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Ankyloglossia: Diagnosis, Challenges, the Role of Frenotomy in Treatment – A Comprehensive Literature Review

Maja Weimann
Internship at Copernicus Hospital, Nowe Ogrody 1/6, 80-803, Gdańsk, Poland
majaweimann.priv@gmail.com
https://orcid.org/0009-0004-1375-1337

Marta Janura
Jan Mikulicz-Radecki University Hospital, Wrocław, Poland
martajanura@gmail.com
https://orcid.org/0009-0004-4606-009X

Martyna Byrska
Jan Mikulicz-Radecki University Hospital, Wrocław, Poland
martyna.helena.byrska@gmail.com
https://orcid.org/0009-0008-1494-3903

Hanna Barska-Kobylińska
Healthcare Complex in Nysa, Bohaterów Warszawy 34, 48-300 Nysa, Poland
hannabarskakobylinska@gmail.com
https://orcid.org/0009-0004-7017-4812

Grzegorz Przywara
Faculty of Medicine, Wroclaw Medical University, Wrocław, Poland
grzegorz.przywara@student.umw.edu.pl
https://orcid.org/0009-0004-4710-0663

Oliwia Biegańska
Faculty of Medicine, Wroclaw Medical University, Wrocław, Poland
oliwia.bieganska@student.umw.edu.pl
https://orcid.org/0009-0006-0442-6211

Emilia Biczak
Faculty of Medicine, Wroclaw Medical University, Wrocław, Poland
emilia.biczak@student.umw.edu.pl
https://orcid.org/0009-0003-7357-1832

Abstract:

Ankyloglossia is a congenital condition where a shortened sublingual frenulum restricts tongue movement, leading to breastfeeding challenges like nipple pain, poor latch, insufficient milk transfer, low infant weight gain and obstructive sleep apnea syndrome (OSAS) in children. Breastfeeding during the early months of a child's life is essential for healthy development. For this reason, researchers have studied ankyloglossia to explore the impact of frenotomy on improving the well-being of children with a shortened lingual frenulum. Based on a studies full lingual frenetomy is an effective intervention for improving speech, feeding, and sleep in children with restricted tongue mobility

Key words: ankyloglossia, frenotomy, frenecotmy, breastfeeding, maternal nipple pain, proper latch, insufficient weight gain, obstructive sleep apnea syndrome.

1. **Introduction**:

Ankyloglossia, commonly known as tongue-tie, is a congenital condition characterized by an anatomical variation in the sublingual frenulum that can restrict normal tongue movement. This condition often leads to challenges with breastfeeding, such as maternal nipple pain, difficulty achieving a proper latch, swallowing problems, inadequate milk transfer, and insufficient weight gain in the infant. Limited tongue mobility can result in orthodontic complications, such as misalignment of teeth, open bite, and gaps between the mandibular incisors.[1-5] Shortened

lingual frenulum may play a role in the development of OSAS (Obstructive sleep apnea syndrome) in children, as the tongue, a key muscle group in the upper airway, is essential for maintaining oropharyngeal openness.[6]

Research [7,8] has demonstrated that children with tongue-tie had a shorter breastfeeding duration than those with normal lingual frenulum, despite the fact that many newborns with ankyloglossia breastfeed without any problems.

2. **Ankyloglossia and problem with breastfeeding**

The medical community, as well as parents, recognizes the critical importance of breastfeeding for the future health and well-being of the young individual. WHO and UNICEF's global guidelines for optimal infant nutrition, outlined in the Global Strategy, recommend exclusive breastfeeding for the first 6 months (180 days), followed by the introduction of safe and nutritionally adequate complementary foods at 6 months, alongside continued breastfeeding until at least 2 years of age or longer. Breastfeeding provides significant short- and long-term benefits for both the child and the mother, offering protection against various acute and chronic conditions. The long-term negative effects of not breastfeeding are becoming increasingly acknowledged as critical.[9]

In the WHO European Region, breastfeeding practices, especially exclusive breastfeeding rates, do not align with WHO recommendations. Significant disparities in breastfeeding practices are observed across countries.[10] The reasons mothers provide for stopping breastfeeding within the first year have been found to differ depending on the child's age at the time of weaning. One of the seven factors for which mothers stopped breastfeeding is trouble with sucking or latching on. This issue emerged as a primary concern among mothers of newborns during the first month of life.[11]

3. **Prevalence and examination of ankyloglossia**

About 4–5% of the general population suffers from tongue tie. It is more common in males than females and is inherited autosomally dominantly through the X chromosome. The phenotypic spectrum includes complete ankyloglossia, in which the ventral portion of the tongue merges with the floor of the mouth, and the lack of clinical importance.[12]

During a clinical examination of an infant's oral cavity, it is crucial to evaluate the tongue's mobility and function while the infant is calm. This includes assessing the tongue's shape and movement by allowing the infant to suck on the examiner's gloved finger. Furthermore, it is important to palpate the lingual frenulum to evaluate its elasticity and tissue length, as well as to examine the placement of its attachments.[13] Another aspect to consider is the tongue's resting position. Typically, in a relaxed state, the tongue rests against the hard palate. However, infants with tongue-tie often keep their tongues on the floor of the mouth. Similarly, when crying, their tongues tend to remain on the floor of the mouth and may lift only at the tip or along the side edges. In contrast, if there is no restriction in tongue mobility, the tongue can lift straight upward.[14,15]

There are three methods for assessing the lingual frenulum: direct measurement, Kotlow's free tongue measurement, and tongue mobility evaluation. Given the technological difficulties and increased likelihood of measurement errors, studies have indicated that direct frenulum measurement which measures the distance between the tongue and the lingual frenulum insertion is less accurate than tongue mobility evaluation.[16,17]

4. Treatment possibilities

Frenotomy is the first- line treatment option for patients presenting ankyloglossia. [18,19] The frenotomy procedure begins with lifting the tongue to expose the frenulum using a grooved director. The tissue is then clamped to provide anesthesia and cut with scissors. If necessary, fingertip pressure is applied to control bleeding.[20] Another option is a frenectomy, which involves the removal of the frenum using a CO2 laser. The procedure begins with a horizontal incision at the center of the frenum, progressing until all fascia and mucosal tension are fully released. Minimal or no bleeding was observed in all cases, with an average laser application time of 15 seconds. Acetaminophen or ibuprofen can be used to treat discomfort following surgery. Parents should be advised to manually stretch the child's wound two to three times daily, as tolerated. Myofunctional exercises were suggested if the youngster could obey instructions.[21]

5. **Literature review**

Multiple studies were conducted to assess the impact of frenotomy on issues related to ankyloglossia. Using PubMed, we searched for articles with the keywords "ankyloglossia" and "frenotomy" to identify publications that met our criteria.

The effectiveness of frenotomy for infant ankyloglossia was the subject of the Buryk study.[20] Determining if frenotomy for newborns with ankyloglossia enhanced nursing capacity and maternal nipple pain was the main goal. Evaluating if frenotomy lengthened the nursing period was a secondary goal. A single-blind, randomized controlled clinical trial was conducted to assess the effectiveness of frenotomy in treating infant ankyloglossia. In order to grade ankyloglossia, examiners used the HATLFF system that evaluates seven functional items (e.g., tongue lateralization and lift) and five appearance items (e.g., tongue appearance and frenulum elasticity), scored from 0 to 2, with a maximum of 10 points for appearance and 14 for function. When it comes to recommending a frenotomy, the HATLFF is incredibly reliable.[20] A frenotomy is recommended for infants scoring either above 11 points in function with failed lactation management or below 8 points in appearance, indicating significant ankyloglossia.[22]

In the Burky study subjects were randomly assigned to the frenotomy or sham group using a computerized random-number generator. The parents were blinded to the group in which their infant was enrolled. Infants in the frenotomy group underwent the procedure, while those in the sham group stayed in the treatment room for the same duration (5 minutes) without the procedure being performed. After the procedure or sham, the infant was returned to the mother for immediate breastfeeding. For the effectiveness of the study, mothers were asked not to look into the baby's mouth. The SF-MPQ and IBFAT assessments were completed before breastfeeding and repeated after breastfeeding following the procedure, after which mothers were informed of their group assignment. A 2-week follow-up in the ENT clinic included HATLFF, SF-MPQ, and IBFAT assessments. Importantly, infants in the sham group were offered the frenotomy procedure before the 2-week follow-up if parents did not notice any improvement in feeding.

Mothers assessed nipple pain using the Short-Form McGill Pain Questionnaire (SF-MPQ), which includes 3 sections: 15 sensory and affective pain descriptors (0-4 scale), a visual analog scale and a present pain intensity measure (0-5 scale). The most severe pain is represented by a maximum score of 50, which is the result of these measurements taken together. The SF-MPQ integrates sensory and affective pain assessments with a visual analog scale and a measure of current pain intensity, both of which are recognized as valid and reliable.[23] Both frenotomy and sham groups showed significant decreases in SF-MPQ scores post-intervention, with greater improvement in the frenotomy group. SF-MPQ scores dropped from 16.77 to 4.9 in the frenotomy group and from 19.25 to 13.5 in the sham group, with an effect size of 0.38.

The Infant Breastfeeding Assessment Tool (IBFAT), completed by the mother in the Burky Study, has 4 response categories scored from 0 to 3 with a maximum score of 15. The IBFAT shows excellent reliability. Higher scores correlate with improved breastfeeding competence, fewer issues, greater maternal satisfaction, and increased milk volumes and intake rates.[24-25] IBFAT scores improved more in the frenotomy group (9.3 to 11.6) compared to the minimal change in the sham group (8.48 to 8.07), with an effect size of 0.31.

A.H. Messner conducted a study evaluating the impact of ankyloglossia on breastfeeding difficulties in comparison to a control group of newborns without the condition.[26] 83% of infants with ankyloglossia were breastfed for at least 2 months, compared to 92% in the control group (statistically non-significant difference, $P = 0.29$). Feeding difficulties (nipple pain lasting more than 6 weeks or difficulty latching onto the breast) were reported by 25% of mothers of infants with ankyloglossia, compared to 3% in the control group ($P < 0.01$). A thick frenulum was more strongly associated with feeding difficulties than a thin one ($P = 0.02$). In most infants, ankyloglossia does not cause breastfeeding problems, but in a minority of cases, it can lead to difficulties such as nipple pain or issues with latching onto the breast.

The Camañes-Gonzalvo study aimed to assess the relationship between a short lingual frenulum (ankyloglossia) and the risk of obstructive sleep apnea syndrome (OSAS) in children.[27] It specifically focused on the impact of a short frenulum on the development of the upper airways and the risk of airway collapse during sleep. In order to answer the PICO (Population/Patient, Intervention, Comparison, Outcome) issue, the

main goal was to determine whether ankyloglossia and children's sleep-disordered breathing are related. The study included children aged 4–17 years with sleep-disordered breathing (SDB) diagnosed through polysomnography. The control group consisted of children without a short lingual frenulum.

According to estimates, between 1 and 4% of children are affected by OSAS, with the maximum incidence happening between the ages of 2 and 8.[28] A short lingual frenulum increased the risk of OSAS by more than threefold. It also restricted tongue mobility, influencing the development of abnormal craniofacial morphology. Because ankyloglossia limits the tongue's superior movement, it is difficult to form a lip seal when swallowing, which leads to tongue pushing. This problem causes an absence of nasal breathing (ascribed to anatomical and muscle tone failure) and improper stimulation of bone growth, which leads to the secondary development of mouth breathing.

The Baxter prospective cohort study [21] sought to evaluate the impact of full lingual frenectomy on speech, feeding, and sleep quality in children with restricted tongue mobility due to ankyloglossia. The effects of the procedure were analyzed in combination with myofunctional exercises.

Surveys were used, completed by participants before the procedure, one week after, and one month later. The surveys included questions on speaking new words, eating duration, sleep quality, and subjective symptom improvement.

The study included 37 children aged 13 months to 13 years, selected due to speech, feeding, or sleep issues linked to ankyloglossia. The Kotlow rating scale assessed the degree of restriction, and frenum removal was performed using a CO2 laser. According to the study, significant improvements were observed in children who underwent a full lingual frenectomy performed with a CO2 laser. Regarding speech, 89% of children showed progress, speaking more new words, mumbling less, and being better understood by parents and others. In feeding, 83% improved, eating more efficiently, encountering fewer issues with food textures, and choking less frequently. Sleep quality also enhanced for 83% of participants, with deeper sleep, fewer apneas, reduced snoring,

and more frequent mouth closure during sleep.

The study of Tae Hoon Kim [29] compared the effectiveness of two surgical methods: simple frenotomy and the more complex 4-flap Z-frenuloplasty, in improving articulation in preschool-aged children. The study included 37 children aged 3 to 7 years with confirmed articulation difficulties and ankyloglossia (Kotlow classification, classes I–III). Both groups showed significant improvement in articulation test results, particularly in consonants. No significant differences in effectiveness were observed between the two surgical methods. Z-frenuloplasty appeared more advantageous in cases of severe ankyloglossia but required a more complex surgical process.

The study conducted by Alan Emond [30] rigorously evaluated the effectiveness of early frenotomy in term infants with mild to moderate tongue-tie presenting breastfeeding challenges. Participants were randomly assigned to one of two groups: an immediate frenotomy group (55 infants) or a standard care group (52 infants). Standard care consisted of routine breastfeeding support provided by experienced lactation consultants and midwives. 35 of 52 mothers in the standard care group requested frenotomy after 5 days due to persistent breastfeeding difficulties. By 8 weeks, only 6 mothers in this group continued breastfeeding without frenotomy. Mothers who underwent frenotomy reported immediate improvements in latch effectiveness and reduced nipple pain. Many noted a noticeable difference during the first post-procedure feeding. This study reinforced the importance of individualized approaches to tongue-tie management, the need for robust lactation support, and the potential benefits of frenotomy for selected cases of breastfeeding difficulty

6. Discussion

According to research, breastfeeding improves a child's cognitive development and reduces the risk of infections, diabetes, obesity, and other health problems. For this reason, the American Academy of Pediatrics recommends breastfeeding for the first year of life.[31]

In the Buryk Study the incidence of ankyloglossia was 1.9%, which is lower than the 3.2% to 10.7% reported in the literature.[26,32] According to the Buryk Study the lower incidence is

because authors not evaluate all infants with ankyloglossia but rather only those who met entry criteria.

The true prevalence of ankyloglossia remains unclear, as it can differ depending on the assessment method used. It is observed more frequently in infants compared to children and adolescents.[33]

The average age at frenotomy in the Buryk research was 6.7 days. As a result, moms had time to exhibit ongoing feeding issues in spite of lactation therapies, and these infants had time to develop nursing routines. Because some infants with ankyloglossia won't have nursing issues, there might be a benefit to giving them more time to get used to breastfeeding before having a frenotomy.[20] The Buryk study suggests that while there is no definitive ideal timing for frenotomy, it is recommended to perform the procedure between two and six days after birth.

Neonatal frenotomy should be considered for selected infants whose mothers report significant feeding difficulties. This procedure is simple, quick and rarely results in complications.[26] Based on the reviewed studies, the authors concluded that frenotomy for clinically significant ankyloglossia results in a significant and immediate decrease in maternal nipple pain and an improvement in infant breastfeeding effectiveness.

Mothers who underwent frenotomy reported immediate improvements in latch effectiveness and reduced nipple pain. Many noted a noticeable difference during the first post-procedure feeding. It is essential to prioritize individualized approaches to tongue-tie management, ensuring that each case is addressed based on its specific characteristics and needs. Equally important is the provision of robust lactation support to help mothers navigate breastfeeding challenges effectively.[34]

There is a significant association between ankyloglossia and OSAS in children.
Assessment of a short frenulum should not be conducted in isolation. Additional factors, such as tongue mobility and a high-arched palate, must also be analyzed. Interventions such as frenotomy and myofunctional therapy may play a crucial role in reducing the risk of OSAS in children.[24] After frenotomy, the tongue rests on the palate instead of the lower jaw. Younger children instinctively adjusted, while older ones required myofunctional exercises. Compliance was challenging for younger children, but speech therapists specializing in oral motor exercises were helpful for those under four.[35]

Based on a study by The Baxter's full lingual frenectomy is an effective intervention for improving speech, feeding, and sleep in children with restricted tongue mobility. The best results were achieved when the procedure was combined with myofunctional therapy. Healthcare providers are encouraged to assess tongue mobility in children presenting with symptoms across multiple areas and to consider frenectomy when restrictions are present.[35]

There is not enough evidence that myofunctional therapy is effective on its own.[36] According to Miranda study, myofunctional therapy may serve as the sole treatment option in certain instances and, when surgery is required, it plays a crucial complementary role in enhancing the overall treatment outcome.[37]

7. **Conclusions**

The analysis of ankyloglossia and the impact of frenotomy on enhancing the well-being of both mother and child is a crucial area of research, particularly in the context of early developmental processes. This topic remains highly relevant and continues to be a focus of active investigation by researchers. Moreover, future studies should aim to standardize diagnostic criteria and treatment protocols while exploring the long-term outcomes of frenotomy. Emphasizing interdisciplinary approaches, such as combining surgical intervention with myofunctional therapy, could further optimize treatment efficacy and improve quality of life for affected individuals.

Conceptualization: Maja Weimann, Marta Janura, Martyna Byrska, Hanna Barska-Kobylińska,

Methodology: Maja Weimann, Marta Janura, Martyna Byrska, Hanna Barska-Kobylińska

Check: Maja Weimann, Marta Janura, Martyna Byrska, Hanna Barska-Kobylińska, Emilia Biczak, Grzegorz Przywara, Oliwia Biegańska

Investigation: Maja Weimann, Marta Janura, Emilia Biczak, Grzegorz Przywara

Writing -rough preparation: Maja Weimann, Marta Janura, Martyna Byrska, Hanna Barska-Kobylińska, Emilia Biczak, Grzegorz Przywara, Oliwia Biegańska

Writing -review and editing: Maja Weimann, Marta Janura, Martyna Byrska, Hanna Barska-Kobylińska, Emilia Biczak, Grzegorz Przywara, Oliwia Biegańska

Supervision: Maja Weimann, Martyna Byrska, Marta Janura, Hanna Barska-Kobylińska

Project administration: Maja Weimann, Marta Janura, Martyna Byrska, Hanna Barska-Kobylińska

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