Review

Shortened Lingual Frenulum in Newborns - Should We Perform Frenectomy? Where Are We Now, and Where Are We Headed?

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Shortened Lingual Frenulum in Newborns - Should We Perform Frenectomy? Where Are We Now, and Where Are We Headed?

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Abstract

Breastfeeding is the only appropriate way to feed healthy newborns. A shortened lingual frenulum in newborns is considered one of the main causes of difficulties in effective breastfeeding, sore nipples, and breastfeeding discontinuation. In recent years, there has been an increasing incidence of diagnosed shortened lingual frenulum in newborns, with this clinical condition being considered the sole cause of breastfeeding difficulties. At the same time, the frequency of frenulum cutting procedures has recently increased dramatically and is becoming an increasingly common procedure in newborns. This raises concerns among both mothers and physicians. Our paper aims to answer whether a frenotomy is always necessary and justified, and whether it has advantages over simple, non-invasive conservative methods.

Keywords: breastfeeding; lingual frenulum; ankyloglossia in newborn; tongue-tie release

1. Introduction

Shortened lingual frenulum in newborns is mentioned as one of the potential causes of difficulties in effective breastfeeding [1,2].

Breastfeeding is the only appropriate way to feed healthy newborns. It is essential for achieving proper growth, development, and shaping the child's health. It is recommended to start breastfeeding within the first hour after birth and continue exclusive breastfeeding for the first 6 months of the child's life, which helps infants develop and maintain health.

Furthermore, it is recommended to start breastfeeding within the first hour after birth, continue exclusive breastfeeding for the first 6 months of the child's life, and expand the child's diet by maintaining breastfeeding for up to 2 years or longer [3,4].

Breastfeeding has numerous proven health benefits for children, including preventing infections, reducing the incidence of malocclusions, improving IQ in both childhood and adulthood, and preventing obesity and diabetes. Additionally, breastfeeding mothers have a lower risk of developing ovarian and breast cancers [5].

Given the proven health benefits of breastfeeding, medical personnel should provide mothers with thorough education and support, as well as assistance in resolving identified problems. Although breastfeeding, by definition, is natural, it is a complex medical process involving the mother's developing lactation and the child's developing breastfeeding skills. These parallel processes are unique to each mother-child pair and have the potential to involve complications at any stage of the World Health Organization (WHO) recommended breastfeeding period. In accordance with medical ethics, any interventions regarding breastfeeding problems must be based on medical

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knowledge, as with any other patient health situation, and cannot be of an intuitive or traditional counseling nature [6].

In recent years, lactation counseling has been noting the rapidly increasing frequency of diagnosing a shortened lingual frenulum in newborns and simultaneously recognizing this clinical situation as the cause of breastfeeding difficulties [7,8]. At the same time, the frequency of frenulum cutting procedures has recently increased dramatically and is becoming an increasingly common procedure in newborns. This raises concerns among both mothers and physicians. Our paper aims to answer whether a frenotomy is always necessary and justified, and whether it has advantages over simple, non-invasive conservative methods.

2. The Lingual Frenulum, Ankyloglossia, and Breastfeeding

A frenulum is defined as a plate or fold that connects two anatomical elements and restricts their mobility. The human body contains many frenula, including the lingual frenulum. Tongue development begins around the fifth week of embryonic life. Growth of the primary oral epithelium leads to the formation of lamellae, which transform into grooves that separate the developing tongue from the floor of the mouth. The tongue bud's complex development (branches I, II, and III) explains its intricate structure and innervation [9]. Traditionally, the lingual frenulum was defined as a fold of connective tissue in the midline that connects the undersurface of the tongue to the floor of the mouth.

The analysis of current data from anatomical and histological studies of the frenulum in newborns provides a more detailed understanding of its structure. In 2019, N. Mills et al. conducted post-mortem examinations of neonates and adults and made a statement on individual qualifications and the differentiation of the anatomical and histological structure of the tongue frenulum [10,11]. It has been unequivocally shown that the frenulum is a dynamic, layered structure. It is built from nerves and supplied to executive devices, and it moves in the final line, secondarily determining the tongue via the genio-glosses cellular network. This muscle acts on the bottom of the tongue and is one of the elements forming the frenulum. Histological studies have shown a variable proportion of type III collagen and elastin content in the components forming the frenulum of the tongue.

Furthermore, there is no simple relationship between the anatomical and histological structure of the frenulum and the resulting visible morphology on external examination or the presence of impairment of tongue function.

Similarly, the location at which the frenulum attaches to the tongue has no direct relationship to the frenulum's internal structure, nor is it an indicator of tongue function as assessed during testing. Rather, it is only one of many elements that determine frenulum structure.

All features of the lingual frenulum's appearance, as captured in commonly used scales for assessing frenulum structure and function (such as Amir and Hazelbaker), are unrelated to the anatomical and histological structure of the sublingual frenulum. Furthermore, in the cited study, the authors emphasize that the lingual frenulum is connected to the anterior part of the tongue and is associated with its mobility. However, there is no anatomical connection between the lingual frenulum and the body and base of the tongue, which have completely different embryological origins.

For this reason, the term "posterior frenulum of the tongue" is incorrect, and the mentioned authors do not recommend its use. When analyzing the structure of the floor of the mouth in the context of qualifying for potential surgical procedures, it should also be considered that the sublingual surface contains, among other things, nerve endings of the hypoglossal nerve, blood vessels, and the openings of the sublingual salivary glands, which can be damaged during surgical intervention. The risk of such a potential complication is difficult to estimate, given the previously emphasized enormous individual variation in the anatomical and histological structure of the frenulum in the studied patients [10–12].

The lingual frenulum is an anatomical structure present in every human being, with rare exceptions. The literature contains case reports of patients with lingual frenulum agenesis who are affected by Ehlers-Danlos disease (which is characterized by a disruption in collagen synthesis). The

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absence of the lingual frenulum is pathognomonic for the diagnosis of Ehlers-Danlos disease [13]. In the healthy population, the lingual frenulum, with its individual anatomical variation, is a typical structure of the floor of the mouth.

A shortened lingual frenulum (ankyloglossia, tongue-tie) is a term used to describe a situation where the structure of the lingual frenulum is considered to restrict proper tongue mobility. This diagnosis is made based on an assessment of the external structure and function of the tongue. This assessment is most often performed by healthcare professionals, who usually use various assessment scales, among others, the Hazelbaker scale, modified by Amir in 2006 [14].

Table 1. Hazelbach Assessment Tool for Lingual Frenulum Function in the modification of Amir (HATLFF).

Functional assessment
The first three criteria are assessed according to the Hazelbaker/Amir scale.
Lateralization 2: complete 1: body of the tongue, but not the tip 0: neither body nor tip
Tongue lift 2: tip to the middle of the mouth 1: only the edges of the tongue to the middle of the mouth 0: tip of the tongue remains at the alveolar ridge or is accessible to the middle of the mouth when the mouth is closed and/or the center of the tongue collapses
Tongue protrusion 2: tip above lower lip 1: tip only to alveolar ridge 0: none of the above, or the anterior or middle part of the tongue curls into a hump and/or collapses
The following four criteria are scored on the full Hazelbaker scale
Anterior tongue protrusion 2: complete 1: moderate or partial 0: slight or absent
Tongue grip 2: the entire tongue edge strongly grips the finger 1: only the lateral edges of the tongue grip the finger, or the gripping force is moderate 0: weak or absent grip
Peristalsis 2: full from the front 1: partial or starting behind the tip of the tongue 0: no peristalsis or reverse peristalsis (thrusting)
Clapping 2: not present 1: periodic 0: often or with every sucking movement
APPEARANCE ASSESSMENT
Appearance of the tongue when raised 2: rounded or square 1: slight gap at the tip 0: heart-shaped
Frenulum Flexibility 2: more than 1 cm or no frenulum 1: 1 cm 0: less than 1 cm

 doi:10.20944/preprints202511.0031.v1

Frenula attachment to the tongue 2: the frenulum occupies 50% or less of the ventral surface of the tongue in the midline 1: the frenulum occupies 50-75% of the ventral surface of the tongue in the midline 0: the frenulum occupies 75-100% of the ventral surface of the tongue in the midline
Frenula attachment to the inferior alveolar ridge 2: attached to the floor of the mouth or far below the gum 1: attached just behind the gum 0: attached to the gum

Table 2. The Hazelbaker Assessment Tool for Lingual Frenulum Function – Assessment summary.

Assessment Summary
14 = Perfect Excellent function score regardless of appearance score. Surgical treatment is not recommended.
11 = Acceptable Acceptable function score if the appearance score is at least 8. The child may be experiencing temporary difficulties, may not have presented well, or may have some other problem that needs to be addressed.
<11 = *The score indicates impaired language function. Frenectomy should be considered if conservative measures fail to improve.* *A function score of 9-10 combined with an appearance score of 8-9 is considered borderline; all other supportive strategies should be exhausted before surgery is considered. Bodywork is recommended.*
< 8 = *Frenotomy is necessary if the score for appearance and function is less than 8 points.*

Table 3. The Assessment Tool for Lingual Frenulum Function in the modification of Amir.

Summary of the assessment according to the modified Amir scale
Amir, assessing the reliability of the scale, stated that three criteria—lateralization, tongue elevation, and tongue protrusion—are fundamental in assessing tongue function. There is agreement among independent observers on this point. There is no agreement between independent observers regarding the remaining four criteria. Therefore, Amir suggests using a simplified scale based on these three criteria. A child can receive a maximum score of 6 points. Frenotomy is recommended for a score of 4 or less for function, and 0-7 for appearance, according to Amir's simplified scale.

3. Frenotomy

Frenotomy is a surgical procedure to cut the frenulum of the tongue. In recent years, this procedure has been widely recommended and performed in newborns [1]. In Poland, there are no clear recommendations regarding the indications and type of procedure for performing frenotomy. This procedure is largely performed commercially, which prevents the data on the exact number of such procedures performed, potential complications, and health outcomes for patients.

Available medical publications emphasize the increasing frequency of both the diagnosis of a shortened lingual frenulum and the performance of frenotomy in recent years [1,7,8]. Data regarding the incidence of ankyloglossia have yielded inconsistent results. The lack of a standardized method for assessing a shortened lingual frenulum is often cited. Researchers use various assessment scales to diagnose a shortened lingual frenulum, which contain numerous elements based on the subjective assessment of the examiner. The Hazelbaker scale presented above, modified by Amir, includes phrases such as "very flexible," "moderately flexible," "small gap," "far in front of the gum," "just in front of the gum," etc. [14]. The different scales used by researchers assess other elements of tongue structure and function, assigning other point values, making it impossible to compare the obtained results [15,16]. It results in the mentioned discrepant data regarding the incidence of the shortened

 doi:10.20944/preprints202511.0031.v1

lingual frenulum, ranging from 0.1% to 11% [8]. At the same time, some publications report an incredibly high incidence of ankyloglossia, amounting to even 20-40% [15,16]. This diagnosis is made more often in male newborns in a 2:1 ratio.

Just as it is difficult to clearly determine the prevalence of a shortened lingual frenulum in newborns, it is equally difficult to assess its impact on breastfeeding.

Researchers attempting to assess this problem use various surveys aimed at mothers, such as the Breastfeeding Self-Efficacy scale, the LATCH Breastfeeding Assessment Tool, which are, by definition, subjective assessments of the mother's situation. Analyses of these surveys indicate that a frenotomy performed on a newborn reduces nipple tenderness during breastfeeding [8]. At the same time, the physiology of lactation indicates that most mothers experience nipple tenderness during the first weeks of breastfeeding, which is determined by several factors and which generally subsides in subsequent weeks of lactation [17]. When analyzing objective data on the effect of frenotomy, such as the duration of breastfeeding, the volume of milk suckling by the child from the mother, or better weight gain, there is no evidence of improvement in any of the above indicators of successful breastfeeding [18]. Despite the lack of clear diagnostic criteria for a shortened lingual frenulum and the lack of evidence for the positive health effects of frenotomy on the child, healthcare professionals dealing with breastfeeding problems commonly recommend and perform frenotomy.

Frenotomy is a surgical procedure involving the severing of the lingual frenulum. The procedure's methodology varies among centers; it can be performed with scissors, a scalpel, or even a laser. No data indicates the superiority of any one method. There is also a lack of evidence and clear recommendations regarding the optimal timing of the procedure [7].

Given current medical knowledge regarding the occurrence and treatment of pain in newborns, the phrase "in the neonatal period, the procedure does not require anesthesia" found in information brochures distributed to parents seems surprising and controversial. The pain intensity score in newborns undergoing frenotomy, as assessed by the CRIES scale, was 5.7 points [19], which requires the use of analgesia, according to the scale's authors. According to available research, pain relief is necessary during frenotomy in newborns [19]. The failure to provide pain relief to a newborn is unacceptable, harmful, and unethical.

4. Our Own Observations

The analysis included 630 cases of newborns assessed by a neuro-speech-language pathologist. These included newborns born both prematurely and at term, at University Clinical Hospital No. 4 in Lublin in 2019 and 2020. The study group represented 12% of all children born during this period. It should be emphasized that, for organizational reasons, the neuro-speech-language pathologist did not assess all newborns born during this period, whose mothers had various types of breastfeeding problems. The neuro-speech-language pathologist diagnosed various types of shortened lingual frenulum (anterior and posterior) in all assessed newborns.

In the analyzed group of patients, the majority were male newborns (ratio 1.2:1). 80% of the children were born by cesarean section, and 20% were born vaginally. In this group, 63% of the children studied during hospitalization required supplemental formula feeding for various reasons. Insufficient maternal milk supply was listed in the medical records as the most common reason for supplemental formula feeding (80%).

64% of the newborns studied were recommended for immediate frenotomy.

The remaining patients remained under the care of an outpatient neuro-speech-language pathologist, and most were also referred for frenotomy in the following weeks. Unfortunately, we do not have data on the possible health effects of the procedure.

5. Discussion

The lingual frenulum is a tissue fold connecting the lower surface of the tongue to the floor of the mouth. Ankyloglossia (tongue-tie) refers to a shortening of the lingual frenulum, which impedes

 doi:10.20944/preprints202511.0031.v1

proper tongue motor function [20,21]. There is significant individual variability in the structure of the frenulum, and the clinical presentation of ankyloglossia can vary.

However, there is no correlation between the degree of its severity and functional impairments related to tongue motor function. There is also no uniform classification of the frenulum structure or established indications for surgical intervention, such as frenotomy. It is recommended that all physiological functions related to tongue motor function (sucking, swallowing, biting, chewing, smacking, and others) be assessed during the examination of a child with ankyloglossia [21–23]. Unfortunately, there is no uniform classification of ankyloglossia, and many assessments are subjective and discretionary, making it challenging to conduct comparable studies on the incidence of ankyloglossia and to establish objective indications for frenotomy [21]. Literature data estimate the prevalence of ankyloglossia at 0.02 to 10.7%, with a predominance in boys [8,20,21], which we also observed in our own studies. Similarly, Walsh et al. observed that, compared to the entire population of patients discharged from the hospital, children with ankyloglossia or frenotomy were more likely to be male, privately insured, and from families with a higher average income [7].

Frenotomy is a surgical procedure that involves incising or cutting the frenulum, a thin band of connective tissue that restricts tongue mobility. In recent years, there has been a disturbing increase in the frequency of diagnoses of ankyloglossia, or a shortened frenulum of the tongue, and the indications for frenotomy in newborns have increased by over 800% [24]. Approximately 38% of these diagnosed newborns undergo surgical treatment [24].

In the United States, there's even been talk of a booming business called "Inside the Booming Business of Cutting Babies' Tongues" due to the frequent use of this procedure and its explicit promotion by some specialists, despite the lack of evidence-based data to justify this sudden increase in frenotomy rates [25,26]. It's also suggested that this invasive procedure often appears unnecessary and may be associated with undesirable complications, while failing to improve breastfeeding exclusivity and duration [26]. Some clinicians suggest that the impact of ankyloglossia (tongue-tie) on breastfeeding may be overestimated, and that surgical treatment of newborns remains controversial [27,28].

Literature data regarding frenotomy surgery also show that only 23% of children undergoing frenotomy achieved improvement in breastfeeding [29]. An increase in post-frenotomy complications such as pain, healing complications, vascular or nerve damage, breathing and feeding difficulties, and weight loss was observed [29]. Given the above findings, parents must always be informed of the possibility of no improvement after the procedure and of the alternative, non-surgical approach, before deciding on frenotomy. No significant difference was found between the surgical techniques used: scissors or scalpel incision, laser incision, or Z-plasty [21]. It is recommended to avoid general anesthesia and perform the procedure under local anesthesia [21].

Currently, the issue of a shortened lingual frenulum is generating much debate and controversy. The management of ankyloglossia and its impact on breastfeeding also remains controversial, with limited scientific evidence, particularly regarding the usefulness of frenotomy [30].

A cohort study conducted by Feldens [28] in a public hospital in Brazil aimed to assess the impact of ankyloglossia in newborns on mothers' self-efficacy in breastfeeding at 14 days of age. The lingual frenulum was clinically assessed and classified using the Bristol Tongue Assessment Tool. For each newborn with diagnosed or suspected ankyloglossia, two newborns without ankyloglossia were included in the study. At 14 days of age, mothers were interviewed at home to collect data on breastfeeding self-efficacy using the Breastfeeding Self-Efficacy Scale-Short Form (BSES-SF). No significant difference was found in BSEF-SF scores between children with and without ankyloglossia. The authors concluded that ankyloglossia at birth had no clinically significant impact on breastfeeding success in children at 14 days of age [27]. Similarly, the study by Dinh et al. [30] found no significant differences between frenotomy and sham procedures in terms of immediate pain relief or improved breastfeeding outcomes. These authors do not recommend tongue frenotomy for the treatment of breastfeeding difficulties in the postpartum period.

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Another multicenter cohort study found no significant difference in the prevalence of exclusive and total breastfeeding at 1, 4, and 6 months between children with defined or suspected ankyloglossia and those without [28]. Another interesting finding of these authors was the prevalence of defined and suspected ankyloglossia, which was 1% and 4.8%, respectively, for a total of 5.8% [27].

Walsh et al. observed that, compared to the entire population of patients discharged from the hospital, children with ankyloglossia or frenotomy were more likely to be male, privately insured, and from families with a higher average income [7]. It appears that in cases of breastfeeding difficulties, non-surgical treatment should first be attempted, as frenotomy does not always resolve breastfeeding difficulties and may cause complications [24].

6. Conclusions

The data in available publications question the validity of subjecting newborns to the invasive, painful surgical procedure of frenotomy. Numerous studies are being conducted to assess the validity of this procedure. It is undoubtedly necessary to monitor patients for significant symptoms associated with a diagnosis of a shortened lingual frenulum in the neonatal period before referring the child for frenotomy.

Based on available medical data, there is no evidence of beneficial health effects for the child resulting from early frenotomy, at least not to the extent currently used [18]. Further research is needed on the biomechanics and identification of factors influencing proper breastfeeding by newborns, as well as a reliable assessment of the role of the lingual frenulum in breastfeeding. It is also essential to collect data on frenotomy procedures to assess the safety of this procedure, as well as to conduct studies assessing the health effects of this procedure in newborns.

Frenotomy does not always resolve breastfeeding problems. Therefore, frenotomy should only be performed in justified cases, when there is no improvement in breastfeeding success after counseling and eliminating other causes through conservative management. Furthermore, it should be emphasized that frenotomy may be associated with complications such as pain, healing complications, vascular or nerve damage, scarring, breathing difficulties, and even feeding difficulties, choking, weight loss, or delays in the diagnosis of other pathologies. It should be emphasized that before the decision about frenotomy is made, parents must be informed about the possibility of a lack of improvement after the procedure and about the alternative of non-surgical treatment.

It seems that collaboration and assessment by a team of specialists (neuro-speech therapists, otolaryngologists, pediatricians, neonatologists, orthodontists, lactation consultants, and physiotherapists) is essential to reduce the incidence of unnecessary frenotomy.

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