REVIEW ARTICLE

# Relationship between Breastfeeding Difficulties, Ankyloglossia, and Frenotomy: A Literature Review

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## Abstract

**Objective:** Breastfeeding plays a key role in the development of the baby, in addition to the benefits to the mother and this dyad. Among the possible difficulties in this process, we have ankyloglossia. Some professionals opt for the frenotomy, although the literature is controversial. This paper aims to present how the literature provides subsidies for health professionals' decision and action in the intersection of the themes: breastfeeding, ankyloglossia, and frenotomy.

**Materials and methods:** The research on the platforms SciELO and PubMed used the terms: "ankyloglossia," "frenotomy," and "lingual frenulum" and the same ones associated with "breastfeeding." A specific inclusion and exclusion criteria were applied and validated by the American Speech-Language-Hearing Association to reduce any bias in the analysis. In the end, 16 papers were included and, by thematic equivalence, divided into two domains: association between lingual frenulum alteration and breastfeeding and between frenotomy and breastfeeding.

**Results:** The literature does not assure that the frenotomy is the "standard conduct" to be adopted in cases of difficulty in breastfeeding and ankyloglossia.

**Conclusion:** Further studies are needed on the different types of ankyloglossia and their direct influence on the sucking function and lactation difficulties.

**Keywords:** Ankyloglossia, Breastfeeding, Frenotomy.

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## Introduction

According to the World Health Organization (WHO),[1] breast milk is the "other standard" food for all newborns and should be the exclusive source of nutrition for the first 6 months of life. It is known that breastfeeding is a process that involves an intimate connection between mother and child, playing a fundamental role in the baby's nutritional development, in addition to promoting their physiological, immunological, cognitive, emotional development and also providing benefits in the mother`s physical and mental health.[2] Therefore, breastfeeding has a major impact on the promotion of comprehensive health in the mother–baby dyad.[2]

At first, every newborn, without anatomical and physiological changes, has conditions and skills for breast sucking,[1] but this process can go through several difficulties and challenges to overcome, such as the mother's lack of experience and knowledge, the anatomy of the breast, nipple pain and fissure, the grip, the baby's posture, fatigue, among others.[3–5] Knowing these issues, one should also be aware of the newborn's mouth anatomy, since any change can provide or enhance these difficulties.[6,7] Among these changes, the literature has cited ankyloglossia as a complicating agent.[8–13]

Ankyloglossia, or "tongue-tie," is a congenital condition, characterized by the abnormal development of the lingual frenulum, which is shortened and/or thick.[14,15] Depending on the complexity of the case, there is interference in the free movement of the tongue, which can lead to complications in the development of the oral cavity and the functions of swallowing, speech, and sucking.[15–19]

For the extraction of breast milk, while the child is breastfeeding, there is simultaneous coordination of oral reflexes, lip sealing, in addition to protruding tongue movements.[20] Therefore, if there is any restriction or difficulty in the activity of the language, this process can be hampered at several levels[10,13,19] and can often lead to early weaning.[19,21,22]

In view of this scenario, many newborns diagnosed with ankyloglossia are submitted to the surgical procedure of frenotomy[23,24] considering that many authors defend the easing and/or resolution of breastfeeding difficulties after the division of the lingual frenulum.[5,25,26] However, the performance of this surgical procedure is still controversial in the literature, where little is discussed about the indication of techniques and their consequences,[16] in addition to some authors considering the strength of the evidence of the benefits of frenotomy to be low.[23,26–28] Others still argue that the indication should be cautious, at the right time and/or, still, only in cases of "symptomatic ankyloglossia," that is, when there is functional interference.[7]

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Thus, although many professionals support the release of the lingual frenulum early as an aid to the breastfeeding process,[29–33] much has been questioned about how this procedure has become routine.[16,27,32,33] Such disparity also occurs because there is a great variation in the way to classify the alterations of the lingual frenulum,[23,24] and the same case can be diagnosed as normal or altered, depending on the criteria of the health professional.[24,34] It is known that although several standards and systems have been developed to analyze and classify the severity of ankyloglossia, none has become a common or universal practice.[23,24]

Therefore, despite the frenotomy being frequently indicated by health professionals, the influences of changes in the lingual frenulum in breastfeeding are still debatable in the literature.[35,36] In addition, there is a large discrepancy in the diagnosis and treatment of ankyloglossia among otorhinolaryngologists, pediatricians, dentists, speech therapists, and breastfeeding consultants,[8,37] including the professional's lack of preparation in relation to these themes.[38]

Knowing all the benefits of breastfeeding for the mother–baby dyad, any and all circumstances that hinder the development of this process should be considered a matter of public health.[29] It is the role of the health professional to understand this breastfeeding process in its original sociocultural and family context, thus taking care of everyone involved.[39]

The present paper aims to present and discuss how the literature provides support for the decision-making and performance of health professionals in the face of the intersection of the themes: breastfeeding, ankyloglossia, and frenotomy.

## Materials and Methods

The bibliographic search was carried out between the 1st of January 2013 and the 31st of May 2020. The databases used were PubMed and SciELO.

The survey of articles was carried out using the following descriptors: (*Medical Subject Headings*—MeSH): ("*ankyloglossia*" [MeSH] OR "(*ankyloglossia*) AND *breastfeeding*" OR "*frenotomy*" [MeSH] OR "(*frenotomy*) AND *breastfeeding*" OR "*lingual frenulum*" [MeSH] OR "(*lingual frenulum*) AND *breastfeeding*" OR "*tongue-tie*" [MeSH] OR "(*tongue-tie*) AND *breastfeeding*").

Initially, specific search filters were used, a feature found in the PubMed database, among them: full text availability (*Text availability: "Full text"*); research in humans (*Species: "Humans"*); Portuguese and English languages (Language: "*English*, *Portuguese*"); ages: children from birth to 18 years; newborns from birth to 1 month of life; infant from birth to 23 months; infant from 1 month to 23 months (*Ages: "Child: birth–18 years; Newborn: birth–1 month; Infant: birth–23 months; Infant: 1–23 months"*). In the SciELO database, these initial selection criteria were performed manually by the researchers. On both platforms, articles published in the previously determined period were selected.

Following the classification proposed by the *American Speech-Language-Hearing Association*,[40] as used by other authors,[41] it was decided to use the evidence level of the articles as a way of selection. Evidence levels 1b (high quality randomized controlled trials), 2b (high quality nonrandomized controlled trials) were included in the present literature review and 3b (cohort studies or low quality randomized controlled trials). Levels 4 (clinical outcome studies: case studies), 5b (case-control studies), 6 (case series), and 7 (expert opinion without evident clinical evaluation) were excluded. Since this is a review in order to score the findings of the literature on the themes in the given period, all types of systematic reviews that already exist were excluded (levels 1a, 2a, 3a, and 5a).

Finally, studies whose sample included syndromic individuals with some type of paralysis, disease, and nasolabial and/or cleft palate were excluded, in addition to studies with an emphasis on various surgical techniques, creation and/or validation of protocols and/or didactic material.

Two researchers first reviewed the abstracts of all selected articles independently to assess their eligibility according to previously determined criteria. Subsequently, a new review was carried out by both researchers together to confirm the excluded articles, and only then were the selected articles read in full to fully confirm their inclusion and extract the necessary data.

## Results

The initial research, according to the selection criteria, identified a total of 552 articles. After excluding duplicate publications due to the occurrence of common descriptors, this number had been reduced to 140 articles. Only two were not available for *download* (*closed access*), resulting in 138 articles evaluated by the elected exclusion criteria.

After evaluating the title and abstract, 21 articles from clinical outcome studies (case studies) (evidence level 4), 25 papers with an emphasis on diverse surgical techniques, creation and/or validation of protocols, 12 articles for not fitting according to the age selection criteria, 23 systematic reviews (evidence levels 1a, 2a, and 3a), 12 opinions/expert cards (evidence level 7), 18 studies with syndromic individuals, some type of paralysis, disease and nasolabial cleft and/or palatal, 6 editorials, 2 case series (level of evidence 6), 1 work on the development of didactic material, 2 studies not in humans.

With the exclusion of 122 articles, a final number of 16 papers were included in this systematic review (Fig. 1). By thematic equivalence, these articles were divided into two domains: 1—association between alteration of the lingual frenulum and breastfeeding (Table 1 and 2) and 2—association between frenotomy and breastfeeding (Tables 3 and 4).

From the analysis of the seven articles in domain 1, it was observed that the age-group studied varied between 0 and 72 months at the beginning of the research, and in one article, this information was not described. When the participants were monitored (two studies), it was 14 days in both, and in the others, the analysis was punctual or retrospective with medical records.

In six cases, studies were conducted directly with mothers and babies. In these situations, the total number of participants was between 100 and 497.

Three studies used a common protocol for the evaluation of the lingual frenulum "protocol for the evaluation of the tongue frenulum in babies" by Martinelli et al.,[10]—"lingual test," and in the others, there was variation between the methods chosen, to obtain. In general, the parameters analyzed were opinion of the breastfeeding women about the breastfeeding experience and initial expectations, challenges, consultations, frenotomy and quality of breastfeeding, clinical examination and anatomical classification of the lingual frenulum and breastfeeding assessment.

In the nine articles in Domain 2, on the other hand, an age range between 0 and 9 months was observed at the beginning of the research, and in two articles, there was no specification. In all cases, the individuals studied were monitored, varying between two and 24 weeks. In total, the studies were carried out directly with mothers

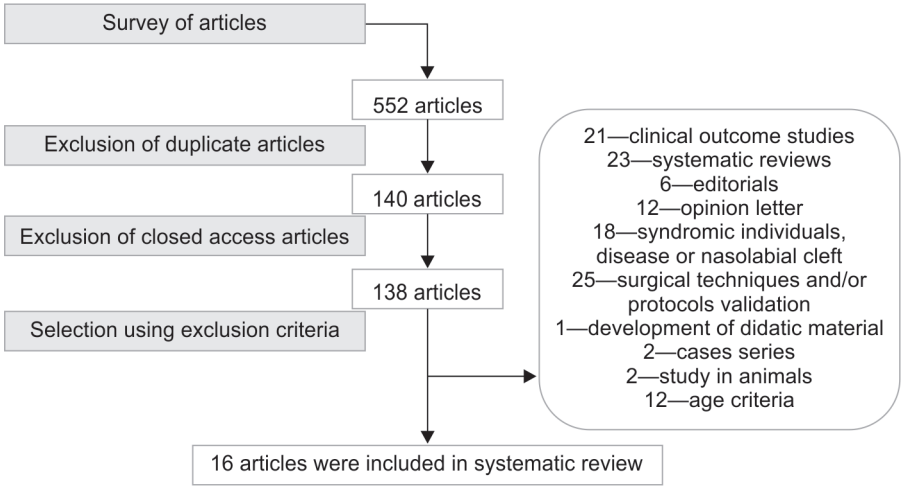


**Fig. 1:** Survey and selection of scientific articles flowchart

**Table 1:** Characterization of the studies of association between lingual frenulum alteration and breastfeeding in terms of type, number of participants, age of infants at the beginning of the study, and follow-up time

| *Authors* | *Type of study* | *Number of participants* | *Infant age at the beginning of the research* | *Follow-up time* |
|---|---|---|---|---|
| Riskin et al.[43] | Unicentric observational | 183 mothers of babies with ankyloglossia<br>314 mothers of babies without ankyloglossia (control) | 1–6 years | No follow-up |
| Haham et al.[7] | Prospective series cohort | 200 infants | 0–3 days | 14 days |
| Pransky et al.[4] | Retrospective review of patient data | 618 | Retrospective review of patient data.<br>Information not provided | Retrospective review of patient data.<br>Information not provided |
| Marcione et al.[5] | Cross-sectional, observational, analytical, with a quantitative approach. | 165 infants | 1–4 months | No follow-up |
| Fujinaga et al.[23] | Cross-sectional exploratory description | 139 dyads | Newborns with more than 15 hours of life | No follow-up |
| Campanha et al.[13] | Cross-sectional study | 130 dyads | 1–5 days | No follow-up |
| Walker et al.[24] | Prospective cohort | 100 dyads | 2 days | 14 days |

and babies involving between 43 and 656 participants, who in some cases were divided and studied as a mother–baby binomial.

Four studies used a common scale, *LATCH scale (Latch/Pega, audible swallowing/audible swallowing*, *nipple type/nipple type, comfort/comfort, hold*/positioning, help from others to keep the child on the chest—Jensen et al.[42]), for the evaluation of the variables involved in the breastfeeding process, and in the others, there was variation between the methods chosen to obtain the data, it was observed that, in general, the parameters analyzed before and after the execution of the frenotomy were quality breastfeeding according to several factors involved and also by the report of the lactating women, clinical examination, and anatomical classification of the lingual frenulum and breastfeeding assessment.

## Discussion

Breastfeeding is a natural and instinctive process in the evolution of humanity whose benefits for the mother–baby dyad have been evidenced with scientific progress,[2] in such a way that the intimate connection of this process to the child's developmental development in terms of nutritional, physiological, immunological, cognitive, and emotional aspects is unanimously consolidated, in addition to providing numerous gains in the mother's physical and mental health.[2,13] Therefore, if breastfeeding is considered to be the "gold standard" for the baby's quality of life[1] and if research is able to highlight, clarify, and strengthen the issues involved in this process, it is expected that these will be frequent and coherent in the scientific community. However, this expected scenario diverged from that demonstrated by the present study in which only 16 articles published in the proposed analysis period could be included, following the chosen criteria and theme.

The importance of breastfeeding is well known, as well as the innumerable barriers inherent to this process[3–5,13] being ankyloglossia cited as one of the complicating factors.[8–12] For Campanha et al.[13] in addition to being aware of the benefits of breastfeeding for the baby and its mother, every health professional who assists the mother–baby dyad must be aware of the prevention and management of the main problems that may occur during breastfeeding, aiming at prevention of early weaning; however, they are often unprepared,[38] as well as, great divergence in diagnoses

**Table 2:** Characterization of the studies of association between lingual frenulum alteration and breastfeeding in terms of evaluated parameters, results, and conclusion

| *Authors* | *Evaluated parameters* | *Results* | *Conclusion* |
|---|---|---|---|
| Riskin et al.[43] | Opinion of lactating women (questionnaire) on:<br>• Breastfeeding experience,<br>• Challenges,<br>• Breastfeeding consultations,<br>• Frenotomy (when performed) | • The overall rates of breastfeeding problems in the first month were similar.<br>• Breastfeeding rates at 6 months were similar.<br>• Mothers of babies with ankyloglossia: reported significantly more problems with latching, prolonged breastfeeding, and exhaustion of the child during feedings; complemented breastfeeding more frequently with expressed breast milk; needed consultation more frequently and a significantly higher proportion reported that the consultation helped.<br>• 87% of mothers were aware of their children's ankyloglossia, but only 50% associated it with breastfeeding problems.<br>• The possibility of frenotomy was mentioned in 69% of mothers and was performed in 35% of cases.<br>• Satisfaction with the procedure was generally poor, except when done to resolve breastfeeding problems. | • Tongue-tie infants had significantly more breastfeeding problems in the first month,<br>• Early diagnosis and lactation consultation may assist mother–infant dyads substantially<br>• More frequently, mothers whose infants underwent frenotomies for breastfeeding found the procedure alleviated breastfeeding problems. |
| Haham et al.[7] | • Classification of the frenulum according to Coryllos<br>• Visual examination of the tongue anatomy<br>• Opinion of the breastfeeding mother regarding the quality of breastfeeding | • All babies, with the exception of one ($n = 199$), had an observable or palpable lingual frenulum.<br>• There was no statistical correlation between the type of frenulum according to Coryllos and the presence of difficulties in breastfeeding. | The lingual frenulum insertion point and Coryllos classification are not correlated with breastfeeding difficulties. |
| Pransky et al.[4] | • Presence of ankyloglossia and classification as anterior subtypes (types I and II) or posterior subtypes (types III and IV)<br>• Presence of upper lip frenulum<br>• Difficulties in breastfeeding | • 290 (47%) had anterior ankyloglossia.<br>• 120 (19%) had posterior ankyloglossia.<br>• 14 (2%) had a upper lip tie.<br>• Some patients had anterior ankyloglossia and upper lip tie (6%) or posterior ankyloglossia and upper lip tie (5%).<br>• In case of anterior ankyloglossia, 78% reported some degree of improvement in breastfeeding after frenotomy.<br>• In case of posterior ankyloglossia, 91% reported some degree of improvement in breastfeeding after frenotomy.<br>• The release of the upper lip also improved breastfeeding (100%). | Anterior and posterior ankyloglossia and upper lip tie are abnormalities of the oral cavity that can contribute to difficulties in breastfeeding in some cases |
| Marcione et al.[5] | Frenulum thickness and insertion based on the "lingual frenulum protocol with scores for infants" | • From 165 babies, 104 were normal and 61 altered.<br>• Among the normal frenulum were prevalent those with the attachment in the middle third and visible from the sublingual caruncles.<br>• Among the altered frenulum were more frequent those with attachment between the middle third and the apex and visible from inferior alveolar crest.<br>• Thin thickness was predominant.<br>• Among the babies with altered frenulum, 24 had altered suction, and of the babies with normal frenulum, 18 had altered suction | • The lingual frenulum was predominant in normal and thin thickness.<br>• Altered frenulum was prevalent in males.<br>• Babies with altered lingual frenulum showed more change of alteration in suction, although the correlation between frenulum and suction was low. |
| Fujinaga et al.[23] | • Anatomofunctional evaluation of the tongue frenulum ("Evaluation protocol of the lingual frenum for infants")<br>• Evaluation of breastfeeding (UNICEF breastfeeding observation protocol) | • From 139 infants, only one infant was verified with a frenum alteration, equivalent to a prevalence of 0.8%.<br>• In the evaluation of breastfeeding, of the 138 binomials, whose infants did not have alteration of the lingual frenum, 82 of them (59.4%) did not demonstrate any difficulty during breastfeeding.<br>• The only infant with lingual frenum alteration did not present difficulties in breastfeeding. | There are insufficient subsidies to establish an association among lingual frenum and breastfeeding. |

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| *Authors* | *Evaluated parameters* | *Results* | *Conclusion* |
|---|---|---|---|
| Campanha et al.[13] | • Anatomofunctional assessment of the lingual frenulum - (lingual frenulum protocol for infants, LFPI)<br>• UNICEF breastfeeding observation aid<br>• Maternal complaints regarding the difficulty or not to breastfeed | • 105 (81%) newborns showed normal frenulum and 25 (19%) ankyloglossia.<br>• Of the 130, 44 did not show ankyloglossia or signs of difficulty in breastfeeding, however, all 25 newborns detected with ankyloglossia (100%) showed signs of possible difficulties in breastfeeding.<br>• 93 (72%) mothers did not report breastfeeding difficulties and 37 (28%) did so. Of the 37 mothers who reported difficulties, 12 (32%) had infants with ankyloglossia.<br>• Statistical analysis revealed an association between ankyloglossia and complaint of difficulty in breastfeeding reported by the mother. | On the first days of life, ankyloglossia is associated with the mother's breastfeeding complaint and with the newborn's sucking difficulty. |
| Walker et al.[24] | • Breastfeeding difficulties (breastfeeding assessment tool—IBFAT)<br>• Distance from the tip of the tongue to the insertion of the lingual frenulum | • Mean tip–frenulum length was 9.07 mm.<br>• A visible cord was identified in 21 subjects (21%). A palpable cord was identified in 59 subjects (59%).<br>• Visible cord and shorter tip–frenulum distance were independently predictive of higher maternal pain scores.<br>• A positive correlation was identified between tip–frenulum length and IBFAT scores for mothers with two or more previous breastfed children. | • Tongue tip–frenulum length correlated with maternal nipple pain and was useful as an objective tool for identifying newborns at risk of ankyloglossia.<br>• Maternal breastfeeding experience appears to be an important factor in the link between tongue anatomy and breastfeeding difficulty.<br>• The presence of a palpable cord was variable across examiners and should be interpreted with caution when evaluating newborns for posterior tongue-tie. |

**Table 3:** Characterization of the studies of association between frenotomy and breastfeeding in terms of type, number of participants, age of infants at the beginning of the study, and follow-up time

| *Authors* | *Type of study* | *Number of participants* | *Infant age at the beginning of the research* | *Follow-up time* |
|---|---|---|---|---|
| Emond et al.[44] | Randomized controlled trial | • 55 infants (intervention group)<br>• 52 infants (control group) | Newborns with less than 2 weeks of life | 8 weeks |
| Dollberg et al.[11] | Prospective follow-up | 264 dyads with infants undergoing lingual frenotomy due to difficulties in breastfeeding | Median of 14 days of life (1–135) | 6 months |
| Martinelli et al.[45] | Prospective longitudinal | 109 infants | 30 days | 35 days |
| Benoiton et al.[46] | Prospective audit | 43 patients | Median of 6.6 weeks (2–20) | 2 weeks |
| Ghaheri et al.[12] | Cohort prospective | 237 dyads | 0–12 weeks | 1 month |
| Billington et al.[19] | Prospective | 100 infants | Median of 17 days (2–88) | 3 months |
| Wakhanrittee et al.[47] | Prospective cross-sectional study | 328 dyads | No information | 3 months |
| Muldoon et al.[38] | Prospective before and after the cohort study | 89 mothers | No information | 1 month |
| Ghaheri et al.[48] | Prospective cohort | 54 dyads | 0–9 months | 1 month |

and conduct.[8,37] This reality is of an alarming nature due to the lack of content in the literature since it is necessary to support any and all actions in consolidated scientific evidence and there is a scarcity of research that provides such foundations.

In addition to the reduced number of studies, also analyzing the availability of information in the literature, the lack of methodological standardization appears as a complicating factor. Among the selected researches, there is a great variation in all the parameters analyzed, as well as in the tools, scales, and methods. These variations even occurred in the diagnosis and classification of ankyloglossia. Such disparities hinder the construction of discussions and conclusions based, creating biases in the literature and clinical practice.

From the selection of all articles present in this review, the presence of two themes was found that subdivided them. And for methodological and didactic purposes, these were divided into two

**Table 4:** Characterization of the studies of association between frenotomy and breastfeeding in terms of evaluated parameters, results, and conclusion

| *Authors* | *Evaluated parameters* | *Results* | *Conclusion* |
|---|---|---|---|
| Emond et al.[44] | • Degree of lingual frenulum (HATLFF abbreviated protocol)<br>• Breastfeeding quality (LATCH scale and BSES scale) | • In the initial evaluation after 5 days (LATCH score), there was no difference between the group with immediate frenotomy and the control group.<br>• Limitations improved due to the lingual frenulum after frenotomy.<br>• After the 5-day consultation, 44 from the control group requested frenotomy.<br>• After 8 weeks, only 6 (12%) were breastfeeding without frenotomy.<br>• After 8 weeks, there were no differences between groups in breastfeeding or in the child's weight. | • Early frenotomy did not result in an objective improvement in breastfeeding but was associated with improved self-efficacy.<br>• The majority of the control group opted for the intervention after 5 days. |
| Dollberg et al.[11] | • Questionnaire (LATCH scale and VAS for pain pattern) and clinical examinations to determine the anatomy of the tongue, indication for frenotomy, and condition of the infant. | • Two weeks after the frenotomy, 89% of mothers were still breastfeeding.<br>• An improvement in breastfeeding was reported by three quarters of mothers, with 3% reporting worsening.<br>• At 3 and 6 months, after the procedure, 68% and 56% of mothers were still breastfeeding, respectively. | • Lingual frenotomy does not always relieve breastfeeding difficulties and rarely gets worse.<br>• No predictor was found for successful breastfeeding after frenotomy. |
| Martinelli et al.[45] | • Changes in breastfeeding after lingual frenotomy (number of suctions, duration of pause between suction and maternal complaints). | • After frenotomy, the number of sucks increased and the pause length between sucking decreased during breastfeeding.<br>• The controls maintained the same patterns observed in the first assessment.<br>• The mothers of the 14 tongue-tied infants' opinion, at 30 days and 75 days, there was an improvement in the coordination between sucking/swallowing/breathing after lingual frenotomy. | • After lingual frenotomy, changes were observed in the breastfeeding patterns of the tongue-tied infants while the control group maintained the same patterns..<br>• All symptoms reported by mothers improved after frenotomy |
| Benoiton et al.[46] | • Questionnaires and clinical examinations to determine the anatomy of the tongue, indication for frenotomy and influence on breastfeeding. | • The most common presenting complaint was latching issues (85%) with mothers' painful nipples being the second (65%).<br>• 62% had a tongue-tie release, 29% had both a tongue-tie and upper lip tie divided, whereas 9% had an upper-lip tie alone divided.<br>• 85% of the patients who had a procedure carried out had an immediate improvement in breastfeeding, while 28 (82%) had a continued improvement at 2-week follow-up. | • Frenotomy is a simple and effective procedure for babies with continuous breastfeeding difficulties who have posterior ankyloglossia and upper lip tie.<br>• Prior assessment and postprocedure support by breastfeeding consultants are imperative in the management of these babies. |
| Ghaheri et al.[12] | • BSES-SF scale, VAS for pain severity, and the revised Children's Gastroesophageal Reflux Questionnaire.<br>• Measure of milk intake. | • The average intake of breast milk has improved 155%. | • Frenotomy results in significant improvement in breastfeeding.<br>• Improvements occur early (1 week after surgery) and continue until 1 month after surgery.<br>• Improvements were demonstrated in both infants with anterior and posterior frenulum. |
| Billington et al.[19] | • Resolution of symptoms (complete, moderate, or minimal).<br>Type of feeding (exclusive breastfeeding, complementary breastfeeding, or exclusive infant formula) | • At 3 months, the resolution of symptoms was complete—80%; moderate—15%, and minimum—5%.<br>• 49% were exclusive breastfeeding, 41% were supplementing with some formula, and 10% used only powdered milk.<br>• Of the 17 mothers who still had symptoms, 5 were exclusive breastfeeding and 8 were persisting with combined foods. | The infants who participated in the research had a higher level of exclusive or supplemented breastfeeding than the general population. |

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| *Authors* | *Evaluated parameters* | *Results* | *Conclusion* |
|---|---|---|---|
| Wakhanrittee et al.[47] | Questionnaires and mothers' reports (LATCH score) to evaluate how nipple pain, grip, and exclusive breastfeeding were evaluated before frenotomy | • The nipple pain score was significantly lower and the LATCH score was higher 24 hours and 1 week postoperatively<br>• At 3 months, the rate of exclusive breastfeeding was 66.67%. | • Frenulotomy could significantly reduce nipple pain and increase LATCH score in tongue-tied infants with breastfeeding difficulty<br>• Several factors are related to exclusive breastfeeding |
| Muldoon et al.[38] | Questionnaires and mothers' reports (LATCH score) to evaluate different breastfeeding variables before the frenotomy | • The most common reason for seeking frenotomy was the difficulty with the handle (38%).<br>• Breastfeeding consultants were the main professionals recommending frenotomy (31%).<br>• Rates of exclusive breastfeeding remained similar before and after frenotomy (58% versus 58%)<br>• Babies' tongue protrusion capacity after frenotomy was significantly greater.<br>• Almost all participants (91%) reported an overall improvement in postfrenotomy breastfeeding.<br>• Breastfeeding pain was significantly reduced after frenotomy and the overall scores on the LATCH score were significantly increased | • The study supports the hypothesis that frenotomy has a positive effect on breastfeeding variables in infants with ankyloglossia.<br>• Those findings, however, are based on a relatively small number of participants from one country only where breastfeeding rates are low.<br>• Larger studies are required to substantiate the findings |
| Ghaheri et al.[48] | • Breastfeeding effectiveness (BSES-SF)<br>• Nipple pain (VAS)<br>• Infant's gastroesophageal reflux (I-GERQ-R) | Significant improvements in postoperative scores (1 week and 1 month) | • Nipple pain, symptoms of infant reflux and the mother's self-confidence may improve after the total release of the lingual frenulum. |

*HATLFF, short form: Hazelbaker Assessment Tool for Lingual Frenulum Function; LATCH score: Latch, Audible swallowing, Nipple Type, Comfort, Hold; BSES: Breastfeeding self-efficacy scale; VAS: visual analogic scale

domains: association between alteration of the lingual frenulum and breastfeeding (1) and association between frenotomy and breastfeeding (2). It is important to note that such domains may correspond to the initial questions of professionals in the conduct and decision making when faced with cases of ankyloglossia in infants. Regardless of the methodological disparities already mentioned, the discussions developed took place in intradomain and interdomain manners.

In both domains, there was a diversity of instruments used to assess, diagnose, and classify ankyloglossia.

The multiprofessional clinical examination without the use of a standardized protocol was used to classify the presence or absence of ankyloglossia in the studies by Riskin et al.,[43] Emond et al.,[44] Martinelli et al.,[45] and Benoiton et al.[46] In the latter, ankyloglossia was also classified as: anterior, posterior, or mixed.

Four studies, Martinelli et al.,[45] Marcione et al.,[5] Fujinaga et al.,[23] and Campanha et al.,[13] adopted the "protocol of assessment of the frenulum of the tongue with scores for babies."[10]

Wakhanrittee et al.[47] and Muldoon et al.[38] classified the lingual frenulum according to its severity: mild, moderate, and severe, whereas Walker et al.[24] aimed to describe the types of frenulum and thus classify them according to the distance between the tip of the tongue and the insertion of the lingual frenulum. Such classifications were made according to the place of insertion of the frenulum in the tongue, however, it is worth noting that its thickness was not taken into account.

The absence of a standardized classification and the common use of evaluation protocols makes the correlation between the frenulum type, the degree of severity of its anatomical alteration, and its respective functional impairment somewhat subjective and inconclusive in studies in recent years. There is a significant diversity of specialties in the health field active in this theme: nurses, pediatric doctors, otorhinolaryngologists, dentists, speech therapists, among others. Without standardization, research findings no longer control variables such as the evaluator's vision, specialization, and experience, which makes the experimental quality questionable and consequently, its conclusions.

Emond et al.[44] emphasized with their findings that the need for better assessment tools since simple inspection of the lingual frenulum is not sufficient to determine which child should undergo the frenotomy procedure. These authors reinforced the importance of including observation and objective measures of breastfeeding effectiveness.

Riskin et al.[43] pointed out that infants with ankyloglossia, regardless of degree or subtype, had significantly more breastfeeding problems in the first month of life. Pransky et al.[4] corroborate this reasoning, but with a nonmandatory relationship. For these, anterior and/or posterior ankyloglossia, in some cases, can contribute to difficulties in breastfeeding. The conditional presented by Pransky et al.[4] is reinforced by the noncorrelation found between ankyloglossia, changes in suction, and breastfeeding difficulties in the studies by Haham et al.,[7] Marcione et al.,[5] and Fujinaga et al.[23] Despite not finding a relationship, Marcione et al.[5] concluded that babies with altered lingual frenulum were more likely to change suction.

Dollberg et al.[11] pointed out that they did not find a significant correlation between the type of frenulum according to Coryllos and the reasons for performing the frenotomy. Contrasting these findings, Ghaheri et al.[12] presented significant data connecting the type of ankyloglossia and problems with breastfeeding.

Such authors evidenced in their sample that 78% of infants with difficulties in breastfeeding had isolated posterior shortened lingual frenulum (class III or IV of ankyloglossia, according to Coryllos).

Campanha et al.[13] showed a 36.07 times higher probability of newborns with ankyloglossia showing signs of difficulty in sucking. Statistical analysis of Campanha et al.[13] revealed an association between ankyloglossia and complaints of difficulty in breastfeeding, in which 32% of mothers with complaints had newborns with ankyloglossia.

For Wakhanrittee et al.[47] and Muldoon et al.,[38] one of the factors associated with the failure of exclusive breastfeeding was the severity of the lingual frenulum, since the greater the severity of the lingual frenulum, the greater the limitation of the tongue and consequently the quality of the grip.

For Walker et al.,[24] the shortest distance between the tip of the tongue and the insertion of the frenulum was positively related to nipple pain and was a useful tool to identify ankyloglossia in newborns; however, they pointed out that the presence of a palpable cord was variable between examiners and should be interpreted with caution when assessing ankyloglossia in newborns.

With the exception of Ghaheri et al.,[12] Walker et al.,[24] and Campanha et al.,[13] all other studies suggested the presence of a correlation between ankyloglossia and breastfeeding but could not prove the existence of this correlation through their findings. It is believed that this is mainly due to the large amount and the lack of control over variables such as the identification and classification of the degree of anatomical impairment and, therefore, the objective diagnosis of its functional impact.

The data found in the literature of the last years analyzed by the present study demonstrate the need for further evidence regarding the relationship between the type of lingual frenulum and difficulties in breastfeeding. They also showed that there is still a need for a tool to assess the anatomical and functional conditions of the lingual frenulum that is universally adopted in order to reduce as much as possible the evaluator's bias in research, thus providing results that favor more assertive interventions based on findings with experimental quality.

The relief, according to the maternal perspective, of breastfeeding problems after frenotomy was pointed out by Riskin et al.,[43] Pransky et al.,[4] Ghaheri et al.,[48] and Campanha et al.[13] being the conduct of the frenotomy often influenced, in a decisive way, by the maternal complaint and not by an anatomophysiological diagnosis.

Management of breastfeeding is carried out by health professionals from different specialties around the world such as gynecologists, mastologists, obstetricians, pediatricians, nurses, speech therapists, physiotherapists, dentists, nutritionists, among others. The formation of these often becomes diversified, which intensifies the need to establish conducts based not only on complaints and subjective observational findings but are also dependent on the training and experience of the professional who assists the lactating-infant dyad.

In order to evidence the change from precondition and postcondition to any type of intervention, an evaluation using validated quantitative and qualitative methods is necessary, as long as they allow nonsubjective verification of the change between the evaluation moments and the magnitude of that change, whether with positive or negative effects. In the present review, it was also possible to verify the diversity of protocols and evaluation methods used to quantify the degree of impairment of the different difficulties experienced in the breastfeeding process.

The LATCH scale[42] was used by Emond et al.,[44] Dollberg et al.,[11] Wakhanrittee et al.,[47] and Muldoon et al.,[38] being the most used standardized tool in recent years.

However, it was possible to observe the use of other methodological standards. Ghaheri et al.[12] and Ghaheri et al.[48] used the abbreviated version of this same scale. Fujinaga et al.[23] and Campanha et al.[13] adopted the UNICEF breastfeeding observation protocol, while Martinelli et al.,[45] Marcione et al.,[5] and Campanha et al.[13] evaluated nonnutritive sucking and nutritive sucking according to part II of the "protocol for the evaluation of the frenulum of the tongue with scores for babies."

Emond et al.[44] evaluated the degree of the lingual frenulum using the shortened version of the Hazelbaker assessment tool for the function of the lingual frenulum as previously described by Ricke et al.[49] and Amir et al.,[50] which showed the subjectivity of the evaluations and the need for experience to use them consistently; Emond et al.[44] used *the Breastfeeding Self-Efficacy Scale*.[51]

Haham et al.,[7] Dollberg et al.,[11] Ghaheri et al.,[12] and Ghaheri et al.[48] used the classification of the frenulum according to Coryllos, while Pransky et al.[4] classified as subtypes anterior (types I and II) or posterior (types III and IV).

The visual analog scale standard of pain[52] was also used in some studies, as a protocol to assess breastfeeding difficulties.[11,12,48] Walker et al.[24] used a questionnaire on breastfeeding difficulties (*Breastfeeding Assessment Tool*).

However, the most used method to assess breastfeeding was still significantly based on the reports of lactating women, collected through structured questionnaires and/or interviews[7,43,46] and/or the classification of the infant in degrees of improvement[4,19] and such tools were not necessarily validated.

The tools that predominantly depend on the maternal vision create a great question about the subjectivity of the findings. It was noted the search of some authors for methods that eliminate this subjectivity, such as the LATCH scale and part II of the "protocol of evaluation of the frenulum of the tongue with scores for babies." However, it is necessary to carry out more research carried out by different study groups focusing on results with greater foundation and in different samples.

Both Emond et al.[44] and Dollberg et al.[11] used the same assessment tool, the LATCH scale, and both did not find a predictor for successful breastfeeding after frenotomy. In contrast to such studies, Wakhanrittee et al.[47] and Muldoon et al.[38] also used the same LATCH scale and presented data that support the positive effect of frenotomy on breastfeeding variables in children with ankyloglossia.

Benoiton et al.,[46] Ghaheri et al.,[12] Billington et al.,[19] and Ghaheri et al.[48] concluded that frenotomy is effective in babies with ankyloglossia and results in a significant improvement in breastfeeding. Ghaheri et al.[12] and Ghaheri et al.[48] characterized the improvement as being early (1 week after surgery) and that it continues until 1 month after surgery. Billington et al.[19] presented findings of complete resolution of symptoms at 3 months in 80% of cases. These data can be justified with the results of Martinelli et al.[45] who objectively demonstrated the change in the suction pattern after frenotomy. There was an increase in the number of sucks and a decrease in the pause time between them, and this finding was a differential in the literature in recent years.

The findings in the scientific literature evidenced in the present bibliographic review allow the statement that conservative conduct should be the guide in the clinical routine. That is, an infant with diagnosed ankyloglossia but without functional impairment

(weight gain and adequate suction pattern) and an infant without breast complications should not be submitted to early frenotomy. If, when diagnosed with ankyloglossia, the only impact on lactation is any breast complication in the lactating woman, one should choose to follow up on lactation management, and only if the frenotomy is not resolved should it be considered.

An infant with ankyloglossia but without functional impairment in lactation should be monitored and the frenotomy only performed when and, mainly, if there is a functional justification, such as at the moment of food insertion if there is any change in chewing and swallowing and/or at the time in which some phonetic distortion is diagnosed.

## Conclusion

The need for universal and more objective tools was evidenced, which reduce as much as possible the bias of the specialty, training, and experience of the evaluator, as well as assistance in the definition of conduct.

The literature of recent years does not guarantee that frenotomy is the "gold standard" procedure to be adopted in cases of difficulty in breastfeeding and ankyloglossia nor does it provide subsidies for this procedure to be indicated safely and accurately.

Further studies are needed on the different types of ankyloglossia and its direct influence on the suction function and breastfeeding difficulties in the lactating-infant dyad.

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