

Tongue-and lip-tie beyond breastfeeding difficulties

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Abstract. The diagnosis and treatment of tethered oral tie tissues, such as ankyloglossia (tongue-tie) and lip-tie, have grown substantially. Although robust evidence indicates that these abnormal anatomic variations are associated with breastfeeding difficulties, impaired craniofacial growth, sleep, speech and posture in children, both diagnosis and treatment of oral ties remain controversial. The oral cavity displays considerable morphological variation across individuals. One of these variations includes tight, restrictive connective tissue between oral structures known as tethered oral tie tissues (TOTs). The clinical view regarding these anomalies has evolved with increasing interest not only in tongue-tie (ankyloglossia) but also in lip-tie [1-3]. Ankyloglossia has been considered a risk factor for breastfeeding difficulties [4-16]. Recent evidence indicates that TOTs can be also associated with whole-body consequences, such as reflux, dental malocclusion, and respiratory disorders, ultimately increasing the risk of sleep and speech disorders, and detrimental changes in posture and eating patterns [13, 17-25]. The prevalence of TOTs is highly variable across populations and is still a matter of ample debate. Currently, there is a lack of consensus on diagnosis criteria, best surgical treatment techniques, and pre- and post-surgery care [19, 26, 27]. Yet, the diagnosis and surgical treatment of TOTs have substantially increased in recent years [28-31]. This mini-review will summarize evidence-based data regarding the cascade of consequences of tongue-tie and lip-tie in children and the main signs and symptoms of these anomalies in newborns. It will also discuss the available evidence on treatment options for TOTs, including pre- and post-surgical care that may enable better outcomes and prevention of possible complications. For a better understanding, tongue-tie and lip-tie will be addressed separately.

Keywords: ankyloglossia, tongue-tie, lip-tie, diagnosis, treatment.

1. General considerations about ankyloglossia

Despite being an anatomical variation described for decades, no definition of ankyloglossia is widely accepted. The “International Affiliation of Tongue-Tie Professionals” (IATP) defined it as a congenital oral anomaly that results from incomplete apoptosis of tissues in the lower portion of the tongue during embryonic development and limits tongue movements [32, 33]. The etiology of ankyloglossia remains poorly understood. However, genetic causes have been postulated in the incidence of ankyloglossia [34, 35]. It has been recently suggested that high consumption of folic acid, which can cause the excessive formation of connective tissues during pregnancy, including the lingual frenulum, could explain (at least partially) the incidence of ankyloglossia [36].

The diagnosis of ankyloglossia remains controversial due to the lack of diagnostic criteria, which causes great uncertainty about its prevalence. For example, Haham et al, described the presence of a lingual frenulum in 99.5 % of newborns evaluated by their study [37]. The authors also observed that not every lingual frenulum restricts tongue movements; thereby suggesting that the diagnosis and treatment of ankyloglossia should not be based only on the presence of the lingual frenulum [37]. Thus, several authors have proposed that the limitation of tongue mobility is a fundamental criterion for diagnosing and treating ankyloglossia, which should be distinguished as symptomatic ankyloglossia [8, 14, 33, 38].

Different classification schemes and grading systems have been proposed but none is universally used [25, 38-43]. Often, ankyloglossia is classified into anterior ankyloglossia and

posterior ankyloglossia based on the location of the frenulum attachment on the ventral surface of the tongue [40]. Noteworthy, posterior ankyloglossia is usually not visible; thus, its diagnosis depends on the palpation of the area, and remains controversial [44-47].

Due to the different diagnostic criteria used, the prevalence of ankyloglossia reported in neonates has ranged from 0.1 % to 46.6 % [5, 6, 8, 47-50]. Despite the great uncertainty regarding ankyloglossia prevalence in the general population, several reports indicate a higher prevalence in males, with ankyloglossia affecting around 1.4 to 3 males for every female [6, 14, 29, 50]. Therefore, intrinsic differences in the studied populations could explain (at least partially) the heterogeneity of ankyloglossia prevalence estimates reported by different studies [35, 49]. In addition, recent reports indicate a significant increase in the prevalence of ankyloglossia in the United States and British Columbia (Canada) between 1997 to 2012 and 2004 to 2013 [29, 31]. According to Walsh et al., the increased awareness of the negative impact of ankyloglossia on breastfeeding and better national and global initiatives to support breastfeeding may have contributed to a higher rate of screening and, therefore, the increase in the prevalence of ankyloglossia observed in the last decades [29].

2. Consequences of untreated ankyloglossia

There is compelling evidence that ankyloglossia can compromise the tongue propulsion needed for the extraction of breast milk affecting newborns breastfeeding and the development of children with ankyloglossia [51]. In fact, ankyloglossia is associated with breastfeeding difficulties, such as poor latching, pain with breastfeeding, ulceration and bleeding of nipples due to the friction created by abnormal tongue movement, poor milk letdown or incomplete emptying due to insufficient suction capacity [4-16, 33]. If not appropriately treated, ankyloglossia can lead to poor weight gain and provoke aerophagia-induced reflux – due to poor latching, ultimately resulting in early abandonment of breastfeeding [24, 25]. Thus, the reporting and identification of early signs and symptoms may allow timely diagnosis of ankyloglossia in newborns [15]. Table 1 summarizes the most common signs and symptoms associated with ankyloglossia.

Table 1. Most common signs and symptoms associated with ankyloglossia

For the mother	For the newborn
Breast engorgement	Biting/grinding while breastfeeding
Blocked ducts	Clicking noises while breastfeeding
Creased, flattened, or blanched nipples after nursing	Colic symptoms
Cracked, bruised, or blistered nipples	Difficulty swallowing (gagging/choking)
Mastitis or nipple thrush	Falls asleep while attempting to nurse
Nipple and breast infections	Frequent loss of latch
Nipple bleeding	Frequent crying
Nipple ulceration	Heart-shaped deformity
Poor milk letdown	Inability to breastfeed
Poor or incomplete emptying	Irritability with feeding
Severe pain when infant attempts to latch	Milk may leak from mouth
Sore nipples	Opened mouth
	Poor latching
	Poor weight gain
	Prolonged feeding
	Reflux symptoms
	Restriction of tongue protrusion
	Snoring
	Tongue mobility restriction
	Unable to hold a pacifier in mouth

In addition to breastfeeding difficulties, studies indicate that ankyloglossia can affect maxillofacial development, being linked with maxillary hypoplasia and soft palate elongation,

craniofacial alterations, and the development of dental malocclusions and mouth breathing [18, 23, 33]. Changes in craniofacial growth can decrease the size of the upper airways, causing breathing disorders such as obstructive sleep apnea. The inability of the tongue to be positioned upward against the palate at rest also contributes to the development of obstructive sleep apnea associated with ankyloglossia, impairing sleep quality [17, 20-22]. Moreover, restriction of tongue movements can provoke articulatory disorders, leading to detrimental compensatory strategies for speech production and eating patterns [20, 52].

The anatomy of the lingual frenulum has been described in detail only recently. Tissue microdissection studies of the lingual frenulum revealed that it is a structure formed by the dynamic elevation of a midline fold on the floor of the buccal fascia and not merely a tissue band [53, 54]. Of note, fascia connects with the rest of the body [19, 33, 55]. Thus, the restriction of tongue movements caused by ankyloglossia can generate tension in the fascial system, contributing to the development of neck and back pain, and postural dysfunctions in both infants and adults [56].

3. Treatment

Ankyloglossia is mainly treated with surgical techniques, which involve a simple cut in the frenulum (frenotomy) or the complete excision of the frenulum (frenectomy). However, no consensus on the indication, time for surgical release, or surgical method exists [8, 14, 27, 45]. Frenotomy is the most common procedure employed for ankyloglossia treatment and, when performed correctly, has a low risk of complications.

Although a number of randomized trials have been conducted showing the benefits of frenotomy for breastfeeding difficulties, most of the evidence comes from observational studies [1, 5, 11-13, 15, 48, 57-61]. O'Shea et al, consider that, the accumulated body of evidence supporting the benefits of frenotomy has a low to moderate certainty [62]. Of note, conducting sham-controlled randomized trials for tongue-tie represents a significant research challenge, as sham frenotomy can be considered an unethical procedure. The replacement of randomized evidence with evidence from observational studies has been suggested as a way to overcome such ethical issues [63, 64]. Overall, a large body of observational evidence indicates that frenotomy is associated with significant improvements in breastfeeding in about 50-79 % of newborns. Frenotomy is also associated with benefit to mothers, who show significant reduction in nipple pain after the procedure [5, 7, 11-15]. Additional reported benefits of frenotomy include improved speech, feeding and sleep patterns, mouth breathing, snoring, bruxism, muscle tension, sleep apnea, and reflux [17, 19-22, 24, 25, 64-66].

Despite the adequate overall safety profile, frenotomy can result in complications, such as pain, bleeding, infections, injuries to the lips and/or to the Wharton ducts, lingual dysfunction, poor feeding, respiratory events, weight loss and delayed diagnosis of alternative underlying medical issues [14, 45, 67, 68]. Besides, frenotomy is also associated with the risk of neuronal damage [53]. Inadequate healing and ankyloglossia recurrence are also possible complications, reported in 0.5-13 % of surgically-treated ankyloglossia cases. Of note, ankyloglossia recurrence is more common in posterior ankyloglossia than in anterior ankyloglossia and may require multiple surgical corrections [9, 45]. Serious adverse events have also been observed after surgical treatment of ankyloglossia. For example, a case of Ludwig angina has been described after frenuloplasty [69]. Two cases of severe bleeding with hypovolemic shock and three cases of airway obstruction were also reported following frenotomy [70-72]. Two out of these three cases of airway obstruction were diagnosed in patients with Pierre Robin Sequence, a neonatal disorder characterized by an underdeveloped jaw (micrognathia), backward displacement of the tongue (glossoptosis) and upper airway obstruction [71, 73]. Thus, practitioners and parents/families should be aware of possible frenotomy-related complications and frenotomy should be considered with caution in infants with Pierre Robin Sequence or other craniofacial anomalies.

While conventional frenotomy employs scissors or a scalpel, most dentists currently use a laser

to perform frenotomy. It has been proposed that the use of a laser could provide several advantages when compared to traditional techniques, as it causes little or no bleeding, and is associated with less pain intensity and inflammation. However, current evidence does not support this notion [27]. Thus, professionals involved in diagnosing and treating ankyloglossia must receive adequate training to judiciously select the infants who can benefit from frenotomy. Moreover, these professionals should be aware of the safety profile of different surgical equipment.

Non-surgical strategies have been also proposed to manage ankyloglossia that can be used either as alternative or complementary to the surgical treatments [33, 74-78]. The non-surgical strategies are generally implemented under the guidance of professionals specializing in breastfeeding and involve using nipple shields, changing babies' positions during breastfeeding, and tongue stretching exercises [79]. It has been suggested that bodywork techniques (Bodywork), manual massage and tummy time could relieve muscle tension associated with ankyloglossia [80]. Physical therapy, speech therapy, and alternative and complementary medicine treatments, including craniosacral therapy, naturopathy, and orofacial myofunctional therapy are also part of the non-surgical armamentarium [14]. However, no randomized trials evaluating the effectiveness and safety of these non-surgical approaches to the treatment of ankyloglossia have been performed to date. Recently, Zaghi et al. described the combination of myofunctional therapy before and after the frenuloplasty procedure as a treatment regime for patients of different ages with ankyloglossia [19]. The authors suggest that this combination is an effective and safe treatment for a select group of patients who present mouth breathing, snoring, bruxism, and myofascial tension. However, additional studies are needed to validate this form of treatment.

The best clinical outcomes for the managing ankyloglossia are more likely achieved with a multidisciplinary health care team, with a patient-centered approach, assessing and treating patients holistically. A simple snip or clip with scissors is unlikely to achieve satisfactory long-term benefits. In this respect, a holistic approach should include a detailed clinical anamnesis, appropriate clinical management (e.g., a complete frenulum release), post-frenotomy stretches and exercises, and comprehensive patient follow-up. Lactation support, feeding therapy, speech therapy, and manual therapy or bodywork are paramount to address fascial restrictions [33]. Finally, it is essential to understand that tongue-tie is not the only limiting factor for normal oral function. The tone, the space and the compensation of the tongue are other factors that need to be taken into account during oral function assessment. Similarly, the detrimental consequences of tongue-tie on the health of the newborn do not comprise uniquely breastfeeding difficulties. Thus, treating tongue-tie with frenotomy without a multidisciplinary approach may be a suboptimal strategy [19, 43].

4. Lip-tie

The superior labial frenum region is not part of the routine newborn physical examination. In 1995, Wiessinger and Miller presented one of the first pieces of evidence that a tight labial frenum can be associated with breastfeeding difficulties [1]. This case report highlighted that the identification, classification, and subsequent treatment decision of TOTs should be reviewed and consider the anatomical variations of the upper labial frenum in neonates [3].

In contrast to the vast literature available on ankyloglossia, the normal morphology of the upper labial frenum is poorly known [3, 81]. In 2004, Kotlow proposed a simple classification system to support the diagnosis and treatment of lip-tie [82]. Besides the breastfeeding difficulties in neonates, lip-tie has been associated with the development of caries in the incisors due to the difficulty of access to cleaning, diastema, and reflux [24, 25, 48, 83]. Altogether, the available evidence support the inclusion of the upper labial frenum region in the routine physical examination of the newborn to comprehensively examine and treat TOTS and their associated complications.

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Data availability

The datasets generated during and/or analyzed during the current study are available from the corresponding author on reasonable request.

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