

ORIGINAL ARTICLES

Newborn Tongue-tie: Prevalence and Effect on Breast-Feeding

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Objective: The purposes of this study were: (1) to determine whether breast-fed infants with tongue-tie have decreased rates of breast-feeding at 1 week and 1 month of age, (2) to determine the prevalence of tongue-tie, and (3) to test the usefulness of the Assessment Tool for Lingual Frenulum Function (ATLFF) in assessing the severity of tongue-tie in breast-feeding newborns.

Methods: A case-control design was used. All infants in the Regions Hospital newborn nursery were examined for tongue-tie. Tongue-tied babies were examined using the ATLFF. Two breast-feeding babies with normal tongues were identified and matched for each case. Mothers were interviewed when the babies were 1 week and 1 month old.

Results: The prevalence of tongue-tie was 4.2%. Forty-nine tongue-tied and 98 control infants were enrolled. Tongue-tied babies were 3 times as likely as control babies to be bottle fed only at 1 week [risk ratio (RR), 3.11; 95% confidence interval (CI), 1.21, 8.03] By 1 month, tongue-tied babies were as likely as controls to be bottle fed only. (RR, 1.00; 95% CI, 0.55, 1.82) Twelve of the tongue-tied infants had ATLFF scores of "Perfect," none had scores of "Acceptable," and 6 had scores of "Function Impaired." The remaining 31 infants had scores that fell into none of these categories.

Conclusions: Tongue-tie is a relatively common condition in newborns. Affected infants are significantly more likely to be exclusively bottle-fed by 1 week of age. The ATLFF was not a useful tool to identify which tongue-tied infants are at risk for breast-feeding problems. (J Am Board Fam Pract 2005;18: 1-7.)

Extensive research documents the diverse advantages to infants, mothers, families, and society from breast-feeding.¹ In order for a breast-feeding infant to properly latch on to a mother's breast, the infant's tongue must be able to thrust to the edge of the lower gum and cup around the areola and the mother's elongated nipple. The infant's tongue movement may be restricted by an abnormal lingual frenulum. Tongue-tie, or ankyloglossia, is the condition in which the frenulum is tight or short, extending to the tip or almost to the tip of the tongue.

In tongue-tie, the lingual frenulum tethers the tongue to the floor of the mouth, prohibiting extension beyond the lower gum. When breast-feeding, the infant is unable to form an appropriate seal. This frequently results in sore nipples for the mother. Ineffective sucking in the newborn may also result in insufficient milk drainage in the mother, which in turn may lead to mastitis, decreased maternal milk supply, and, eventually, poor infant weight gain secondary to non-nutritive sucking.² Tongue-tie does not normally cause a problem with the more passive efforts involved with bottle feeding.

The lack of a uniform definition for tongue-tie has led to a wide variation in reported prevalence. Other factors contributing to varying prevalences include different study methods and patient populations. The importance of the functional assessment of the tongue has been emphasized by several authors, who note that a frenulum may look short but still have sufficient elasticity to function.³ The only available tool designed to assess breast-feeding

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newborns with tongue-tie for the severity of the problem is the Assessment Tool for Lingual Frenulum Function (ATLFF).³ A quantitative tool, the ATLFF has been used in one previous study to assess newborns with tongue-tie.⁴

The primary purpose of this study was to use a case-control design to determine whether breast-fed infants with tongue-tie have lower rates of breast-feeding at 1 week and 1 month of age compared with similar infants without tongue-tie. A secondary purpose was to determine the prevalence of tongue-tie in infants born in an inner-city hospital. The final purpose was to test the usefulness of the ATLFF in assessing the severity of tongue-tie in breast-feeding newborns.³

Methods

The study took place at Regions Hospital, a 420-bed hospital in St. Paul, MN. The HealthPartners Institutional Review Board approved the study.

The study used a case-control design, with 2 controls individually matched to each case. Before the study began, one of the investigators (NJB) met with the supervisor of the nursery nurses, who helped coordinate communication with the nursing staff. The investigator talked with the day and evening shift nursery nurses about identifying tongue-tie. A workbook with several photographs of infants with tongue-tie was placed in the nursery for reference. From October 1, 2000, through May 1, 2002, when all newborns were admitted to the normal newborn nursery, the nurses looked at the infants' oral cavity and documented whether or not they thought the infants had tongue-tie, based on appearance only.

Study investigators used the ATLFF to assess the infants' tongues. The ATLFF was developed by Alison Hazelbaker for her Master's degree thesis in human development.³ The ATLFF consists of 5 items assessing newborn tongue appearance and 7 items assessing tongue function.⁴ The function scores and appearance scores are combined to determine a total score that may fall into a category of "Perfect," "Acceptable," or "Function Impaired." Hazelbaker has modified the ATLFF scoring system, and the following definitions were used in this study: Perfect, function score of 14, regardless of appearance score; Acceptable, function score of 11, if appearance score is 10; and Function Impaired, function score <11.

All infants with function scores of 13 and 12, and infants with function scores of 11 and with appearance scores <10, fall into none of the above categories. Before the study began, Ms. Hazelbaker came to Regions Hospital and personally instructed the investigators in the use of the ATLFF and was videotaped demonstrating the use of the tool.

One of the 3 investigators examined the tongues of all babies thought by the nurses' initial screening to have the appearance of tongue-tie. If the investigator agreed that the baby had the appearance of tongue-tie, she formally assessed the tongue using the ATLFF. When possible, more than one investigator examined the same baby using the ATLFF. When more than one investigator performed the ATLFF on the same infant, the highest rating was used for the data analysis. After the first 9 infants had been examined by more than one examiner, the inter-rater reliability of the ATLFF was determined. After the inter-rater reliability was calculated, the investigators discussed where their examinations differed and how they could be more alike in the future.

The investigators or a research assistant invited mothers of breast-feeding tongue-tied babies to participate in the study. Mothers were excluded for the following reasons: speaking neither English nor Spanish; having their infant in the Intensive Care Nursery; being unable to be contacted by telephone; having infants with mid-facial abnormalities or significant hypotonia; or having twins.

After obtaining informed and written consent from the mothers, the investigators or research assistant briefly interviewed them. Questions about potentially confounding factors, such as previous breast-feeding experience, were asked. The investigators or assistant then showed the mothers how to complete the Infant Breast-feeding Assessment Tool (IBFAT) for each feeding during their stay at the hospital.⁵ The IBFAT is a short assessment and measurement of infant breast-feeding competence designed to be used easily by mothers or nurses. The range of scores for each of 4 components is 0 to 3, with a total score ranging from 0 to 12. A score of 12 indicates a vigorous, effective feeding. Because 85% of the mothers completed an IBFAT score for only one feeding, the initial IBFAT score was used for the analyses. Unfortunately, many babies take 36 to 48 hours to achieve the optimum IBFAT score, so that the later scores are most relevant.⁵ The HealthPartners Survey Center con-

ducted follow-up telephone surveys with the English speaking mothers when the infants were 4 to 7 days old and 1 month old.

The investigators identified 2 breast-fed infants who were not tongue-tied, and these infants were used as control subjects for each case. Mothers of control infants were excluded for the same reasons as the mothers of the tongue-tied infants. Control infants were matched to a tongue-tied infant by the following factors: maternal age within 5 years, maternal race, first versus later birth-order child, and sex of the child. Control infants were examined with the ATLFF. Although the ATLFF is not designed to be used with normal infants, all the function items and some of the appearance items can be tested on normal infants. Therefore the function of the tongues of the control infants was examined and documented in a uniform manner. The mothers of control infants were taught how to complete the IBFAT while in the hospital. They also received follow-up telephone calls at 1 week and 1 month of age. All study materials were translated into Spanish. A Spanish-speaking research assistant obtained consent and conducted the interviews for mothers who spoke Spanish.

To minimize potential bias, the purpose of the study was described to the mothers as to determine whether the appearance and movement of babies' tongues affect how well they breast-feed. The investigators or research assistant did not tell the mothers that their babies had tongue-tie. In addition, the investigators' findings on the ATLFF were not communicated to the lactation consultants, the infant's primary physician, or to the mothers. If the lactation consultants were concerned that an infant was having breast-feeding problems because of tongue-tie, the consultants followed an already established, written hospital protocol regarding this issue. The protocol describes an assessment process including obtaining a clinical history, doing a visual oral inspection, and oral-digital examination. The protocol includes suggestions regarding nursing positioning, suck/tongue training, and nipple shields. According to the protocol, the lactation consultant discussed the need for a frenotomy with the infant's own physician. If a study infant received a frenotomy, his or her study data up until the time of the frenotomy was included in the analysis.

Statistical Analyses

The Mantel-Haenszel χ^2 statistic for matched data was used to compare baseline, 1-week, and 1-month characteristics of the study mothers and to test for differences in breast-feeding status between groups. The relative risks were estimated using the Mantel-Haenszel logit method controlling for the variable which tracks the matching. The inter-rater reliability of the ATLFF was measured using the κ statistic for those infants for whom more than one investigator performed the ATLFF on the same infant. The overall rating of function impaired versus not function impaired was compared. Analyses were performed using SAS statistical software, version 8.02 (SAS Institute Inc, Cary, NC).

Results

An outline of the study is shown in Figure 1. During the study period, 3490 infants were cared for in the normal newborn nursery at Regions Hospital. Of these, 148 were identified as having tongue-tie by the nursery nurses and confirmed by the study investigators, for a prevalence of 4.24%. 103 of the affected newborns were male and 45 were female (male-female ratio, 2.3:1.0).

Among the 148 tongue-tied infants, 78 met eligibility criteria. Of the 78 eligible, 22 were not enrolled for the reasons shown in the figure. Six infants were dropped because their mothers did not complete an IBFAT, and one infant was dropped from the study because no controls could be identified. Therefore, 49 tongue-tied infants remained in the study, which is 63% of the eligible infants.

Two control infants without tongue-tie were identified for the 49 infants. They were successfully matched except for 2 infants. One of the selected control infants matched for all but maternal age, and one for all but sex and maternal age.

Ninety-three (95%) of the control infants had function scores of 14 of 14 on the ATLFF. Four control infants had function scores of 13, and one control infant had a function score of 12 on the ATLFF. Characteristics of the case and control mothers, including their previous breast-feeding experiences, are shown in Table 1.

The mean IBFAT score of the tongue-tied infants was 10.08 and the mean score for control infants was 9.97. Four of the tongue-tied infants had frenotomies. Their IBFAT scores were 12, 9, 7, and 9. They all continued to breast-feed for the

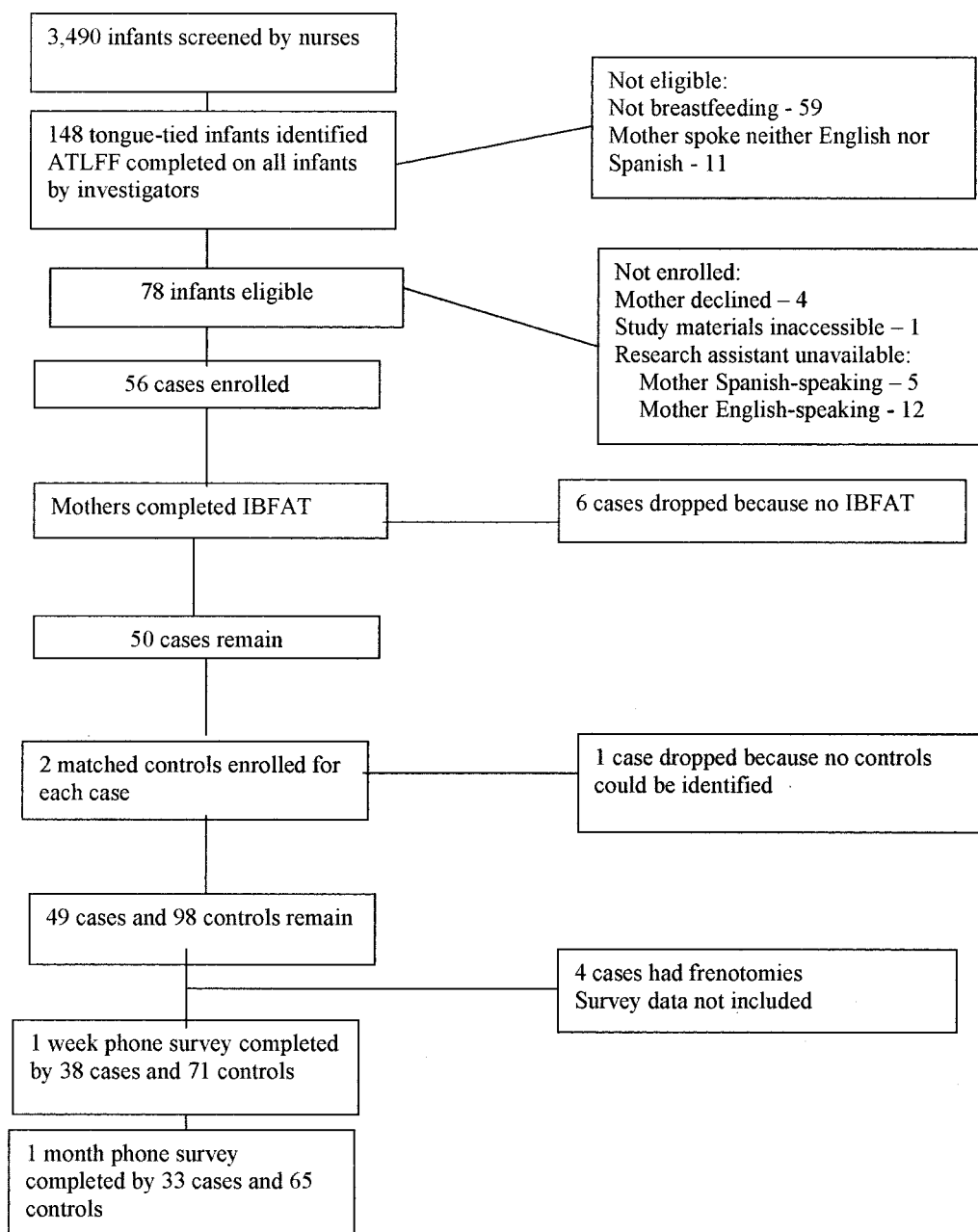


Figure 1. Study outline. ATLFF, Assessment Tool for Lingual Frenulum Function; IBFAT, Infant Breast-feeding Assessment Tool.

month of follow-up, but their survey results are not included in the remainder of the analyses.

Telephone follow-up was available for 38 of the now 45 cases (84%) and 71 (72%) of the controls at 1 week. As shown in Table 2, tongue-tied babies were approximately 3 times as likely as control babies to be bottle-fed only at 1 week [risk ratio (RR), 3.11; 95% confidence interval (CI), 1.21, 8.03]. Telephone follow-up was available for 33 (73%) cases and 65 (66%) controls at 1 month. As

shown in Table 2, by 1 month of age, tongue-tied babies were as likely as control subjects to be bottle fed only (RR, 1.00; 95% CI, 0.55, 1.82).

The responses to the 1-week and 1-month survey by the breast-feeding mothers are shown in Table 3. Case and control breast-feeding mothers reported similar concerns and symptoms. Over twice as many breast-feeding mothers of tongue-tied infants reported extremely sore nipples and breast pain at the 1-month survey. The differences

Table 1. Characteristics of Study Mothers

	Case Subjects (n = 49)	Control Subjects (n = 98)	P value
Mean age, years (standard deviation)	25.9 (6.2)	26.5 (5.9)	.193
Race			
White	28 (57.1%)	56 (57.1%)	.974
Hispanic	15 (30.6%)	30 (30.6%)	
Black	4 (8.1%)	8 (8.1%)	
Asian	2 (4.0%)	4 (4.0%)	
Male newborn	32 (65.3%)	63 (64.2%)	.480
Nulliparous	22 (44.8%)	44 (44.8%)	.999
Decided to breast-feed			.149
Before pregnancy	29 (59.2%)	69 (70.4%)	
During pregnancy	18 (36.7%)	26 (26.5%)	
After delivery	2 (4.1%)	3 (3.1%)	
Prepared for breast-feeding by			.367
Classes	11 (22.9%)	15 (15.8%)	
Reading	15 (31.3%)	31 (32.6%)	
Were breast-fed themselves	25 (51.0%)	52 (53.1%)	.811
The baby's father was breast-fed	22 (44.9%)	48 (49.5%)	.655
Number of previous babies breast-fed			.394
None	25 (53.2%)	45 (48.9%)	
≥1	22 (46.8%)	47 (51.1%)	
Has household/family member who feels should NOT breast-feed	1 (2.0%)	2 (2.0%)	.999

in case and control mothers in these 2 symptoms are not statistically significant, possibly because of the small number of women (ie, a type II error).

The responses of bottle-feeding only case and control mothers to both the 1- week and 1-month surveys are combined in Table 4, because of the small number of respondents. Bottle-feeding case and control mothers reported similar concerns and symptoms.

Two of the 3 investigators (LAR and DJM-K) examined 95% of the infants. Their inter-rater agreement on whether or not the infant had a score of Function Impaired on the ATLFF was moderate ($\kappa = 0.49$).⁶

Twelve of the tongue-tied infants had ATLFF scores of Perfect, none had scores of Acceptable, and 6 had scores of Function Impaired. The remaining 31 infants had scores that fell into none of

these categories. The outcomes at 1 week are known for 8 of the infants with Perfect scores and 5 of the infants with Function Impaired scores and are shown in Table 5.

Discussion

One aim of our study was to determine the prevalence of tongue-tie in infants born in an inner-city hospital. Our finding of a prevalence of 4.24% is consistent with recent reports of the prevalence of tongue-tie in newborns of 4.4%⁷ and 4.8%.⁸ Tongue-tie was found in 4.4% of five hundred neonates from well baby nurseries in Memphis, TN, that were examined for oral anomalies by 2 investigators.⁷ In a 2000 report, 4.8% of 1041 newborns in Palo Alto, CA, were found to be tonguetied, with a male-female ratio of 2.6:1.0. Several

Table 2. Feeding Status of Infants at 1 Week and 1 Month of Age

	1 Week*		1 Month†	
	Case Subjects (n = 38)	Control Subjects (n = 71)	Case Subjects (n = 33)	Control Subjects (n = 65)
Bottle-feeding only	8 (21.05%)	4 (5.63%)	10 (30.3%)	15 (23.08%)
Breast-feeding and bottle-feeding	15 (39.47%)	32 (45.07%)	20 (60.61%)	40 (61.54%)
Breast-feeding only	15 (39.47%)	35 (49.3%)	3 (9.09%)	10 (15.38%)

* P = .035.

† P = .46.

Table 3. Survey of Mothers Breast-Feeding at 1 Week and 1 Month of Age

	1 Week			1 Month		
	Case Subjects (n = 30)	Control Subjects (n = 67)	P Value	Case Subjects (n = 23)	Control Subjects (n = 50)	P Value
Breast-feeding going well:			.398			
Yes	29 (96.7%)	63 (94%)				
No	1 (3.3%)	1 (1.5%)				
Don't know		3 (4.5%)				
Extremely sore nipples	7 (23.3%)	19 (28.4%)	.216	5 (21.7%)	5 (10%)	.439
Nipples cracked or bleeding	4 (13.3%)	13 (19.4%)	.216	2 (8.6%)	6 (12%)	.398
Mother worried baby not growing well	1 (3.3%)	6 (9.0%)	.355	3 (13%)	7 (14%)	.999
Baby's doctor worried baby not growing well	1 (3.3%)	2 (3.0%)	.695	1 (4.3%)	2 (4%)	.617
Baby always hungry				9 (39.1%)	19 (38%)	.522
Breast pain				8 (34.7%)	8 (16%)	.425
Breast infection				3 (13%)	4 (8%)	.674
Not enough milk				1 (4.3%)	4 (8%)	.808

reports have noted a male predominance of cases, as did we.^{4,7-9}

The tongue-tied infants in this study were 3 times more likely to be exclusively bottle-fed at 1 week than matched control subjects with normal tongues. However, 80% of the tongue-tied infants did successfully breast-feed at 1 week. Moreover, at 1 month of age, breast-feeding rates for tongue-tie and control infants were similar, if disappointingly low. Other factors not addressed in this study are likely to have more influence on the continuation of breast-feeding at 1 month of age. Therefore, tongue-tie appears to affect the breast-feeding ability of a minority of the affected infants. It is the function, not the appearance, of the tongue that is crucial for successful breast-feeding. Although all the case subjects were tongue-tied, most were able to move their tongues well enough to successfully breast-feed.

A similar conclusion was reached by Messner et al.⁸ In their case control study of 41 infants, 1 of 2 investigators subjectively graded tongue-tie of newborns. Eighty-three percent of tongue-tied infants and 92% of control infants successfully breast-fed for 2 months ($P = .29$). Breast-feeding difficulties were experienced by 25% of the mothers of the tongue-tied infants compared with 3% of the control mothers ($P < .01$).

However, Ballard et al⁴ came to a very different conclusion in their study. One investigator used the ATLFF to identify the babies with tongue-tie. No intra-rater reliability testing was reported. This study found that 3.2% of all infants scored Function Impaired on the ATLFF! At that time, the criteria for the Function Impaired score were different: a function score of ≤ 11 and/or an appearance score of ≤ 8 . All the Function Impaired tongue-tied babies had breast-feeding difficulties—either with poor latch or

Table 4. Survey of Bottle Feeding Mothers at 1 Week and 1 Month Combined

Problems when breast-feeding	Case Subjects (n = 10)	Control Subjects (n = 15)	P Value
Baby always hungry	5 (50.0%)	9 (60.0%)	.225
Nipples extremely sore	8 (80.0%)	10 (66.7%)	NA*
Cracked or bleeding nipples	6 (60.0%)	8 (53.3%)	.317
Breast pain	8 (80.0%)	11 (73.3%)	.617
Breast infection	1 (10.0%)	4 (26.7%)	.072
Not enough milk	2 (20.0%)	7 (46.7%)	.086
When breast-feeding, mother worried baby not growing well	4 (40.0%)	2 (13.3%)	.808
When breast-feeding, baby's doctor worried baby not growing well	2 (20.0%)	1 (6.7%)	NA

* NA, not able to be calculated.

Table 5. Outcomes at 1 Week of Age Based on Initial Newborn Assessment Tool for Lingual Frenulum Function Score

	Function Impaired (n = 5)	Perfect (n = 8)
Bottle-feeding only	2 (40%)	1 (12.5%)
Breast-feeding and bottle-feeding	1 (20%)	3 (37.5%)
Breast-feeding only	0	4 (50%)
Frenotomy followed by breast-feeding only	2 (40%)	0

nipple pain. Only about 32% of tongue-tied infants did not receive a score of Function Impaired on the ATLFF (Ballard JL, personal communication). The disparity in findings between our study and that of Ballard et al is surprising, particularly because both used the ATLFF to attempt to objectively evaluate the infants for tongue-tie.

There clearly is a need for a tool that clinicians can use to identify those tongue-tied infants who are likely to have breast-feeding problems. However, based on our study findings, the ATLFF is not adequate for this purpose. There are many subjective components to this objective measure, which can lead to the differing study results discussed above. These subjective components also contributed to the only moderate inter-rater agreement, despite the investigators being personally trained by the developer of the tool. The average clinician occasionally dealing with a tongue-tied infant is unlikely to perform any better with the tool. In addition, the scoring system is problematic when it completely excludes 63% of the subjects, as in our study. The results shown in Table 5 suggest that the infants with Perfect scores have better outcomes at 1 week than those with Function Impaired scores. However, more infants need to be studied to answer that question definitively.

In Ballard's study, frenotomies were performed on 123 infants without complications.⁴ Latch improved in all cases, and maternal pain levels fell significantly immediately after the procedure. No long-term follow-up was available. The 4 infants in our study who received frenotomies were all breast-fed exclusively at 1 week. At 1 month, 3 were breast-fed exclusively, and 1 was breast-fed and bottle-fed. Our results suggest that, if a frenotomy is to be done, it should be performed before 1 week of age, when significant numbers of tongue-tied infants will have already stopped breast-feeding.

A limitation of our study is the large number of infants lost to follow-up at 1 week and 1 month. As a result, some analyses had few subjects and therefore limited power. The breast-feeding status of the nonrespondents is unknown and may not be similar to that of the respondents.

Because the ATLFF was not a useful tool to determine which tongue-tied infants will have difficulty with breast-feeding, the development and testing of such a tool remains a research priority. In addition, the role of frenotomy in the management of tongue-tied infants with breast-feeding problems needs further evaluation. To date, only case series have been reported of this procedure, with no control groups or long-term follow-up.^{4,10,11} A randomized study of intensive lactation support versus frenotomy would provide better evidence to determine the role of this procedure.

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