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Ankyloglossia in infants and its impact on breastfeeding - etiology, diagnosis, treatment. Literature review

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ABSTRACT

Ankyloglossia is a condition of limited tongue mobility caused by a restrictive lingual frenulum. Proper tongue mobility is essential for proper food intake (breastfeeding, speech development or proper development of facial bones). There is no clear classification that would indicate the need for surgical intervention or conservative treatment.

Material and method: A literature review was conducted by searching the PubMed database, Google Scholar, literature using the keywords 'ankyloglossia', ‘tongue tie', 'frenotomy,' ‘breastfeeding’, :lingual frenulum’. Focusing on the most recent reports, the selected articles were published from 2019 to 2024.

Brief description of the state of knowledge: The aim of the study was to review the literature on current recommendations regarding etiology, diagnostics, typical clinical manifestations of ankyloglossia in newborns and infants with particular emphasis on the effect on breastfeeding, indications for surgical treatment, complications after surgery and its effectiveness.

Result and conclusion: There is no clear consensus in the literature on the management of surgical treatment. However, There is a consent that surgery should be performed as soon as possible after an infant has been diagnosed with ankyloglossia and breastfeeding problems that do not improve with conservative treatment.It should be noted that surgical treatment is not the only method of dealing with ankyloglossia.

Keywords : 'ankyloglossia", tongue tie ", ‘frenotomy’,’breastfeeding’, ‘lingual frenulum’

INTRODUCTION

The frenulum of the tongue is a remnant of tissue – a three-dimensional, dynamic structures located in the midline of the tongue between the ventral surface and the floor of the mouth. The pathology may concern both the abnormal location of the lingual insertion, the lower attachment and the abnormal length of the frenulum itself. Its sufficient length, as well

as proper mobility, is needed to perform the necessary functions, such as food intake, correct pronunciation.

From birth, we may encounter a problem with a disturbance in the sucking mechanism and difficulties in taking breast food. If the mobility of the structures responsible for sucking is disturbed, the child will engage his body more – he will tense up by presenting himself, e.g. clenching his fists. This, in turn, will lead to stiffening of the tissue - stiffness of the neck or chest, increased tension. It should also be remembered that the limited mobility of the tongue affects its resting position, which is essential for the development of the maxilla and mandible, or the correct type of swallowing. It should also be remembered that the limited mobility of the tongue affects its resting position, which is essential for the development of the maxilla and mandible, or the correct type of swallowing.

In the case of a diagnosis of pathology, it is necessary to intervene starting with conservative methods (consultation with a lactation consultant, functional myotherapy) and if these do not bring results, making a decision about frenotmi. However, the lack of a uniform classification makes diagnosis difficult.

Results:

Tongue frenulum – structure, definitions

The frenulum of the tongue is a three-dimensional dynamic structure that connects the floor of the mouth to the ventral part of the tongue. It is located centrally in the floor of the mouth. They are formed by a fold made up of the connective tissue of the fascia.[1] The fascia attaches to the inner surface of the lower jaw - forming the floor of the mouth. Movements of the tongue cause tension in the middle part of the floor of the mouth, lifting the median fold - and the frenulum of the tongue is formed. [2]

There are anatomical varieties that differ in the height of the posterior attachment to the tongue, the height of the attachment to the mandible and the length of the frenulum. Ankyloglossia is a condition of limited tongue mobility caused by a restrictive lingual frenulum. Experts in 2020 came to a consensus on the definition of anterior and posterior ankyloglossia. Anterior ankyloglossia is a condition in which the frenulum attaches to the tip of the tongue or close to it, and thus limits its mobility. In a study by Ghaheri et al. 78% of newborns experiencing breastfeeding problems were diagnosed with 7 posterior ankyloglossia. Posterior ankyloglossia is more controversial because it is recognized by part of the scientific community as a regularity. Posterior ankyloglossia is a condition when the frenulum of the

tongue attaches to the posterior, ventral part of the tongue, restricting its mobility, as well as that it is a submucosal attachment of the frenulum of the tongue. [3]

Occurrence

Over the past decades, the number of diagnoses of ankyloglossia and the number of cases treated with frenotomy have increased.[4] Currently, the prevalence is estimated at 2-10%. In children under 1 year of age, it is about 8%. More often in males in the proportion of 2.5 : 1.2 , a relationship with the linked X chromosome or with TBX22 mutations is assumed. [4,6] Ankyloglossia may be sporadic or familial, and may be part of rare syndromes, such as Moebius syndrome.

A study in a hospital in Brasila showed that the degree of kinship that linked the children to the diagnosed short frenulum was a cousin.This increase in cases is attributed to increased awareness of the benefits of breastfeeding for both mother and baby[5 ,29] The WHO recommends exclusive breastfeeding during the first 6 months. [7,25]

Classifications, diagnostics of ankyloglossia

There are very many classifications of the lingual frenulum. I will present a few of them dedicated mines for newborns and infants.

1.Coryllos classification - determines the type of frenulum of the tongue, it is based only on the anatomical structure , without taking into account the function or criterion of ankyloglossia.

Type I- Frenulum attachment to the tounge tip, usually in front of the alveolar

Type II- Frenulum attachment 2-4 mm behind the tongue tip and on or just behind the alveolar.

Type III: The frenulum is thick, fibrous and less elastic, and attached to the mid-tongue. The tongue cannot be lifted, and is shaped like a scoop when extension is attempted.

Type IV: The frenulum is not visible, but when touching the area, the examiner can feel a fibrous or submucous, thick and slightly elastic anchor, from the base of the tongue to the floor of the mouth. The tongue cannot be lifted and performs asymmetric movements when extension is attempt [4]

2. Hazelbaker’s Assessment Tool for Lingual Frenulum Function Tabela 1.

Function Items	Clinical Features	Points
Lateralization	Complete	2
	Body of tongue but not tongue tip	1
	None	0
Lift of tongue	Tip to mid-mouth	2
	Only edges to mid-mouth	1
	Tip stays at the alveolar ridge or rises only with jaw closure	0
Extension of tongue	Extends the tip over the lower lip	2
	Extends the tip over the lower gum only	1
	Neither of above	0
Spread of anterior tongue	Complete	2
	Moderate or partial	1
	Little or none	0
Cupping	Entire edge, firm cup	2

The side edges only, moderate cup	1

3. Kotlow's classification divided the ankyloglossia into 4 classes: which evaluates the length of the free part of the tongue and measures the distance from the tip of the tongue to the place of attachment on the tongue.

– Class I: mild ankyloglossia (12–16 mm);

– Class II: moderate ankyloglossia (8–11 mm);

– Class III: severe ankyloglossia (3–7 mm);

– Class IV: complete ankyloglossia (less than 3 mm)[8,9]

4.Marchesan described a clinically useful classification that measures the difference (given as a percentage) between the maximum mouth opening with the tongue resting on the floor of the mouth and the tongue touching the palatine papilla. On the basis of Martinelli's classification, a newborn language screening test was performed from the Lingual Frenulum Protocol for Infants (LFPI)[9,27]

Initially, the resting position of the lips while the infant was asleep was assessed; The lips could be closed, open, or semi-open. Then, when the newborn was awake, it was observed whether the position of the tongue during crying was midline, raised, midline with the sides raised, or the tip of the tongue down with the sides raised. The shape of the tip of the tongue raised during crying or lifting maneuvers could be round, with a slight V-shaped slit, or heart-shaped. It was then observed whether it was possible to visualize the frenulum of the tongue or make it visible during the manoeuvre. The thickness of the frenulum was classified as thin or thick. The attachment of the frenulum to the sublingual surface of the tongue can be in the middle third, between the middle third and the apex, or in the apex. The TABBY tool consists of 12 images that show the appearance of the tongue, gum attachments and mobility limitations Score on a scale of O to 8. It is a clear, simple addition to the assessment of ankyloglossia.

Look at the table with description below [10]

TABBY category	**Guidance on use of TABBY**
What does the tongue-tip look like	This is usually the most obvious and most likely to be noted by parents. A notch in the tip of the tongue may only be noticed when the baby lifts the tongue(0-2 pkt)

Where it is fixed to the gum?	With some training and experience this can be assessed visually. If it is difficult to see, then the assessor can [with parental consent] gently use their index finger to feel where the frenulum is attached.(0-2 pkt)
How high can it lift (wide open mouth)?	This can be the most difficult to teach. The assessor needs awareness of normal tongue lift in infants.(0-2 pkt) The tongue may curl back when restricted and so appear to lift. The lift is most easily viewed if the infant is awake and crying. If the baby is not awake, then the assessor can digitally lift the tongue to assess.(0-2 pkt)
How far can it stick out?	This is not always easy to assess in newborn infants. It can be helpful to ask parents what they have noticed, and the pictures can be helpful in discussing this. The easiest way to assess protrusion is to watch the baby as they latch to the breast; are they able to bring the tongue out to latch?(0-2 pkt)

Results:
- 8 Normal Functioning
- 6.7 is the limit of the correct value
- 5 and lower impairment of function
- if 4 or less + breastfeeding problems - indication to consider frenotomy

6.Na also noteworthy is the Ostapiuk classification, which combines the anatomical and structural and clinical aspects in the study.
It takes into account:
- frenulum structure thin, flexible - under the frenulum there is a transparent membrane over 1 mm thick - palpable resistance
b.coarse pow 2
-lower attachment - between the lower incisors, near the lower gums, near the mouth of the sublingual salivary gland
-the shape of the dorsal part of the tongue should also be assessed
-condition of the mucous membrane – anemia of the mucous membrane when raising the tongue to the palate and the presence of additional folds

-ability to perform primary functions and articulation - free achievement of vertical-horizontal position [11]

Noteworthy is the scale of clinical assessment of feeding

Infant Breastfeeding Assessment Tool Tabela 3.

Items	**3 points**	**2 points**	**1 point**	**0 point**
To get the baby to feed, you:	Placed the baby on the breast as no effort was needed	Used mild stimulation such as unbundling, patting, or burping	Unbundled baby, sat baby back and forward, rubbed baby's body or limbs vigorously at the beginning and during feeding	Could not be aroused
Rooting	Rooted effectively at once	Needed coaxing, prompting, or encouragement	Rooted poorly even with coaxing	Did not root
How long from placing baby on the breast to latch & suck?	0 – 3 min	3 – 10 min	Over 10 min	Did not feed
Sucking pattern	Sucked well throughout on one or both breasts	Sucked on & off but needed encouragement	Sucked poorly, weak sucking; sucking efforts for short periods	Did not suck

Consequences of a short frenulum

It has been reported that in 25-44% of infants with anterior ankyloglossia, restricted tongue movements may impair sucking and swallowing. [10,12]Difficulties are not observed in bottle-fed infants.

Breastfeeding requires a proper grasp of the breast, i.e. opening the mouth wide, protruding the tongue, sealing the areola. The suction mechanism was analysed by dynamic ultrasound and electromyography, taking into account the work of suprahyoid muscles. It consists of two phases: the first - the suction phase when the mouth is wide open, the tongue lowers downwards, and a negative pressure is created inside the mouth.

If breastfeeding problems have been found, the first step is to expand the history and further differential diagnosis. To rule out potential possible causes such as: abnormal development of the palate, maxilla or mandible; neurological or cardiovascular disorders; and upper respiratory tract obstruction or reflux. It is essential to take into account various lactation factors, such as the mother's experience in breastfeeding. If you have any problems with breastfeeding, you should start with the advice of a lactation consultant.[4,10]

Consequences of Ankyloglossia

Ankyloglossia affects the incorrect resting position of the tongue, as it is impossible to latch on to the palate. This results in limited ventilation and translates into habitual mouth breathing, snoring. In addition, it can interfere with the development of the correct adult type of swallowing. No correlation was found between speech impediment and short frenulum [5]

There is a hypothetical conclusion that a short frenulum causes malocclusion, especially chewing progenia. No correlation was found between speech impediment and short frenulum [5]There is a hypothetical conclusion that the short frenulum causes malocclusion, especially mandibular progenia, this is due to the low resting position, which exerts pressure on the mandible forward.[4]

Infants with a short frenulum have been noted to have greater salivation. Infants with a short frenulum have been noted to have greater salivation.

Breastfeeding problems

To decide whether surgical intervention is necessary, it is also needed to assess the quality of breastfeeding.[5,4]

Signs, symptoms in the mother and in the child - suggesting a problem which is ankyloglossia.

- poor weight gain
- injured, painful nipples, recurrent mastitis
-difficulty latching, poor latching, calluses after breastfeeding
-facial asymmetry, hypertrophy of the cheeks
-prolonged or very frequent feedings, irritability

It consists of two phases: the first - the suction phase when the mouth is wide open, the tongue lowers downwards, and a negative pressure is created inside the mouth. The second - compression - there is a closure of the maxilla and mandible, the tongue moves upwards and milk is swallowed. Limited mobility of the tongue leads to poor sealing around the nipple and consequently to ineffective suction. The infant tries to compensate for this by catching the gingival papilla, which causes pain and cracking of the nipple.

The second - compression - there is a closure of the maxilla and mandible, the tongue moves upwards and milk is swallowed. Limited mobility of the tongue leads to poor sealing around the nipple and consequently to ineffective suction. The infant tries to compensate for this by catching the gingival papilla, which causes pain and cracking of the nipple. As a result, milk flow is impeded, which can lead to early weaning.[16]

If tongue mobility is reduced, the infant increases the suction pressure. The milk duct circuit, which is flexible and has low pressure, collapses with instead of increasing flow, leading to reduced efficiency. The infant's effort increases, as does the local pressure in the breast11. The infant's effort increases, as does the local pressure in the breast[11]

How to examine a newborn?

According to a pilot study of the UFMS University Hospital, carried out by trained persons.-Initially, the resting position of the lips during the infant's sleep (closed, open or semi-open) was assessed. Then, when the newborn was awake, it was observed whether the position of the tongue during crying (raised in the midline, in the midline with the elevation of the sides or the tip of the tongue down with the elevation of the sides.

The shape of the tip of the tongue raised when crying or when it is raised (round, with a slight V-shaped or heart-shaped slit). It was then observed whether it was possible to visualize the frenulum of the tongue or during the maneuver. Thickness of the frenulum (Thin , thick). Attach the frenulum to the sublingual surface of the tongue (middle 1/3 J, between the middle third and the tip of the tongue or at the apex). Attach the frenulum to the floor of the mouth (from the sublingual side or the alveolar crest).[5,7,12] The frenulum should be carefully palpated to check its response to lateral and posterior pressure[20]. Surgical treatment - frenotomy It is important to note that not all babies with a short frenulum need to have a problem with breastfeeding. Studies show that some infants with ankyloglossia are able to effectively breastfeed without surgery. [4]According to Ricke et al., 80% of newborns diagnosed with ankyloglossia are able to breastfeed effectively thanks to the adaptability of the tongue. In Kumar's study, and in half of the children studied, they had a short lingual frenulum that did not impede breastfeeding. [23] The first step after finding a short frenulum, which affects the disorder of the normal function of the tongue and the accompanying difficulties in breastfeeding, is conservative treatment. (consultation with a lactation consultant and myofunctional therapy) including extraoral and intraoral stimulation to improve sucking reflexes in infants. The goal of the therapy is to create awareness of the posture and function of the tongue in the mouth, to strengthen and tone the muscles of the tongue and the orofacial complex, and to eliminate harmful habits][13,22.] The therapy is carried out by a speech therapist or neurologopedist. S. Zaghi recommends OMT at least 1 month before surgery and 2 months after. [5,9,14,15]

Contraindications to the procedure are neuromuscular disorders, hypotonia, retrognathia and mcrognation. In these anomalies, frenotomy can block the airway, make swallowing difficult, through (glossptosis) collapse of the tongue.e.g. Pierre ROBIn [4,5.] Do not forget about coagulation system disorders, where the child should be properly prepared and the operator should be an experienced operator with appropriate facilities.

The course of the procedure

Frenotmi is recommended for infants diagnosed with ankyloglossia who have difficulty breastfeeding after other conservative treatments have failed. [17] The infant is immobilized with a diaper or a Papoose board, the assistant holds the baby's head by pulling down his chin. The doctor lifts the tongue with a spatula, making the frenulum visible, then cuts the tissue. If the frenulum is thin and clearly visible, one incision is made to the groove connecting the tongue and the floor of the mouth at the thinnest point of the frenulum, parallel to the tongue.

Inspect with your eyes or finger. Sometimes it is necessary to gently cut the groove under the tongue. However, if the access is difficult (or if the posterior frenulum is affected) it is necessary to first cut to a depth of 1 mm in the thinnest free part of the frenulum, and subsequent incisions diverging in the parts of the frenulum on the left and right sides. When fully released, a diamond-shaped wound[4.11] is obtained. Be careful not to damage the tongue, sublingual muscle, salivary glands, gums or chin-lingual muscle. Applying pressure with the index finger to the remaining part of the frenulum is held in place by a gauze pad soaked in 0.9NaCL There is no need for stitches. The baby can be put to the breast immediately. The use of local anesthesia is not recommended up to 4 months. However, after 4 months, some authors recommend performing frenotmi under general anesthesia due to the greater awareness and strength of the infant. [26] Acetaminophen may be considered [16]. To reduce discomfort, 24 % sucrose can be applied a few minutes before the procedure. In a randomized trial, inhalation of lavender oil had positive effects on relieving discomfort in infants 19In the Griffiths study, the average crying time during a frenotomy was 15 seconds. [26]After the procedure, the child should stay for 20-30 minutes of observation. Paracetamol may be recommended when the child is concerned. A follow-up visit should take place a few days after the procedure.

Recommendations after the procedure

Massages should be intense and take place before each feeding for at least two weeks. because that's how long it takes to heal [11]They consist of lifting the tongue or stretching exercises to stretch the tissues after surgery. However, there are no studies proving the legitimacy of such a procedure. CML . The risk of complications is rare, about 9%. These include bleeding – estimated at 3%, airway obstruction, damage to surrounding structures, cyst formation, scars [4.5, 20]

Benefits and prognosis after surgery

However, clinical work and general studies and case reports show that frenotomy of the lingual frenulum is associated with improved breastfeeding based on appropriate indications [16,24,28]-In a randomized trial conducted by Buryk and Cochrane with ankyloglossia aged 1 to 35 days, frenotomy was found to immediately correct breastfeeding and reduce breast pain in the mother.[8,30] After re-evaluation of the LATCH scale and the maternal pain scale, it was found to show improvement in breastfeeding.

Discussion

The frenotomy procedure is performed in the neonatology, otolaryngology or on an outpatient basis. The timing of the frenotomy is debatable. In the Prabin study, it was found that infants who underwent frenotomy before 8 days of age gained more weight than after 8 days of age. Some recommend observation of 2.3 weeks in infants with a mildly shortened frenulum. [26,33]Studies show that delaying the procedure until the infant was 4 weeks old resulted in a high rate of breastfeeding discontinuation. However, some authors recommend waiting until the age of 4 to undergo the procedure, when pronunciation assessment can be performed[24]

If ankyloglossia is diagnosed and there is no improvement after conservative treatment, a frenotomy procedure should be performed as soon as possible. [30]Thus, although many specialists support the early release of the frenulum of the tongue as an aid in the breastfeeding process, many have been questioned. Frenotomy can be performed using a laser or a scalpel. The latest AAO-HNSF consensus states that due to the lack of adequate research on a better treatment method, She also emphasizes that if ankyloglossia is diagnosed and there is no improvement after conservative treatment, a frenotomy procedure should be performed as soon as possible.[30]Tissue studies have shown, however, that scalpel incisions made on pig tongues showed less thermal tissue damage and inflammation compared to laser, electrosurgery or constant voltage electrocoagulation. The use of laser reduces postoperative bleeding and minimizes postoperative swelling and pain[7] . The NICU O research is also]promising, showing that laser phrenomy is effective and safe. Laser frenotomy resulted in low pain and very few complications both intra- and post-operative, and all wounds healed within thirty days with no recurrence of ankyloglossia. It has also shown significant improvements in breastfeeding and growth in newborns with a reduction in nipple pain in mothers. . In a study by Haytac et al. et al. showed that patients treated with CO2 laser had lower pain values on the visual analogue scale (VAS) compared to patients treated with traditional methods. It is important to note that the laser does not cut through tissues, but causes denaturation and coagulation[6] The number of exercises and the period of time for which myofunctional therapy should be carried out are also debatable. Postoperative tongue stretching exercises vary depending on the authors. They range from 1 to 6 times a day.

Applications

It is known that, although several standards and systems have been developed to analyze and classify the severity of ankyloglossia, none of them has become universal, which makes it

difficult to diagnose and justify surgical treatment The same case can be recognized as an indication or not, depending on the criteria of the doctor's evaluation.

Before starting any treatment, the infant should be evaluated for any other conditions affecting breastfeeding. An infant diagnosed with ankyloglossia, but without functional disorders in lactation, should be monitored and frenotomy should be performed only if and mainly when there is a justification. It should be emphasized that frenotomy, leads to an improvement in feeding . However, this is not the only method to deal with a short frenulum. Prior consultation with a lactation consultant is very important, as well as the increasingly appreciated myofunctional therapy carried out by speech therapists. This therapy has a justified effect, because in the histological structure of the frenulum there is type III collagen and elastin, which is stretched. The possibility of working with the frenulum, and greater awareness of the parents, can protect against unnecessary frenotomy treatment. And the combination of myofunctional therapy with the treatment gives the best results.

Disclosures

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