

Effect of Frenotomy on Breastfeeding in Infant with Ankyloglossia: A Rapid Review

Mustika Praja Kurniawati¹, Naninda Berliana Pratidina², Eriska Riyanti^{2*}

1. Faculty of Dentistry, Universitas Padjadjaran, Indonesia.

2. Department of Pediatric Dentistry, Faculty of Dentistry Universitas Padjadjaran, Indonesia.

Abstract

Ankyloglossia or tongue-tie is a congenital disorder characterized by a short lingual frenulum resulting in limited tongue movement, therefore causing difficulty in breastfeeding. Frenotomy is indicated if there is interference in breastfeeding. The objective of this study is to determine the effect of frenotomy on breastfeeding in infant with ankyloglossia.

Rapid review was conducted using PRISMA guidelines through Pubmed, Scopus, and Embase using keywords according to research questions based on PICO. Risk of bias was assessed using JBI Critical Appraisal Tools.

Five articles consisting of four randomized controlled trial and one quasi-experimental were included for review. Two studies reported nipple pain reduction after frenotomy, while the other two studies did not. Effectiveness of breastfeeding were found to increase in two studies, whereas one study found no increase. Two studies using different instrument to assess the effectiveness of breastfeeding did not show an increase after frenotomy. Maternal self-efficacy were found to be elevated in one study. Maternal subjective assessment reported improvement in breastfeeding in three studies. All studies reported no serious complications after frenotomy.

Frenotomy was found to be inconsistent in improving breastfeeding ability in infant with ankyloglossia. Practitioners should do case selection carefully by taking into account the assessment of tongue-tie and breastfeeding, as well as conservative approach (lactation support) before performing frenotomy.

Review (J Int Dent Med Res 2022; 15(4): 1769-1779)

Keywords: ankyloglossia, frenotomy, breastfeeding

Received date: 05 July 2022

Accept date: 14 August 2022

Introduction

Ankyloglossia or commonly known as tongue-tie is a congenital disorder characterized by a short lingual frenulum, resulting in limited tongue movement.¹ The prevalence of ankyloglossia in newborns has been reported to range from 0.2% to 10.7%.² The majority of infants have asymptomatic ankyloglossia and does not show difficulty in suckling. However, there are 25% of babies with this condition have difficulty in breastfeeding,³ This is because of changes in the latch due to alteration in tongue movement.⁴

Breastfeeding is a complex and multifactorial process, and the presence of dysfunction can

cause different symptoms and signs in both mother and baby.⁵ During breastfeeding, the restricted frenulum causes ineffectiveness of the baby in breastfeeding, inadequate transfer and intake of milk, and nipple pain. These can cause adverse effects on infant feeding such as poor weight gain and leading to early weaning.^{5,6}

Breastfeeding is the ideal standard nutrition for babies. The World Health Organization (WHO) recommends exclusive breastfeeding for the first 6 months and continued for up to 2 years with complementary foods.⁷ Breastfeeding is also believed to be one of the environmental factors that influence craniofacial growth, apart from genetic factors.⁸ During breastfeeding, the movement of muscles is activated thereby stimulating the development of stomatognathic system, while during bottle-feeding the stimulation is limited.⁹ Therefore, it is important to support mothers to breastfed their children directly, identify the obstacles that can interfere, one of which is ankyloglossia, and appropriate treatment to overcome them.

*Corresponding author:

Department of Pediatric Dentistry, Faculty of Dentistry
Universitas Padjadjaran, Indonesia.
E-mail: eriska.riyanti@unpad.ac.id

Surgical reduction of the abnormal lingual frenulum is indicated if it interferes breastfeeding.¹⁰ However, there is controversy regarding the benefits of frenotomy in reducing breastfeeding difficulties. Srinivasan et al.¹¹ reported that frenotomy can increase breastfeeding and reduce nipple pain in short term. In addition, the long-term effects of frenotomy were also reported by Ramoser et al.¹² which include an increase in attachment during breastfeeding, an increase in tongue mobility with reduced milk coming out of the tip of the mouth, and an increase in body weight. Nevertheless, an RCT study conducted by Emond et al.¹³ on 107 infants reported no change in the effectiveness of breastfeeding in the intervention and control groups 5 days and 8 weeks post-frenotomy. Therefore, in this study the authors aim to critically evaluate scientific research related to the effect of frenotomy treatment on breastfeeding ability in infants with ankyloglossia.

Materials and methods

This study was written using the rapid review method following the Preferred Reporting Items for Systematic Review and Meta-analysis (PRISMA) flow chart guidelines. The formulation of research questions follows the PICO framework (population, intervention, comparison, and outcome) as follows: (1) infants with ankyloglossia and breastfeeding difficulties (infant, ankyloglossia); (2) intervention: infants with ankyloglossia who undergo frenotomy (frenotomy); (3) comparison: infants with ankyloglossia who did not undergo frenotomy; (4) outcome: the effect of frenotomy on breastfeeding in infants with ankyloglossia (breastfeeding). Article searches were conducted through Pubmed, Scopus, and Embase databases according to predetermined research questions. The search method is carried out using a boolean operator that combines keywords using "AND" and "OR". Keywords used in Pubmed database are ("Infant"[MeSH Terms]) AND ("Ankyloglossia"[MeSH Terms] OR "Ankyloglossia"[Title/Abstract] OR "tongue tie"[All Fields]) AND ("frenotomy"[All Fields]) AND ("Breast Feeding"[MeSH Terms] OR "Breast Feeding"[Title/Abstract]). Keywords used in Scopus database are (infant) AND ((ankyloglossia) OR ("tongue tie")) AND (frenotomy) AND (("breast

feeding") OR (breastfeeding) OR (breastfed) OR ("breast fed")). Keywords used in Embase database are (infant) AND (ankyloglossia) OR (tongue-tie) AND (frenotomy) AND (breastfeeding). Additional article searches were also carried out manually by checking references from the articles obtained and would be used if they were relevant to the research topic.

Inclusion criteria were articles published in the last 15 years (2006-2021), discusses the indicated frenotomy in infants with ankyloglossia who have difficulty breastfeeding, randomized controlled trial, cohort, or quasi-experimental studies with a control group, and articles in English or Indonesian. Exclusion criteria were infants with certain medical conditions that could interfere breastfeeding, such as born prematurely (<37 weeks), down syndrome, cleft lip and/or cleft palate, congenital heart defects and case series, case report, and narrative review study design. The articles obtained were assessed for risk of bias using the Joanna Briggs Institute (JBI) Critical Appraisal Tools. The overall quality of the articles is divided into three categories, (1) low risk of bias (meets >75% of the assessment results), (2) moderate risk of bias (meets 50%-74% of the assessment results), (3) high risk of bias (meets <49% of the assessment results).

Results

Article search was carried out using PRISMA flowchart (see figure 1). In the initial search, a total of 560 articles were found, including 559 articles obtained from Pubmed, Scopus, and Embase, and 1 article obtained through manual hand searching. Subsequently, multiple articles were screened, 171 articles were excluded, leaving 389 articles for screening. A total of 372 articles were excluded because the title and abstract did not match the research question. Then, the remaining 17 articles were selected for eligibility and 12 articles were excluded. A total of 5 articles used in this review, including 4 randomized controlled trial (RCT) and 1 quasi-experimental study.

Based on the results of the data extraction of the articles obtained (see table

1), three studies carrying out research in the UK^{13,14,15}, one study conducting research in the US¹⁶, and another study was conducted in Israel¹⁷. Two studies^{13,15} compared intervention group that underwent frenotomy to a control group without frenotomy and both groups were given lactation support, while three studies^{14,16,17} compared the frenotomy group to a control group who had a sham surgery, which is a procedure where all the steps were the same as the original procedure, the only difference was whether or not an incision was made to the lingual frenulum. Total participants were 289 infants aged between 1-178 days. Gender proportions of the participants mentioned in three studies^{14,16,15} found more boys than girls, while two studies^{13,17} did not mention the number of participants by gender.

Indications for frenotomy in each study are different. One study¹³ performed frenotomy if the tongue-tie assessment based on the Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF) had a score of 6-12 (mild-moderate tongue-tie) and the breastfeeding assessment based on Latch, Audible swallowing, Type of nipple, Comfort, Holding (LATCH) had a score ≤ 8 , which indicates breastfeeding difficulties. Indication of frenotomy based on HATLFF score was also used in another article¹⁶ and accompanied by nipple pain or difficulty in breastfeeding after lactation management. Another study¹⁷ performed frenotomy if tongue-tie, which is defined as the inability of the infant to protrude the tip of the tongue into the lower gums with the tip of the tongue tied to the floor of the mouth by a tight frenulum based on physical examination was detected as well as the presence of nipple pain. Another study¹⁴ performed a frenotomy on infant who was referred for a frenotomy and the presence of nipple pain and tongue-tie was confirmed, but did not mention how the tongue-tie assessment was performed. Another study¹⁵ perform a frenotomy if there were complaints in breastfeeding such as nipple pain and poor latch and diagnosed

having a tongue-tie, but did not explain how the assessment is carried out.

Several assessment tools were used to assess tongue-tie and breastfeeding (see table 2). Two studies^{13,16} assessing ankyloglossia using a validated tool, HATLFF, while another three studies^{14,15,17} did not explain the details assessment of ankyloglossia. Breastfeeding assessment using validated tools, including LATCH, was carried out in two studies^{13,17}, Infant Breastfeeding Assessment Tool (IBFAT) in two studies^{13,16}, Breastfeeding Self-Efficacy Scale-Short Form (BSES-SF) in one study¹³, Short Form-McGill Pain Questionnaire (SF-MPQ) in one study¹⁶, and Visual Analogue Scale (VAS) in three studies^{13,14,17}. The maternal subjective assessment was carried out in three articles¹³⁻¹⁵. Timing of breastfeeding assessment after frenotomy varied between immediately after the intervention to follow-up 3 months later.

Technique for performing frenotomy is described in four studies¹⁴⁻¹⁷, whereas another article does not explain how frenotomy is performed.¹³ The technique includes lifting the tongue using a finger or a grooved director, then making an incision on the frenulum using surgical scissors carefully and not damaging vascular tissue, submandibular and sublingual ducts. Bleeding is stopped by applying light pressure with gauze or operator's finger on the incision area. All studies did not use anesthesia.

Two studies^{16,17} reported significant difference in nipple pain reduction between intervention and control groups, while the other two studies^{13,14} did not. Effectiveness of breastfeeding based on IBFAT scores in intervention group were found to be elevated in two studies^{15,16}, while another study¹³ found no significant difference in improvement between intervention and control groups. Two studies^{13,17} using LATCH to assess breastfeeding effectiveness did not show significant improvement difference between intervention

and control groups after frenotomy. Improvement of tongue-tie after frenotomy based on HATLFF score was found in one study¹³. Maternal self-efficacy based on BSES-SF scores were found to be elevated after frenotomy in one study¹³. Three studies¹³⁻¹⁵ found an increase in breastfeeding based on maternal subjective assessment through interviews. All studies¹³⁻¹⁷ reported no complications following frenotomy.

All randomized controlled trial studies¹³⁻¹⁷ have low risk of bias and one quasi-experimental study¹⁵ has moderate risk of bias. (see table 3 dan 4)

Discussion

Several studies have reported association between ankyloglossia and breastfeeding difficulties.^{18,19,20} Short lingual frenulum can interfere seal during breastfeeding. Changes in the sucking mechanism can lead to low milk transfer, prolonged breastfeeding, and nipple pain due to pressure from gums. These can then lead to early weaning and potentially switching to bottle feeding.⁹

Nipple pain is often experienced by mothers with ankyloglossia babies.^{21,22,23} Breastfeeding involves the peristaltic movement of the tongue so that limited movement of the tongue can cause friction from the tongue or the gum pads on the nipples.²⁴ Study by Dollberg et al.¹⁷ showed significant nipple pain decrease between intervention and control group immediately after frenotomy ($p < 0.01$) Buryk et al.¹⁶ found a placebo effect, which is significant nipple pain reduction both in the intervention and control group immediately after frenotomy, although the intervention group increased significantly more than the control group ($p < 0.001$). The placebo effect may occur because the mother can detect whether her child has had a frenotomy by the first breastfeeding after procedure. A decrease in nipple pain may occur due to an increase of baby's ability to protrude the tongue beyond

the gums or lower lip after frenotomy.²⁵ Two weeks after the intervention, Buryk et al.¹⁶ reported a decrease in nipple pain scores in both groups, although there was no significant difference between the two groups. This could be due to a cross-over sample, where all infants in the control group except for one infant chose to undergo frenotomy before follow up assessment at two weeks, therefore could affect the results.²⁶

In contrast to the two studies, Emond et al.¹³ and Berry et al.¹⁴ did not find a significant difference in nipple pain reduction between intervention and control groups. Emond et al.¹³ found no significant difference in nipple pain after frenotomy between the intervention group and the lactation-supported group at 5 days and 8 weeks follow up. This could be due to differences in the sample criteria, which excluded infants with severe ankyloglossia and only included infants with mild-moderate ankyloglossia. Berry et al.¹⁴ also found no significant difference in the reduction of nipple pain between the two groups immediately after intervention. This could be due to the fact that the mean age of infants in the study conducted by Berry et al.¹⁴ was several days older (32 days), whereas in Buryk et al.¹⁶ was 6 days so that it could be related to longer cumulative trauma to the mother's breast as a result of ineffective latch.²⁷ Lactation support that can be provided includes skin-to-skin contact, breastfeeding more often, and fixing breastfeeding positions such as propping baby's head toward the breast with nose facing forward, touch the baby's lower lip with nipple, then bring the baby close to the breast by pressing his back and shoulders, and put breast as much as possible in baby's mouth so that only a small part of the areola is visible.²⁸

Infant Breastfeeding Assessment Tool (IBFAT) was used to measure infant breastfeeding behaviour. Buryk et al.¹⁶ reported a significant difference in the increase of IBFAT scores immediately after frenotomy between intervention and control

group ($p=0.029$). However, in two weeks follow up, IBFAT scores increased but did not show significance between groups.¹⁶ Sharma et al.¹⁵ reported a significant increase in IBFAT scores before and after frenotomy in the intervention group ($p=0.0001$), while in the control group there was no significant increase ($p=0.16$).¹⁵ Emond et al.¹³ reported no significant difference in IBFAT scores between the frenotomy and the lactation-supported group at 5 days and 8 weeks of follow-up ($p=0.76$ dan $p=0.58$). Different results could be due to the fact that the participants in the study by Emond et al.¹³ only included infants with mild-moderate ankyloglossia and excluded severe ones.

Effectiveness of breastfeeding based on LATCH assessment was not found in the two studies^{13,17}. Dollberg et al.¹⁷ found no significant difference in LATCH scores between intervention and control group. Emond et al.¹³ also reported that there was no significant difference in the LATCH scores between intervention and lactation-supported group after 5 days and 8 weeks post-intervention ($p=1.0$ and $p=0.41$). This could be because there are components of the LATCH assessment that cannot be modified by the frenotomy, such as nipple type and holding aspect.²³ This is supported by a study conducted by Muldoon et al.²⁹ who looked at changes in each aspect of the LATCH component after frenotomy and found no change in aspects of the nipple type. Therefore, assistance by a lactation consultant is still needed to ensure breastfeeding is maintained after frenotomy.¹³ This is in compliance with study conducted by Rasteniene et al.²² that all mothers who continue breastfeeding after frenotomy are those who receive lactation consultations after.

Study by Emond et al.¹³ found no change in the breastfeeding variables (nipple pain, IBFAT score, LATCH score) as previously mentioned. However, the tongue-tie is improved, indicated by a significant

increase in HATLFF scores in the intervention group compared to the control group on the fifth day of follow-up ($p<0.0001$). This indicates that a better assessment tool is needed because simple inspection of the lingual frenulum is not sufficient to determine which infant requires a frenotomy procedure.¹³ This is in line with study by Schlatter et al.³⁰ that only the HATLFF function score <11 and peristalsis item score indicates breastfeeding problems in infants with tongue-tie, while other appearance or function parameters from the HATLFF assessment were not significant risk factors for breastfeeding problem.

Mother's confidence in breastfeeding was found to be significantly increased in the intervention group compared to the control group ($p=0.002$) based on BSES-SF.¹³ This is in line with study by Ghaheri et al.³¹ which showed a significant change in the BSES-SF score before and after frenotomy. In spite of that, study by Emond et al.²⁵ could not show an objective and sustained improvement in breastfeeding after frenotomy because majority of the sample in the control group ended up receiving frenotomy on the fifth day of follow-up, thus affecting results of the study.

Breastfeeding improvement based on subjective maternal assessment was reported to increase in three studies¹³⁻¹⁵. Emond et al.¹³ conducted interviews with parents and found that most parents reported improved breastfeeding after the intervention and reduced nipple pain. Sharma et al.²⁰ also found a significant increase in breastfeeding between intervention group compared to the control group immediately after frenotomy ($p=0.0074$). Berry et al.¹⁴ also found significant difference breastfeeding improvement between intervention group and control group immediately after frenotomy ($p<0.02$), including better latch, reduced pain, and more effective breastfeeding. This is in line with other studies that have found subjective improvements in nipple pain.²⁴ Breastfeeding is a subjective experience, so

the mother's assessment is considered a good assessment to see whether or not there is a change in breastfeeding.¹⁴

Complications after frenotomy are rare, but some complications such as excessive bleeding, infection of the incision area, injury to surrounding structures, reattachment of the lingual frenulum due to excessive wound, and oral aversion can occur.^{3,32,33} All studies¹³⁻¹⁷ reported no complications after frenotomy. Two studies^{14,17} reported bleeding which can be controlled in seconds. Emond et al.¹³ also found no side effects occurred, however on the fifth day of follow-up there were 63 infants who had small white patches at the base of the frenulum and it took an average of 7 days (between 1-30 days) to heal. White patches are part of wound healing process from the incision. Stretching the frenulum can be done after the procedure to avoid reattachment by pressing the index finger on the wound upwards routinely 5-6 times a day for 7 to 10 days.³⁴ However, no studies have reported its effectiveness in preventing frenulum reattachment. In order to avoid complications, clinicians must understand the surgical technique, master the landmarks, and have the ability to identify and treat complications that can occur.³⁵

This study has several limitations, including the non-uniformity of assessment in defining ankyloglossia so that the inclusion criteria of participants are not clear and the severity of ankyloglossia in each study may differ, the high variation of assessment tools in measuring breastfeeding ability so that the authors find it difficult to compare experiences between individuals, and only a few articles met the inclusion criteria because the lack of studies with control groups on this topic which could be due to ethical constraints. Future research could use uniform assessment so that which groups of infants would benefit from frenotomy can be determined.

Conclusions

Frenotomy was found to be inconsistent in improving breastfeeding ability in infant with ankyloglossia. Practitioners should do case selection carefully by taking into account the assessment of tongue-tie and breastfeeding, as well as conservative approach (lactation support) before performing frenotomy.

Acknowledgements

The author would like to thank Prof. Dr. Achmad Syawqie, drg., MS, Dr. Kartika Indah Sari, drg., M.Kes, dan Farah Asnely Putri, drg., Sp.BM(K) who have given suggestions to this manuscript.

Declaration of Interest

The authors report no conflict of interest.

No	Author	Study design	Sample characteristics			Indication	Ankyloglossia assessment	Breastfeeding assessment	Follow-up	Results
			Age	Gender	Groups					
1.	Sharma et al/ 2015	Quasi-experimental (non-randomized)	15-178 days (mean 38 days)	Boys = 23 Girls = 19	Intervention= 36 Control (lactation support) = 6	Diagnosed tongue-tie, poor latch and maternal nipple pain	N/A	IBFAT and maternal subjective assessment	1 month	1. There was a significant difference in the increase in breastfeeding between the intervention and control groups based on subjective maternal assessment (p=0.0074) 2. There was an increase in IBFAT score before and after frenotomy in the intervention group (p=0.0001) 3. There was no significant increase in IBFAT score before and after frenotomy in the control group (p=0.16)
2.	Emond et al./ 2013	Randomized controlled trial (single-blind)	<2 weeks	N/A	Intervention= 55 Control (lactation support) = 52	LATCH score ≤8, HATLFF score 6-12 (mild-moderate tongue-tie)	HATLFF	LATCH, IBFAT, BSES-SF, VAS, maternal subjective assessment (open question)	5 days and 8 weeks	5 days follow-up: 1. There was no significant difference in LATCH scores (p=1.0), IBFAT (p=0.76), BSES-SF (p=0.53), and VAS (p=0.13) between the two groups. 2. The increase in BSES-SF scores was greater in the intervention group than the control group (p=0.002) 3. There was an increase in the HATLFF score in the intervention group (p<0.0001) 8 weeks follow-up: 1. There was no significant difference in LATCH scores (p=0.41), IBFAT (p=0.58), BSES-SF (p=0.62), VAS (p=0.41), and infant weight (p=0.41), =0.54) between the two groups 2. Maternal subjective assessment reports an improvement in the way the baby breastfed and a reduction in nipple pain
3.	Berry et al./ 2012	Randomized controlled trial (double-blind)	5-115 days (mean 32 days)	Boys= 40 Girls = 20	Intervention= 30 Control (sham procedure) = 28	Referred for frenotomy and confirmed tongue-tie and nipple pain, difficulty in latching or ineffective breastfeeding	N/A	LATCH and IBFAT modification, VAS, maternal subjective assessment (open question)	Immediately after frenotomy dan 3 months	Immediately after frenotomy: 1. There was a significant difference in breastfeeding improvement (better latch, less pain, and more effective breastfeeding) based on subjective assessment in the intervention group compared to the control group (p<0.02) 2. There was no difference in the decrease in VAS scores between the two groups (p=0.13) 3 months follow-up: 92% (54 of 59) mothers reported an improvement in breastfeeding

4.	Buryk et al./ 2011	Randomized controlled trial (single-blind)	1-35 days (mean 6 days)	Boys=38 Girls=20	Intervention= 30 Control (sham procedure)=28	The presence of significant ankyloglossia based on HATLFF, breastfeeding difficulties and maternal nipple pain after lactation management	HATLFF	SF-MPQ, IBFAT	Immediately after frenotomy and 2 weeks	Immediately after frenotomy: 1. SF-MPQ scores in both groups decreased significantly, with the intervention group decreasing significantly more than the control group (p<0.001) 2. IBFAT score significantly increased in the intervention group compared to the control group (p=0.029) 2 weeks follow-up: There was a decrease in SF-MPQ and IBFAT scores in the two groups, but not significant between the two groups
5.	Dollberg et al./ 2006	Randomized controlled trial	1-21 days	N/A	Intervention = 14 Control (sham procedure) = 11	Presence of nipple pain and presence of a tongue-tie which was defined as the inability to protrude the tip of the tongue into the lower gums with the tip of the tongue tied to the floor of the mouth by a tight frenulum based on physical examination	N/A	LATCH, VAS	Immediately after frenotomy	1. There was a significant difference in the decrease in VAS scores in the intervention group compared to the control group (p<0.01) 2. There was an almost significant difference in the increase in LATCH scores in the intervention group compared to the control group (p=0.06)

Notes: Latch, Audible Swallowing, nipple Type, Comfort, Hold (LATCH); Infant Breast Feeding Assessment Tool (IBFAT); Breastfeeding Self-Efficacy Score (BSES); Visual Analogue Scale (VAS); The Short Form McGill Pain Questionnaire (SF-MPQ); Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF); N/A : *Not Available*

Table 1. Data extraction.

Variables	Instrument
Breastfeeding	<ol style="list-style-type: none"> <i>Latch, Audible Swallowing, Type of nipple, Comfort, Holding (LATCH)</i> <ol style="list-style-type: none"> Five aspects assessed: <i>latch, audible swallowing, type of nipple, comfort, holding</i> on a scale of 0-2 Maximum score 10 points The higher the score, the higher the effectiveness in breastfeeding <i>Infant Breastfeeding Assessment Tool (IBFAT)</i> <ol style="list-style-type: none"> Four aspects assessed: <i>readiness to feed, rooting, latching on, sucking</i> on a scale of 0-3 Maximum score 12 points The higher the score, the higher the effectiveness in breastfeeding <i>Breastfeeding Self-Efficacy Scale-Short Form (BSES-SF)</i> <ol style="list-style-type: none"> Assessing maternal confidence in breastfeeding using a Likert scale 1-5 consisting of 14 question items Maximum score 70 points The higher the score, the higher the mother's confidence in breastfeeding (<50 predicted weaning)

Nipple pain	<p>1. <i>Visual Analogue Scale (VAS)</i> a. 0-10 point scale (<i>no pain, mild, moderate, severe, very severe, worst pain</i>) b. The higher the score, the more pain felt</p> <p>2. <i>Short-form McGill Pain Questionnaire (SF-MPQ)</i> a. Consists of 3 parts: i. 15 words that describe the sensory and affective aspects of pain (0-3 scale; <i>none, mild, moderate, severe pain</i>) ii. VAS iii. Descriptors for pain intensity (0-5 scale) b. Maximum score 50 points c. The higher the score, the more pain felt</p>
Severity of tongue-tie	<p><i>Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF)</i> a. Assess 5 aspects of tongue appearance and 7 aspects of tongue function b. Frenotomy indicated If the function score is <11 and appearance score is <8 c. The higher the score, the lower the severity of the tongue-tie</p>

Table 2. Assessment tools used in all articles.

No	JBI Checklist Questions	Emond et al. 2013	Berry et al. 2012	Buryk et al. 2011	Dollberg et al. 2006
1	Was true randomization used for assignment of participants to treatment groups?	Yes	Yes	Yes	Yes
2	Was allocation to treatment groups concealed?	Yes	Yes	Unclear	Yes
3	Were treatment groups similar at the baseline?	Yes	Yes	Yes	Yes
4	Were participants blind to treatment assignment?	Yes	Yes	Yes	Yes
5	Were those delivering treatment blind to treatment assignment?	No	Unclear	Unclear	Unclear
6	Were outcomes assessors blind to treatment assignment?	Unclear	Yes	Unclear	Yes
7	Were treatment groups treated identically other than the intervention of interest?	Yes	Yes	Yes	Yes
8	Was follow up complete and if not, were differences between groups in terms of their follow up adequately described and analyzed?	Yes	Yes	Yes	Yes
9	Were participants analyzed in the groups to which they were randomized?	Yes	Unclear	Yes	Unclear
10	Were outcomes measured in the same way for treatment groups?	Yes	Yes	Yes	Yes
11	Were outcomes measured in a reliable way?	Yes	Yes	Yes	Unclear
12	Was appropriate statistical analysis used?	Yes	Yes	Yes	Yes
13	Was the trial design appropriate, and any deviations from the standard RCT design (individual randomization, parallel groups) accounted for in the conduct and analysis of the trial?	Yes	Yes	Yes	Yes
Result		84,61% (low risk of bias)	84,61% (low risk of bias)	76,92% (low risk of bias)	76,92% (low risk of bias)

Table 3. Risk of bias assessment for randomized controlled trials studies.

No	JBI Checklist Questions	Sharma et al. 2015
1	Is it clear in the study what is the 'cause' and what is the 'effect' (i.e. there is no confusion about which variable comes first)?	Yes
2	Were the participants included in any comparisons similar?	Unclear

3	Were the participants included in any comparisons receiving similar treatment/care, other than the exposure or intervention of interest?	No
4	Was there a control group?	Yes
5	Were there multiple measurements of the outcome both pre and post the intervention/exposure?	Yes
6	Was follow up complete and if not, were differences between groups in terms of their follow up adequately described and analyzed?	Yes
7	Were the outcomes of participants included in any comparisons measured in the same way?	Yes
8	Were outcomes measured in a reliable way?	Unclear
9	Was appropriate statistical analysis used?	Yes
Result		66,67% (moderate risk of bias)

Table 4. Risk of bias assessment for quasi-experimental study.

References

- Rowan-Legg A, Cummings C, Gander S, et al. Ankyloglossia and breastfeeding. *Paediatr Child Heal*. 2015;20(4):209-213.
- Bargale S, Deshpande A, Macwan C, Sikligar S. Ankyloglossia in the Infant and Young Child : A Review. *J Adv Med Dent Sci*. 2014;2(1):63-67.
- Nowak AJ, Christensen JR, Mabry TR, et al. *Pediatric Dentistry: Infancy Through Adolescence*. 6th ed. Philadelphia: Elsevier; 2019:406.
- Genna CW, Saperstein Y, Siegel SA, et al. Quantitative imaging of tongue kinematics during infant feeding and adult swallowing reveals highly conserved patterns. *Physiol Rep*. 2021;9(3):e14685.
- Ghaheer BA, Cole M, Fausel SC, et al. Breastfeeding improvement following tongue-tie and lip-tie release: A prospective cohort study. *Laryngoscope*. 2017;127(5):1217-1223.
- Ghaheer BA, Cole M, Mace JC. Revision Lingual Frenotomy Improves Patient-Reported Breastfeeding Outcomes: A Prospective Cohort Study. *J Hum Lact*. 2018;34(3):566-574.
- Marcadante KJ, Kliegman RM. *Nelson Essentials of Pediatrics*. 8th ed. Philadelphia: Elsevier; 2019:99.
- Sum FHKMH, Zhang L, Ling HTB, et al. Association of breastfeeding and three-dimensional dental arch relationships in primary dentition. *BMC Oral Health*. 2015;15(1):1-9.
- Brzęcka D, Garbacz M, Micał M, et al. Diagnosis, classification and management of ankyloglossia including its influence on breastfeeding. *Dev period Med*. 2019;23(1):79-87.
- Dean JA, Jones JE, Walker Vinson LA, eds. *McDonald and Avery's Dentistry for the Child and Adolescent*. 10th ed. St.Louis: Elsevier; 2016:70.
- Srinivasan A, Al Khoury A, Puzhko S, et al. Frenotomy in Infants with Tongue-Tie and Breastfeeding Problems. *J Hum Lact*. 2019;35(4):706-712.
- Ramoser G, Guóth-Gumberger M, Baumgartner-Sigl S, et al. Frenotomy for tongue-tie (frenulum linguae breve) showed improved symptoms in the short- and long-term follow-up. *Acta Paediatr Int J Paediatr*. 2019;108(10):1861-1866.
- Emond A, Ingram J, Johnson D, et al. Randomised controlled trial of early frenotomy in breastfed infants with mild-moderate tongue-tie. *Arch Dis Child Fetal Neonatal Ed*. 2014;99(3):F189-F195.
- Berry J, Griffiths M, Westcott C. A double-blind, randomized, controlled trial of tongue-tie division and its immediate effect on breastfeeding. *Breastfeed Med*. 2012;7(3):189-193.
- Sharma SD, Jayaraj S. Tongue-tie division to treat breastfeeding difficulties: Our experience. *J Laryngol Otol*. 2015;129(10):986-989.
- Buryk M, Bloom D, Shope T. Efficacy of neonatal release of ankyloglossia: A randomized trial. *Pediatrics*. 2011;128(2):280-288.
- Dollberg S, Botzer E, Grunis E, et al. Immediate nipple pain relief after frenotomy in breast-fed infants with ankyloglossia: a randomized, prospective study. *J Pediatr Surg*. 2006;41(9):1598-1600.
- Walker RD, Messing S, Rosen-Carole C, et al. Defining Tip-Frenulum Length for Ankyloglossia and Its Impact on Breastfeeding: A Prospective Cohort Study. *Breastfeed Med*. 2018;13(3):204-210.
- Campanha SMA, Martinelli RLC, Palhares DB. Association between ankyloglossia and breastfeeding. *CODAS*. 2019;31(1):e20170264.
- Riskin A, Mansovsky M, Coler-Botzer T, et al. Tongue-tie and breastfeeding in newborns-Mothers' perspective. *Breastfeed Med*. 2014;9(9):430-437.
- Barberá-Pérez PM, Sierra-Colomina M, Deyanova-Alyosheva N, et al. Prevalence of ankyloglossia in newborns and impact of frenotomy in a baby-friendly hospital [Prevalencia de la anquiloglosia en el recién nacido e impacto de la frenotomía en un hospital de la iniciativa para la humanización de la asistencia al nacimiento. *Bol Med Hosp Infant Mex*. 2021;78(5):418-423.
- Rastenienė R, Puriene A, Aleksejuniene J. Tongue function characteristics in infants experiencing breastfeeding difficulties and changes in breastfeeding after frenotomy procedures. *Clin Oral Investig*. 2021;25(8):4871-4877.
- Wakhanrittee J, Khorana J, Kiatipunsodsai S. The outcomes of a frenulotomy on breastfeeding infants followed up for 3 months at Thammasat University Hospital. *Pediatr Surg Int*. 2016;32(10):945-952.
- Praborini A, Purnamasari H, Munandar A, et al. Early Frenotomy Improves Breastfeeding Outcomes for Tongue-Tied Infants. *Clin Lact*. 2015;6(1):9-15.
- Muldoon K, Gallagher L, McGuinness D, et al. Effect of frenotomy on breastfeeding variables in infants with ankyloglossia (tongue-tie): A prospective before and after cohort study. *BMC Pregnancy Childbirth*. 2017;17(1):373.
- Gupta S. Intention-to-treat concept: A review. *Perspect Clin Res*. 2011;2(3):109.
- Francis DO, Krishnaswami S, McPheeters M. Treatment of ankyloglossia and breastfeeding outcomes: A systematic review. *Pediatrics*. 2015;135(6):e1458-e1466.
- Yohmi E, Partawi IGAN, Pambudi W, et al. Diagnosis dan Tata Laksana Ankyloglossia (Tongue-Tie). *Pandu Prakt Klin Ikat Dr Anak Indones*. 2017:1-17.
- Muldoon K, Gallagher L, McGuinness D, et al. Effect of frenotomy on breastfeeding variables in infants with ankyloglossia (tongue-tie): A prospective before and after cohort study. *BMC Pregnancy Childbirth*. 2017;17(1):1-9.
- Schlatter S-M, Schupp W, Otten J-E, et al. The role of tongue-tie in breastfeeding problems—A prospective observational study. *Acta Paediatr Int J Paediatr*. 2019;108(12):2214-2221.
- Ghaheer BA, Cole M, Mace JC. Revision Lingual Frenotomy Improves Patient-Reported Breastfeeding Outcomes: A

- Prospective Cohort Study. *J Hum Lact.* 2018;34(3):566-574.
32. Connor O, Connor MEO, Gilliland AM, et al. Complications and misdiagnoses associated with infant frenotomy: results of a healthcare professional survey. *Int Breastfeed J.* 2022:1-9.
 33. Rajput N V, Reid N, Rajput N V. Acute feed refusal followed by *Staphylococcus aureus* wound infection after tongue-tie release. *J Paediatr Child Health.* 2014;51(12):1030-1031.
 34. Kenny-Scherber AC, Newman J. Office-based frenotomy for ankyloglossia and problematic breastfeeding. *Can Fam Physician.* 2016;62(7):570-571.
 35. Power RF, Murphy JF. Tongue-tie and frenotomy in infants with breastfeeding difficulties: Achieving a balance. *Arch Dis Child.* 2015;100(5):489-494.