

Tongue-tie – development, evolution and treatment

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ABSTRACT

Over the past decade, there has been an exponential increase in the number of children diagnosed and treated with ankyloglossia, a condition where the lingual frenulum attaches near the tip of the tongue (anterior tongue-tie) or at the root of the tongue (posterior tongue-tie) and may be short, tight and thick. There have been immense controversies regarding clinical significance of this condition. Recent studies suggest that speech, solid feeding, and sleep difficulties may be linked to restricted tongue function. This article presents the latest evidence on the diagnosis and management of tongue-tie.

Keywords: tongue-tie, tongue-tie division, ankyloglossia, frenulum, frenectomy, breastfeeding, speech disorders, temporomandibular joint, sleep apnea, ADHD, infant feeding, breastfeeding consultant

INTRODUCTION

The tongue is a highly mobile organ made up of longitudinal, horizontal, vertical, and transverse intrinsic muscle bundles. The extrinsic muscles are the fan-like genioglossus which is inserted into the medial part of the tongue and the styloglossus and hyoglossus into the lateral portions [1].

The lingual frenulum is a dynamic structure, formed by a midline fold in a layer of fascia that inserts around the inner arc of the mandible, forming a diaphragm-like structure across the floor of mouth. This fascia is located immediately beneath the oral mucosa, fusing centrally with the connective tissue on the tongue's ventral surface. The sublingual glands and submandibular ducts are enveloped by the fascial layer and anterior genioglossus fibers are suspended beneath it. Lingual nerve branches are located superficially on the ventral surface of the tongue, immediately deep to the fascia. The lingual frenulum is not a discrete midline

structure. It is formed by dynamic elevation of a midline fold in the floor of mouth fascia [2].

Tongue-tie, or ankyloglossia, is a condition in which the lingual frenulum may either be attached too close to the tip of the tongue [3] and too far forward towards the inferior alveolar ridge, or it may be attached in a more posterior position on the tongue and the floor of the mouth, but be so short as to impede movement. When the tongue is lifted, the tip of the tongue may form a heart shape [4].

An expert panel of pediatric otolaryngologists was assembled with nominated representatives of otolaryngology organizations in 2020 [5]. The panel achieved clear consensus in describing ankyloglossia as a "condition of limited tongue mobility caused by a restrictive lingual frenulum".

No specific embryological cause of ankyloglossia has been identified so far [6]. In large cross-sectional studies of the condition in newborns, the prevalence has ranged from 4% to 10% [7]. Boys are affected more than girls, with the sex ratio being

about 2:1. There is no clear ethnic predilection [8].

In neonates referred because of breastfeeding problems, the prevalence of tongue-tie varies across studies from 12.8 % to 56 % [7,9,10]. The large increase in prevalence may be due to increased focus on the condition, previous under-reporting or a tendency to overdiagnose in recent years.

Ankyloglossia was also found associated in cases with some rare syndromes such as X-linked cleft palate syndrome [11], Kindler syndrome [12], van der Woude syndrome [13] and Opitz syndrome [14]. Nevertheless, most ankyloglossia are observed in persons without any other congenital anomalies or diseases.

CLASSIFICATION OF TONGUE-TIE

The term free-tongue is defined as the length of tongue from the insertion of the lingual frenulum into the base of the tongue to the tip of the tongue. Clinically acceptable, normal range of free tongue is greater than 16 mm. The ankyloglossia can be classified into 4 classes based on Kotlow's assessment, as follows: class I – mild ankyloglossia, 12 to 16 mm, class II – moderate ankyloglossia, 8 to 11 mm, class III – severe ankyloglossia, 3 to 7 mm, class IV – complete ankyloglossia, less than 3 mm. Class III and IV tongue-tie category should be given special consideration because they severely restrict the tongue's movement [15]. A normal range of motion of the tongue is indicated by the following criteria: The tip of the tongue should be able to protrude outside the mouth; without clefting, the tip of the tongue should be able to sweep the upper and lower lips easily; without straining, when the tongue is retracted, it should not blanch the tissues lingual to the anterior teeth; and the lingual frenulum should not create a diastema between the mandibular central incisors.

The appearance of the tongue is not sufficient on its own to make a diagnosis, as the thickness and elasticity of the frenulum also vary widely and affect the extent to which normal tongue movements are inhibited.

Anterior ankyloglossia is much more common and readily managed when compared to posterior ankyloglossia. Posterior ankyloglossia is a poorly recognized condition that may contribute to breastfeeding difficulties [16]. The diagnosis is difficult due to the subtle clinical findings but relevant health care providers should be aware of this condition.

The concept of 'posterior lingual frenulum' is controversial. The term can also be misleading because the reasons for reduced tongue mobility may be complex, and not necessarily related to the lingual frenulum itself [17]. According to the guidelines of the Norwegian Society of Pediatricians,

'posterior lingual frenulum' simply means a somewhat thicker submucosa in the posterior part of the lingual frenulum; the guidelines emphasize that there is only a single lingual frenulum. The diagnosis of "posterior" tongue-tie also applies a reductionist, medicalized theoretical frame to the complex problem of impaired tongue function, risking unintended outcomes. Impaired tongue function arises out of multiple interacting and co-evolving factors, including the interplay between social behaviors concerning breastfeeding and mother-infant biology [18].

The extensive variation in the reported prevalence of ankyloglossia reflects the differing diagnostic criteria that are used. There is no consensus on either definition or grading system.

Martinelli designed in 2010 a new lingual frenulum protocol with scores for infants and is considered to be an effective tool for health professionals to use for assessing and diagnosing the anatomical alterations of the lingual frenulum, and its possible interference with breastfeeding [19].

The most comprehensive clinical assessment used today is the Hazelbaker Assessment Tool for lingual frenulum function (HALF) [20].

The ATLFF was designed to be used on all babies, regardless of feeding method.

A reliable screening tool needs to be developed and validated. Education for nurses to assess infants with ankyloglossia in a systematic fashion should be explored [21]. Investigation of psychological sequelae, including maternal stress, postpartum depression, and mother-infant bonding is an important next step in the research of infants with ankyloglossia.

TONGUE-TIE IMPACT IN BREASTFEEDING

The significance of ankyloglossia in children remains controversial, both within, and between, specialty groups [22]. Tongue-tie is linked to breastfeeding difficulties, gagging, choking or vomiting food, delayed development or deterioration of speech, dental and behavioral issues [23]. Ankyloglossia is a relatively common finding in the newborn population and represents a significant proportion of breastfeeding problems. Poor infant latch and maternal nipple pain are frequently associated with this finding. Careful assessment of the lingual function, followed by frenulum division when indicated, seems to be a successful approach to the facilitation of breastfeeding [1,24].

The movements of the tongue during infant feeding have been studied by ultrasound [25,26]. Ultrasound reveals some similarities between the movements made by the baby when either breast or bottle feeding [27], but also some important differ-

ences [28]. The tongue is projected further forward in breast feeding [29] and the human nipple elongates during each suck in a way that an artificial teat cannot do. During feeding, the artificial teat, or the nipple together with some breast tissue, is held fully in the mouth with the tongue covering the lower gum ridge. The nipple is protected from damage and pain at the back of the baby's mouth. The baby's lower jaw is then elevated, compressing the artificial teat, or the breast immediately behind the nipple, while the front of the tongue moves up to aid the expression of milk. In breast feeding, this is by compression of the milk ducts under the areola. A wave of upward movement of the medial part of the tongue progresses backwards, and the expression of the milk is further facilitated by negative pressure generated by downward movement of the back of the tongue and the lower jaw and, in breast feeding, by the active expulsion of milk once the let down occurs.

In coordinated feeding, the sucking, swallowing and breathing movements follow in a 1:1:1 sequence. This can take several days to become established in healthy full term infants. In pre-term infants and in some term infants a variety of poorly coordinated feeding movement patterns are observed and sometimes persist [30]. Swallows did not occur on their own. Younger babies' swallowing invariably was associated with a pause in breathing. Older babies generally showed better co-ordination of sucking, swallowing and breathing than younger ones, whether breast- or bottle-feeding [26]. Antenatal ultrasound studies [31] show that mouth and tongue movements are already well developed in association with intra-uterine yawning and crying.

The impact of posterior tongue-ties on the pharyngeal phase of swallowing is not well documented in the literature. A video-fluoroscopic swallow study (VFSS) allows for visualization of the oral, pharyngeal, and esophageal phases of the swallow. When decreased base of tongue movement, impaired pharyngeal pressure generation, and presence of pharyngeal residue are noted during a VFSS, a neurologic etiology can be suspected. However, in the setting of a normal MRI with normal motor development, other etiologies need to be explored [32]. Patients with impaired base of tongue movement and impaired pressure generation resulting in pharyngeal residue in the setting of a normal neurologic workup could possibly present with a posterior tongue-tie which should be examined and included in the differential diagnosis.

Tongue-tie has been cited as a cause of poor breastfeeding and maternal nipple pain [3]. Good assessment and selection are important because 50% of breastfeeding babies with ankyloglossia will not encounter any problems [20].

Several randomized clinical trials report a significant reduction in maternal nipple pain after frenotomy. Frenotomy is a low-procedure surgery, but the benefits are sparsely documented. Parents should be presented with risk and benefits, before a division is offered [33].

A study conducted by Herzl-Goldfarb Breast-feeding Clinic between April 2014 and April 2015, published in 2019 [34] concluded that frenotomy for posterior ankyloglossia may improve breastfeeding and nipple pain. Subjective improvement in breastfeeding was reported by 90% of mothers immediately after frenotomy and 83% of mothers later.

Waterman's [35] article about mothers' experiences of breastfeeding a child with tongue-tie revealed that most participants described an overwhelming desperation to figure out what was going on with their child. Many reported feeling guilt for what their child had endured, blaming themselves for not being able to 'breastfeed properly', and for feeling dread each time their child latched onto the breast. Many of the participants felt let down by their breastfeeding experience and had a distinct sense of disappointment. Some participants, especially first-time mothers, spoke to the fact that they did not anticipate breastfeeding to be so difficult. A Cochrane review update from 2020 concluded that frenotomy reduced breastfeeding mothers' nipple pain in the short term [36].

A prospective observational study published in 2019 evaluated what determined breastfeeding problems in a non-selected mother-infant cohort, with special reference to tongue-tie and improvements in breastfeeding following frenotomy. Tongue-tie had a significant impact on breastfeeding and so did low birth weights and prematurity. Frenotomy proved helpful when breastfeeding problems were reported [37].

A prospective, cohort study from June 2014 to April 2015 in a private clinic concluded that surgical release of tongue-tie/lip-tie results in significant improvement in breastfeeding outcomes. Improvements occur early (1 week postoperatively) and continue to improve through 1 month postoperatively. Improvements were demonstrated in both infants with classic anterior tongue-tie and less obvious posterior tongue-tie. This study identifies a previously under-recognized patient population that may benefit from surgical intervention if abnormal breastfeeding symptoms exist [38].

Ballard and colleagues [39] reported a marked fall in maternal pain scores after the procedure. Hogan and colleagues [40] randomized their cases to immediate or delayed intervention and found that frenotomy was much more effective than advice from a lactation counsellor. They reported dramatic

and rapid, often immediate, improvement after the procedure in most of their cases; improvement was noted in 95% of babies.

A 24-month prospective New Zealand Pediatric Surveillance Unit study from 2020 concluded that poor feeding, pain, bleeding, weight loss and delayed diagnosis of an alternative underlying medical condition are important complications that require hospital assessment and admission [41]. Practitioners and parents/families need to be aware of these possibilities. Centralized guidelines with access to specialist second opinions should be developed.

TONGUE-TIE IMPACT IN SPEECH AND ORAL DEVELOPMENT

One of the first symptoms of tongue-tie in adults is poor oral health. When the tongue has limited mobility, it becomes difficult to remove food and debris from the teeth after eating. Tongue-tie can also cause a gap between the bottom front teeth. Adults with tongue-tie may experience frequent cavities, gum inflammation, gum disease, bad breath, and other oral health problems.

Another major symptom of tongue-tie in adults is temporomandibular joint(TMJ) dysfunction. When the tongue is unable to move in a full range of motion, the mouth naturally adapts to cope. One of the ways the mouth copes is by swallowing incorrectly. Normally, the tongue will push food around in the mouth to the back to swallow but when a tongue-tie is present, this cannot happen. Instead, people with tongue-tie have food move around in their mouth when they eat. This can lead to incorrect swallowing. With consistent incorrect swallowing, teeth can come misaligned and the jaw will develop incorrectly. The misaligned teeth and jaw development issues can cause an imbalance in the jaw joints which results in TMJ disorders. TMJ disorders can lead to serious symptoms like frequent headaches and migraines, jaw, neck, and back pain, clicking and popping jaw, and much more [42].

Localization of the frenulum insertion on the gingiva seemed to be of importance for gingival sequelae because insertion of the lingual frenulum in the area of the papilla had the highest association with gingival recession [15].

An abnormally low position of the tongue may cause mandibular prognathism with maxillary hypodevelopment due to an exaggerated anterior thrust leading to Class III malocclusion [22]. Whereas, somewhat higher position of tongue in the mouth may lead to tongue thrust causing posterior or anterior open bite. Moreover, excessive forces while retrusion of tongue by patient may cause blanching of tissues, gingival recession, and midline diastema in lower central incisors.

Speech problems can occur when there is limited mobility of the tongue due to ankyloglossia. The difficulties in articulation are evident for consonants and sounds like “s, z, t, d, l, j, zh, ch, th, dg” [43] and it is especially difficult to roll an “r”. These articulation problems are, however, less common than tongue-tie itself, and children and adults characteristically use various compensatory techniques of mouth opening and tongue movements [44].

Tongue-tie restricts the physiologic movements of the tongue and results in various functional, behavioral and speech abnormalities along with the development of frontal and lateral lips. Tongue-tie may cause problems which may exist since birth such as breastfeeding and swallowing to problems which may persist through lifetime such as dysarthria, mechanical problems, and social issues [45].

TONGUE-TIE IMPACT IN SLEEP BEHAVIOR AND MORE

Sleep is often impacted, beginning in infancy. If tongue-ties remain untreated, they can lead to structural and functional changes in the craniofacial-respiratory complex and can impact sleep throughout the lifespan [46]. Tongue-ties and low tongue resting postures often lead to or exacerbate mouth breathing. Mouth breathing prevents the brain from experiencing the deepest level of sleep. As a result, people who mouth breathe at night often awaken un-refreshed. Children and adults may be getting the right quantity of sleep at night, but many are not getting the quality of sleep that they need.

Snoring is a red flag and signals that obstructive sleep apnea or sleep-disordered breathing is likely. Tongue-tie can cause sleep apnea as a result of an underdeveloped palate which can cause a smaller airway. When someone with a smaller palate goes to sleep, they're at a higher risk of their upper airway collapsing – which is the cause of sleep apnea [42]. Sleep apnea can cause daytime sleepiness, morning headaches, difficulty concentrating, memory problems, moodiness, and result in car accidents. Without treatment, it can also increase the risk of heart disease, stroke, diabetes, high blood pressure, and more.

Upper airway resistance and poor nasal breathing can be caused by a smaller-than-normal nasal cavity, deviated septum, or high arched palate. All of those may develop when there is insufficient pressure from the tongue against the palate both in utero and during infancy [47]. These common tethers keep the tongue in a low position in utero, so the palate does not receive its natural resting pressure from the tongue, and instead of a broad, flat palate, the baby is born with a high arched or “bubble” palate which leaves less room for the base of the nose

and less volume available for the nasal cavity. It is said that nursing is nature's palatal expander—meaning that the action of breastfeeding works to broaden the palate and increase the size of the nasal cavity. Many airway issues that continue into childhood and even adulthood can be traced back to factors in infancy which promote the high palate; for example, low tongue posture (often from a tie), bottle-feeding, and a defective swallow. A tongue-tie release in infancy along with breastfeeding can help prevent adult airway constrictions by allowing the infant to develop a broad, flat palate and spacious nasal passages and sinuses [46].

Narrow palates and soft palate elongations have been associated with tongue-ties in a recent study [48]. After full tongue-tie releases, children and adults are often found to sleep more deeply, snore less, exhibit fewer movements, and feel more refreshed in the morning.

Often the parents and patients themselves report better concentration and less hyperactivity as well [46]. This suggests that tongue-tie might impact an ADHD-like behavior, as a consequence of sleep-disorder breathing. More research is needed and this might have a huge impact in approaching the ADHD therapy, especially in young children, where tongue-tie release might be considered as part of the therapy.

Mechanical (nonspeech-related) symptoms and social concerns due to impaired tongue mobility may not become apparent until late childhood [49].

TONGUE-TIE TREATMENT AND MANAGEMENT

Considerable controversy regarding the diagnosis, clinical significance, and management of the condition remains, and great variations in practice have been recorded [50]. Although most tongue-tie babies are asymptomatic without feeding difficulties, operative corrections may be necessary in some cases to improve their breastfeeding [51].

Operative interventions were proposed already in Greek medicine. In the Middle Ages, competition arose between midwives, who used their nails to detach the frenulum, and surgeons, who were allowed to use instruments. During the last century, the justification for frenotomy shifted from improved language to enhanced feeding. Despite the high frequency of the intervention, little research has been performed on its benefit, which remains a controversial topic among different groups of health professionals [52].

Anterior tongue-tie is accepted in most clinical practices as a potential risk for breastfeeding difficulty, and good evidence exists that division of an anterior tongue-tie leads to improved breastfeeding outcomes [53]. Posterior tongue-ties and upper lip-

ties are being studied more intensively to provide sound, evidence-based recommendations on their diagnosis and treatment.

Tongue-tie division is a safe procedure with minimal complications. Surgical techniques for the therapy of tongue-ties can be classified into three procedures. Frenotomy is a simple cutting of the frenulum. Frenectomy is defined as complete excision, i.e., removal of the whole frenulum. Frenuloplasty involves various methods to release the tongue-tie and correct the anatomic situation. There is no sufficient evidence in the literature concerning surgical treatment options for ankyloglossia to favor any one of the three main techniques [15]. The commonest complication is minor bleeding. Recurrence leading to re-division occurs with rates of 0.003-13% reported; this appears to be more common with posterior than anterior ties. Surgical release of the lingual frenulum has become an increasingly common procedure, performed from birth through to adulthood. There are limited reports indicating that prophylactic frenotomy may promote subsequent speech development; however, evidence is currently insufficient to condone this practice and further good quality research into this area is warranted [54].

Infant tongue-tie can cause breastfeeding problems, which may be improved by frenotomy, by correcting the restriction of the tongue movement and allowing more effective breastfeeding with less maternal nipple pain [3]. Prospective, cohort study from June 2014 to April 2015 [38] concluded that surgical release of tongue-tie/lip-tie results in significant improvement in breastfeeding outcomes. Improvements occur early (1 week postoperatively) and continue to improve through 1 month postoperatively. Improvements were demonstrated in both infants with classic anterior tongue-tie and less obvious posterior tongue-tie. This study identifies a previously under-recognized patient population that may benefit from surgical intervention if abnormal breastfeeding symptoms exist.

A recent prospective cohort study from 2020 concluded that that speech, solid feeding, and sleep difficulties may be linked to restricted tongue function. Overall, speech improved in 89%, solid feeding improved in 83%, and sleep improved in 83% of patients as reported by parents. Fifty percent of speech-delayed children said new words after the procedure, 76% of slow eaters ate more rapidly and 72% of restless sleepers slept less restlessly. After tongue-tie releases paired with exercises, most children experience functional improvements in speech, feeding, and sleep. Providers should screen for oral restrictions in children and refer for treatment when functions are impaired. Parents should be educated about the possible long-term effects of

tongue-tie while their child is young (< 1 year of age), so that they may make an informed choice regarding possible therapy [4].

Robust policies regarding newborn screening for tongue-tie and community referral processes for assessment and management are needed. Furthermore, the findings will help inform the development of educational programming, clinical practice guidelines and hospital and community policies for health care professionals [35]. Physicians can use these self-reported maternal perspectives on frenotomy to guide counseling and improve shared decision making for parents [55].

CONCLUSIONS

Recent studies suggest that breastfeeding, speech, solid feeding and sleep difficulties may be linked to restricted tongue function.

Neonates with tongue-tie are at increased risk for breastfeeding difficulties. An early recognition of this association by primary care provider and prompt referral to a lactation consultant is important. In cases with clearly documented breastfeeding difficulties, frenulum division often results in rapid improvement in symptoms. Although this is a simple low risk procedure, it should be carried out only by those who have been trained in the procedure.

It is fascinating that a hidden string can have such a dramatic impact on human physiology and quality of life, from birth to adulthood. Although treating tongue-tie as young as possible is preferred, it can still drastically increase the quality of life for adults who have tongue-tie. It's never too late to improve someone's life, especially when it is that easy to do it.

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