment is another common

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Table 2. Department of Health Infant Feeding Profile. Trend in number of inpatient & outpatient tongue tie (Ankyloglossia) corrective surgeries in infants aged less than 1 year old at Strategic Health Authorities in England.

Table 3. Forecast for Trend in total number of inpatient and outpatient tongue tie (ankyloglossia) corrective surgeries in infants aged less than 1 year old at SHA’s in England from the year beginning 2007.

[Graph showing trend from 2007/2008 to 2020/2021]
Table 4. Forecast for trend in total number of inpatients and outpatient tongue tie (Ankyloglossia) corrective surgeries in infants aged less than 1 year old per 1000 live births at SHA’s in England from the year beginning 2007.

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<th>Upper Confidence Bound</th>
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The Bristol Tongue Assessment Tool (BTAT) was developed aiming to produce an objective tool to provide consistent assessment of tongue appearance and function for infants with ankyloglossia.27 This tool scores infants from 0-2 in four different categories. The four categories are tongue tip appearance, attachment to the lower gum ridge, lift of the tongue and protrusion of the tongue. When the score is summed, totals of 0-3 are considered indicative of more severe reduction in tongue function.27 This tool was found to have good consistency (0.760) and showed significant correlation with Hazelbaker’s assessment tool (0.89) which indicates the simpler tool could be a more objective alternative.27 Other tools do exist, but these demonstrate the variety of assessments employed by health care providers and may explain some of the inconsistencies in diagnosis.

Current Treatment

As there are various treatments available in the approach to AG care, the choice can depend on the age of the infant or child at diagnosis, the practitioner responsible for intervention, preferences of the guardian and location of the patient.3 Patients may immediately have a LF division via frenotomy or may trial manual care to assess improvements. Lactation consult, or BF council may be approached to see if there is any advice applicable to mother and infant to improve feeding efficiency.38 In older patients beginning to communicate, speech and language therapists (SLT) can be used as part of a multidisciplinary team to improve and aid the function of the tongue.39

assessment tool. This appraises the frenulum physically, observing it situated in position with the tongue and the mouth floor.24 There are four types of classification including two types of anterior and two posterior types as seen in table 7 by Maya-Enero.25 Type 4 is unique in that it seems to be tight lingual fascia, and an indication that the baby should be examined for related musculoskeletal issues. Amir et al,23 reviewed the Hazelbaker assessment tool in 2006. In their study diagnostic items were condensed to improve diagnostic effectiveness.2,23 The three items considered included tongue lateralization, lift and extension, each also showed a high inter-observer agreement with a Kappa index of 0.65.23 Kotlow26 also produced a tongue tie assessment tool evaluating individuals under three diagnostic criteria.26 These include symptoms exhibited (by mother or infant) which can be associated to poor latch, appearance and function of tongue and maxillary lip, and assessment for lip tie.
• Latching difficulty.
• Painful and/or cracked nipple.
• Prolonged or very frequent feeds.
• Tongue cannot extend beyond the lips.
• Stunting. (Growth).
• Recurrent mastitis.
• Callus from breast feeding. (This is formed on the breast).
• Facial asymmetry.
• Twisted mouth when open.
• Tongue remains down during crying.
• Gothic palate. (Impaired or high palate).
• Cheek hypertrophy.

Table 5. Manifestations of AG.2

<table>
<thead>
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<th>Appearance items</th>
<th>Functional Items</th>
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<td>Appearance of tongue when lifted</td>
<td>Lateralization</td>
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<tr>
<td>Elasticity of frenulum</td>
<td>Tongue Lift</td>
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<tr>
<td>Length of LF when tongue is lifted</td>
<td>Tongue Extension</td>
</tr>
<tr>
<td>Attachment of LF to tongue</td>
<td>Spread of Anterior Tongue</td>
</tr>
<tr>
<td>Attachment of LF to inferior alveolar ridge</td>
<td>Cupping</td>
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<td>Peristalsis</td>
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<td>Snapback</td>
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Table 6. Items of Hazelbaker Assessment Tool.21

Table 7. Coryllos Classification Types (Maya-Enero, López-Vilchez, Luis Ruiz; 2021)25 Photos by Dr. Luis Ruiz-Guzman; with permission from Dr. Silvia Maya-Enero.

Manual therapists such as chiropractors and osteopaths deploy tactile skills to assess areas of tension surrounding the jaw, cranium, neck and floor of the mouth, before soft mobilization techniques are used to decrease tissue tension.30

The complication rates of these interventions are hard to predict because of the lack of data recorded on the affects they have on patients with AG complaints. Despite this, Dixon et al.31 discusses the benefits of a multifaceted approach to AG prior to frenotomy.31 They suggest patients should receive a thorough AG diagnosis using the BTAT and lactation consult.31 Their findings showed a significant improvement in breastfeeding rates with no difference from those who received a frenotomy.31 A recent AG consensus carried out in Australia proposed a similar package of care in the form of a management pathway for those with AG diagnoses and feeding issues.28 This suggested a non-surgical management and a reassessment of feeding before approaching surgical consult.28 Following a management pathway will allow infants with feeding difficulties to trial conservative care and management under a range of medical professionals prior to seeking operative attention. Research evidence, over-all, is lacking for treatment options for AG.

Currently, surgical treatment is recommended for AG if it is associated with BF problems that are not resolving conservatively.2 Some researchers believe, depending on the degree of restriction, that operative intervention can affect subjects’ speech, oral function and oral posture.32 However, evidence behind current surgical division is inconsistent. A systematic review by Visconti et al.33 highlighted that although improvements are made in many surgical trials, the diagnosis and assessment vary, and progress is unpredictable.33 Ghaheri et al34 produced a randomized controlled trial on frenotomy for infants with diagnosed posterior tongue-ties (PTT).34 They claimed that division both subjectively and objectively improved infant feeding. Ghaheri et al34 found the ‘within-burst’ suck period of infants improved in surgical groups to -0.14 from 0.9, compared to 0.27 from 1.0 found in control groups.34 This suggested that in a burst of sucks, the infant can produce faster sucks if the tongue is divided.34 Using the Breastfeeding Self-Efficacy Scale Short Form (BSES-SF) they subjectively determined BF confidence scores improved from 37.5 to 50.9.34 However, the study itself had a small sample size and there was no follow up beyond 10 days.34

Surgery involves the division of the LF; this is a fast procedure which can be completed in the office of a trained professional, establishing it as the most common technique performed on infants.127 These professionals include dentists (DDS, DMD), oral surgeons and other medical physicians (MD’s with specialties like (but not
limited to) otolaryngology, pediatric surgery, general pediatrics), physician assistants (PA) and advanced practice registered nurses (APRNs), midwives and properly trained international board of certified lactation consultants (IBCLC). Depending on the regulations in their country, state or province, IBCLCs would be required to have additional credentials that qualify them to perform this procedure under their scope of practice.

The procedure is sometimes accompanied by anaesthesia (24%) in the form of sucrose both pre and post-surgery. Further, a 2016 study conducted by Shavit et al investigated the topical administration of some anaesthetizing agents used within frenotomy procedures, finding them ineffective (benzocaine and tetracaine). Shavit et al suggested that the search should continue for effective pain management for infants during frenotomy procedures. After surgical intervention, the infant should be placed in skin-to-skin contact with the mother immediately to initiate feeding and assist with pain relief. Sucrose could be reapplied if necessary.

When performing the frenotomy, the infant may be wrapped securely in a blanket or in a structured papoose consisting of a board, head rest and Velcro fastened wraps to effectively limit movement. To keep the mouth open, an assistant gently holds down the chin before the clinician lifts the tongue with their fingers or an appropriate tool. A small incision is then made approximately 2mm in the central mucous membrane using surgical scissors or a laser. Special care is taken to avoid damage to the base of the tongue due to its vascularization and the position of the Wharton duct. Kenny-Scherber and Newman discuss applying pressure with the index finger on the incision area, after the initial cut, to broaden any division made.

Frenectomy and frenuloplasty are different procedures that often get interpreted as frenotomy. Frenectomy involves the complete removal of the frenulum either using a scalpel or laser. Whereas a frenuloplasty is a restructuring of the frenulum using plastic surgery to minimise any scar tissue development. These techniques are beyond the scope of this paper.

Complications

Current rates of complications with frenotomies are relatively low ranging from 1-9% across different studies. Despite this, there are many different complications which have been observed from this procedure. Previously frenotomies have been described as both pain and risk free. However, there is now an understanding that this is not the case, as infants can experience prolonged pain and the possible onset of other symptoms. Pain is not a complication that should be overlooked in this vulnerable period of infant development. Victoria and Murphy discussed that an exposure to pain in early life can result in hyper- or hyposexual phenotype in response to short term or lasting pain and stress in later life.

Walsh and Tunkel reported a series of different complications and their prevalence including bleeding (3-5%), recurrence (5%), lip or Whartons duct injury, infection, injury to the lingual nerve, formation of mucous retention cyst, pain and failure of technique to improve BF. Rates of oral or breast aversion are also significant enough to be documented in studies as infants negatively associate objects in their mouth with pain or stress, disrupting a crucial relationship between a mother and their infant. Hale et al emphasised other complications often missed in other papers reporting incidences after division. They noticed infants were having delays in the diagnosis of underlying conditions, which had been overlooked in favour of treating AG. One case involving a cardiac disorder, another severe dehydration and weight loss and one case involved a failure to adequately establish feeding. The same paper also mentioned the complication of financial burden which often gets overlooked as there can be a high cost for the procedure.

Other case reports have highlighted additional circumstances where further complications have arisen such as Ludwig Angina, two cases of severe hypovolaemic shock and two cases of frenotomies leading to airway obstructions in patients with Pierre Robin Sequence.

Financial burdens and complications to infant not only interfere with BF but also affect a parent’s ability to consent to continued BF. They could feel additional pressure to continue BF due to the possible trauma placed on the infant or the financial sacrifice made to carry out the procedure.

It is not uncommon for invasive procedures to have secondary impacts and complications. There are risks of complication in all surgical interventions such as wound dehiscence, infection, excess bleeding, nerve injury or scar and tissue adhesions, which could affect future function of the body. It is important to address other types of therapies as surgical procedures are not without serious side effects. Therefore, should surgical procedures be used in any but the most egregious cases? There may be other considerations with AG that should be considered first.

A 2019 study by Mills et al declared the lingual frenulum is less a band of elastic tissue waiting to be snapped back into place, but instead a fibrous fold in a layer of fascia. Fascia itself is known to have various anatomical functions such as stabilization, imparting of strength, maintaining vessel patency, separation of muscles and enclosure of different organs. However, the function of LF is to stabilize the tongue under tension against resulting diverse vectors
of forces. This static fascial role would suggest that the LF cannot be tight at all. It is more a fascial captive of the surrounding tissue, only to be considered taut if the attaching muscles or structures are dysfunctional. This emphasizes the concern of whether the LF should be cut at all.

It is considered in some literature, that the lingual frenulum plays part in a chain of fascia and muscle from the tongue and floor of the mouth, down the anterior neck and abdomen, via the genioglossus, then geniohyoid and then mylohyoid muscles before inserting onto the hyoid bone in the neck.53-55 Myers55 explored this theory further when discussing the next linked tissues. They suggested that stresses on the hyoid bones could be conceived in the anterior chest, then in the abdomen, before continuing down these myofascial and kinematic chains until reaching the ankles and feet.55 Due to the relationship between these muscles and tissues, it is reasonable to assume that there is a chain reaction through the body.55

Compensatory muscular tension is a regular complication of AG. If the muscle surrounding the jaw and oral cavity becomes tight due to a restriction in tongue movement, associated muscles will also be affected.37 These longitudinal muscular continuums still function; however, adaptations can interfere with central pattern generators located within the brain stem and spinal cord.37,56 These important areas, responsible for the production of movements and roles governing an infant’s ability to suck swallow and breathe, may now perform suboptimally.37,56 The immediate release of these regionally affected structures could induce a fast increase to the motion in an ill prepared infant leaving them orthopedically susceptible.57,56 A 2012 investigation by Borstad and Szucs57 in adults, demonstrated this hypothesis in breast cancer suffers, revealing that patients who had breast surgery were consequently vulnerable to further orthopedic problems as kinematics in the shoulder had been altered.57 These patients also scored significantly higher on pain scores and had altered recreation and active function.57

If restriction at the LF can affect up to the occiput and down to the abdomen and lower limbs, it is feasible that these areas can equally affect the LF causing tension in other areas of the kinematic chain.55 Therefore, consequent tension caused by birthing trauma can follow these anatomical cascades increasing tension at the LF putting the infant at a greater risk of an AG diagnosis.36,58 Gottlieb59 suggested that an increase in minor birthing trauma correlates with an increase in the levels of intervention in the current birth process.59 These interventions have musculoskeletal consequences on the infant.58,59 Therefore, it is not irrational to hypothesize that frenotomy populations accordingly could decrease if tension from traumatic events were treated effectively.

When considering contributing factors to a blanket diagnosis of AG, the palate is a vital part of the feeding mechanism that should not be dismissed. When feeding, the tongue works in conjunction with the palate of the mouth to create a negative pressure to initiate a milk ejection reflex in the breast.37,60 If the palate is too high, from a congenital abnormality or dysfunctional muscles such as the palatoglossus, when the infant draws the breast into the mouth, they will not be able to feed effectively.57 This ineffective ability to suckle will likely, in individuals with a taut LF, be diagnosed as AG. However this judgement is ambiguous.56 When treating the palate of an infant, Williams58 suggested that the skull is a useful tool for assessment and therapy.58 Williams looks at the maxilla, which can be affected by birthing trauma, to be a contributor to hard palate deformation.58 This can be treated with soft internal manipulation using a touch and hold technique as advocated by Williams.58 However, the maxilla should not be a practitioner’s only therapeutic focus. Instead, look to the occiput to influence any tightness or imbalance within the sphenoid prior to attempting to treat the maxilla.61

Davis discussed how the kinesiopathological pattern of an infant can be optimised by restoring typical function of the sphenobasilar mechanism.61 Although there is insufficient research to support manual therapy alone in the treatment of AG, it is logical that considerations need to be made for all of the vital surrounding tissues in the newborn.

The relationship of the hypoglossal nerve should also be discussed as a factor in relation to AG. This nerve supplies the tongue, travelling through the hypoglossal foramen at the occipital condyle to innervate the tongue in the mouth. In neonates the occipital condyle is cartilaginous and in two separate parts. Carriero62 discussed how unwanted distortion of these components may compromise the lumen the nerve travels through.62 This could lead to reduction in sucking activity from irritation to the hypoglossal nerve at the level of the occipital canal.62 Furthermore, an obstruction from biomechanical compromise, in the sensorimotor integration mediating part of the central nervous system (which informs the musculoskeletal system of environmental demands) can present as dysfunction in an infant such as ineffective feeding.46 This draws interest to the base of the skull when thinking about treating AG. Anatomical science demonstrates how the oropharyngeal system is effectively tethered to the base of the cranium. The muscles of the tongue attach to the mandible, the temporal bone and the hyoid, while the hyoid attaches to the temporal bones via the stylohyoid ligaments and muscles and digastric.58 Therefore, it is plausible to assume that if the range and combined movements of these associated tissues and bones are dysfunctional, the suckling and feeding of the neonate will be affected. Treating these areas of dysfunction may improve the struggling infant’s ability to feed efficiently.
All procedures used to treat ankyloglossia in infants need to be properly researched to determine the best and safest evidence-based approach to improve the infant’s comfort and function. Chiropractic care has been found safe using manual therapies in infant care. At a minimum, all clinicians who treat these infants should keep accurate and complete records and share them across professions to help determine clinically useful treatment options.

Conclusion
Ankyloglossia is a common diagnosis in breastfeeding difficulties. Frenotomy is a quick procedure that in some cases facilitates the latching of the infant at the breast almost immediately. However, it is not without risks and lacks high-quality research evidence. There are many variables that need to be revisited before a practitioner can assess infants for ankyloglossia with absolute clarity. The perfect clinical scenario calls for a universal assessment tool and diagnosis pathway to be utilized by all clinicians, allowing practitioners to approach feeding difficulties conservatively prior to the division of the lingual frenulum. This will ensure all care is individualized and potentially harmful surgical intervention is avoided where possible.

References:


Treatment options for ankyloglossia: A research informed review


