

Ankyloglossia, Breastfeeding, and Infant Weight Gain: A Mixed-Methods Study

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BACKGROUND: Ankyloglossia may cause breastfeeding difficulties, potentially leading to early cessation of exclusive breastfeeding (EBF). However, increases in rates of frenotomy have not resulted in increases in EBF rates, and, in some regions, ankyloglossia is not regarded as a major barrier to successful breastfeeding. We sought to elucidate the contribution of untreated ankyloglossia to early cessation of EBF.

METHODS: We conducted a sequential explanatory mixed-methods study at an urban tertiary care hospital for women and children in Hyderabad, India. We evaluated for ankyloglossia and breastfeeding difficulties in mother–infant dyads recruited within 48 hours after birth. Primary outcomes of EBF and infant weight gain were compared between infants with (tongue-tie [TT]) or without (No-TT) ankyloglossia. Semistructured interviews were conducted to identify reasons contributing to early cessation.

RESULTS: A total of 476 dyads were recruited over 3 months. There was no significant difference in rates of EBF at 6 months (No-TT 81.4% vs TT 78.6%; $P = .6$) or infant weight gain velocity from 0 to 3 months (No-TT 26.3 g/d vs TT 26 g/d; $P = .86$) or from 3 to 6 months (19.3 g/d vs 20.4 g/d; $P = .49$). Maternal concern for inadequate supply was associated with lower EBF odds at 6 months (odds ratio 0.22; $P = .003$). Mothers with early cessation in either group rarely reported issues with infant breastfeeding skill or nipple pain.

CONCLUSIONS: Although ankyloglossia may affect breastfeeding experiences, ankyloglossia alone does not appear to affect breastfeeding maintenance or infant weight gain. Improving breastfeeding outcomes should include multidisciplinary management to focus on all potential causes and not only ankyloglossia.

abstract



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Dr Raol conceptualized and designed the study, designed the data collection instruments, collected data, coordinated and supervised data collection, drafted the initial manuscript, and critically reviewed and revised the manuscript. Ms Silamkoti collected data, carried out the initial analyses, and critically reviewed and revised the manuscript. Dr Syed collected data and critically reviewed and revised the manuscript. Ms Hosek carried out the initial analyses and critically reviewed and revised the manuscript. Dr Theetla and Dr Madireddy conceptualized and designed the study and critically reviewed and revised the manuscript for important intellectual content. All authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

WHAT'S KNOWN ON THIS SUBJECT: Ankyloglossia interferes with breastfeeding in some children and may increase the risk of early cessation of breastfeeding. Lingual frenotomy can result in improved short-term outcomes such as decreased nipple pain and improved maternal breastfeeding self-efficacy.

WHAT THIS STUDY ADDS: Untreated ankyloglossia in itself, particularly if mild to moderate, may not result in early breastfeeding cessation or poor infant weight gain. Maternal concerns regarding breastmilk supply are associated with early cessation regardless of presence of ankyloglossia.

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INTRODUCTION

Ankyloglossia, or tongue-tie (TT), occurs in 8% of the pediatric population aged less than 1 year,¹ with reported prevalence ranging from 0.5% to 21% from infancy to adolescence.^{2–13} The impact of ankyloglossia can be wide reaching. In a meta-analysis of 39 studies of mother–infant dyads presenting to a clinician for evaluation of ankyloglossia, approximately one-half reported experiencing breastfeeding difficulties such as a poor latch, prolonged feeds or frequent feeding, poor weight gain, and even failure to thrive.¹⁴ These same mothers reported nipple pain (30%) and early undesired weaning (20%), with a weighted mean breastfeeding self-efficacy score of 43.7, which is well below the “at-risk for early cessation” cutoff of 50.¹⁴ However, these data are from self-selected groups of patients seeking care for ankyloglossia; therefore, the broader impact of TT on the population is not known.

Frenotomy is the definitive surgical treatment of ankyloglossia and has been demonstrated to be a valuable procedure with improvement in a number of breastfeeding-related feeding measures following intervention.^{15–19} Importantly, most existing studies focused on pre-/postprocedural assessment without comparison with those who had no surgical intervention. Therefore, it is not clear which, if any, of these children would have improved without treatment. Indeed, in Messner’s 2000 study, 30 out of 36 children with untreated ankyloglossia were still breastfeeding at 2 months compared with 33 out of 36 children without ankyloglossia.²⁰ However, this study had a limited 2-month follow-up period and did not specify whether it tracked the incidence of any degree of breastfeeding or specifically the incidence of exclusive breastfeeding (EBF). Therefore, the impact of untreated ankyloglossia on long-term breastfeeding outcomes remains unclear.

Given continued uncertainty surrounding the impact of untreated ankyloglossia, we undertook a sequential explanatory mixed-methods study to evaluate the impact of untreated ankyloglossia on EBF maintenance rates at 6 months (EBF6) compared with those without ankyloglossia. As a secondary objective, we assessed weight gain and velocity of weight gain between the 2 cohorts. Finally, using qualitative interviews, we identified maternal experiences that correlated with early cessation of breastfeeding in both cohorts. We hypothesized that those with ankyloglossia would have higher rates of early cessation and slower weight gain than those without ankyloglossia.

METHODS

Data Collection

Participant Enrollment

Between October and December 2023, a convenience sample of study patients were recruited from mother–infant

dyads who arrived for infant vaccination within 48 hours after birth in the postpartum inpatient setting at Niloufer Hospital for Women and Children in Hyderabad, Telangana, India. Owing to the high volume of deliveries, use of paper charts, and open ward setting at this hospital in which patients frequently change bed locations, it was not possible to approach all dyads. Verbal consent was obtained from mothers for study participation. Ethics committee approval from Osmania Medical College and institutional review board approval from Emory University were obtained prior to study commencement.

Inpatient Assessment

Following enrollment, the principal investigator (N. R.) performed evaluation of the infant’s oral cavity. The Bristol Tongue Assessment Tool (BTAT)²¹ was used to grade the infant’s tongue. The scale ranges from 0 to 8, with a score of 0 to 3 indicating more severe reduction of tongue function,²¹ and a score of 8 indicating no ankyloglossia. For this study, any infant whose BTAT score was less than 8 was considered to have ankyloglossia. A BTAT less than or equal to 5 has been shown to be associated with a higher likelihood of frenotomy.²² The mother or the infant’s additional caregiver was queried regarding breastfeeding difficulty. If breastfeeding difficulty for any reason was endorsed (ie, difficulty latching, concerns regarding supply, uncertainty regarding position), the study team visited the mother–infant dyad following oral evaluation, and lactation counseling was provided by a study team member (S. M. S.) with specific training in breastfeeding medicine and lactation. The lactation consultant was aware of the ankyloglossia status prior to counseling. However, because ankyloglossia evaluation was not part of the standard of care in this region and the investigators did not want to introduce bias, mothers were not informed of the infant’s ankyloglossia status after evaluation. Prior to discharge, demographic information, including mother’s age, pregnancy and delivery history, perinatal history, method of delivery, infant gestational age, infant birthweight, and delivery complications, were extracted from the medical record.

Follow-up

Following discharge, mothers were contacted by a study team member who was blinded to the ankyloglossia status and fluent in the mother’s primary language (ie, Telugu or Hindi) at 3 postdelivery time points: 2 to 4 weeks, 3 months, and 6 months. At each follow-up, mothers were asked 7 questions regarding breastfeeding and weight gain (Supplemental Material Appendix A). The mother was then counseled on topics such as frequency of feeding, mother’s diet, expected infant weight gain, and strategies to improve supply.

Qualitative Interviews

Following the 3- and 6-month follow-up phone calls, 9 mothers from each group (4 groups total: +TT/+EBF6, +TT/-EBF6, -TT/+EBF6, -TT/-EBF6) were selected for interviews through purposive sampling. Semistructured interviews via telephone (Supplemental Material Appendix B) lasting less than 30 minutes were conducted by one researcher (S. B.) trained in qualitative research and interview techniques who was a native speaker of both Telugu and Hindi, primary languages of participants, but who was not involved in the patient's medical care. Prior to beginning the interview, all caregivers were reminded of the study design and aims, and verbal consent for an audio-recorded interview was confirmed. Audio recordings of the interviews were transcribed and translated into English by hand by the interviewer. The interviewer also recorded field notes during and after each interview to document impressions. Because thematic saturation, defined by the point at which new interviews failed to generate novel codes or information, and future interviews would be unlikely to yield new themes,²³ was achieved with 9 participants in each group, no further interviews were conducted.

Data Analysis

Quantitative Data Analysis

Demographic characteristics were summarized with mean (SD) or proportions, and differences in categorical and continuous demographic variables were tested with a χ^2 test and Student *t* test, respectively. Rates of EBF at follow-up visits were calculated and compared using a χ^2 test. Infant growth velocity was calculated by taking the change in weight between the time points of interest and dividing by the number of days between time points. Differences in growth velocity were compared using a Student *t* test. Logistic regression was performed to identify variables that were associated with early cessation of breastfeeding. Statistical analysis was performed using Stata/SE (version 16, StataCorp LLC).

Sequential Explanatory Mixed-Methods Analysis

To explore reasons for early cessation of breastfeeding, qualitative data analysis was performed using a hybrid of inductive and deductive methods. Interview transcripts were coded and queried using Taguette, a free and open-access software program designed to facilitate the management of qualitative data. First, all coded data were transferred to Excel (2024 version for Mac, Microsoft Corporation). An initial sample of 4 interviews were inductively coded by 2 separate analysts (S. B. and N. R.), each of whom developed their own unique codes and codebook. A composite codebook was created by S. B. and approved by other members of the research team. Subsequent interviews were deductively coded using the composite

codebook. Four randomly selected interviews were double-coded at various time points to ensure continued concordance among the analysts. Strong intercoder consistency was determined by ensuring that the study included all 8 aspects of intercoder reliability in qualitative research, as described by Cofie et al.²⁴ The codebook was a dynamic document that was updated and refined to accommodate additional codes and themes identified in successive interviews. The team frequently returned to the transcripts for content familiarization. Transcript coding and thematic analysis was periodically reviewed by the entire research team to confirm consistency and resolve any disagreements.

RESULTS

Patient Demographics

A total of 476 dyads were included in the study, with 366 (76.9%) dyads in the no-ankyloglossia (No-TT) cohort and 110 (23.1%) in the ankyloglossia (TT) cohort (Table 1). For those with TT, only 2 out of 110 had severe restriction of tongue function with a BTAT less than or equal to 3, but 33 out of 110 had a BTAT less than or equal to 5 (Table 1). The median age of mothers at the time of delivery was 24 years, with no significant difference in number of prior children, weeks of gestation at time of birth, method of delivery, or comorbidities (Table 1). Infants in the TT cohort were significantly more likely to be of male sex, consistent with prior reports,¹⁴ but birthweight was similar across cohorts (Table 1). The number of participants in each time point varied, with 51.5% follow-up at 2 to 4 weeks, 64.3% at 3 months, and 70% at 6 months. A total of 34.5% (164 out of 476) of dyads had data for all 3 follow-up time points.

Evaluating the Impact of the Presence of Ankyloglossia on Breastfeeding Outcomes

Given prior data suggesting that ankyloglossia leads to early cessation of EBF,¹⁴ we first assessed rates of EBF at 3 chronological age-based time points. We found no differences at the 2- to 4-week (No-TT 88.5% vs TT 92.6%, 95% CI for difference: -12.4% to 4.2%; *P* = .39), 3-month (84.6% vs 77.7%, 95% CI for difference: -3.8% to 17.5%; *P* = .18), or 6-month (81.4% vs 78.6%, 95% CI for difference: -7.9% to 13.5%; *P* = .6) follow-up phone calls (Figure 1). When looking specifically at dyads with TT, no difference emerged in ankyloglossia severity based on the BTAT between infants who were or were not exclusively breastfed at 6 months (mean [SD] 5.93 [1.02] vs 5.87 [0.74], mean difference -0.61, 95% CI: -0.62 to 0.5; *P* = .78).

When looking specifically at those dyads with complete data, rates of EBF were similar at each time point: 2- to 4-week (No-TT 90% vs TT 94.1%, 95% CI for difference: -13.6% to 5.3%; *P* = .46), 3-month (86.2% vs 76.5%, 95% CI for difference: -5.8% to 25.1%; *P* = .17), or 6-month

TABLE 1. Mother–Infant Dyad Demographics				
Participant Characteristic	No Ankyloglossia (n = 366)	Ankyloglossia (n = 110)	Total (N = 476)	P value
Mother's age				.14
Mean (SD)	24.9 (4.15)	24.4 (4.24)	24.8 (4.17)	
Median (IQR)	24 (10, 42)	24 (18, 37)	24 (10, 42)	
Number of live births				.51
Mean (SD)	2.0 (1.03)	1.9 (0.95)	2.0 (1.01)	
Median (IQR)	2 (1, 7)	2 (1, 5)	2 (1, 7)	
Method of delivery, n (%)				.77
Cesarean section	206 (57.9)	63 (59.4)	269 (58.2)	
Spontaneous vaginal delivery	150 (42.1)	43 (40.6)	193 (41.8)	
Amniotic fluid index				.36
Mean (SD)	12.3 (4.86)	12.9 (4.76)	12.4 (4.84)	
Median (IQR)	12.0 (1.5, 29.5)	12.5 (4.5, 32.5)	12.3 (1.5, 32.5)	
Maternal comorbidities, n (%)				
Hypothyroidism	57 (15.6)	16 (14.5)	73 (15.3)	.79
Anemia	15 (4.1)	1 (0.9)	16 (3.3)	.1
Hypertension	6 (1.6)	4 (3.6)	10 (2.1)	.2
Diabetes mellitus	2 (0.5)	2 (1.8)	4 (0.8)	.2
Gestational hypertension	17 (4.6)	4 (3.6)	21 (4.4)	.65
Gestational diabetes	4 (1.1)	3 (2.7)	7 (1.5)	.21
Baby's gestational age at birth, number of weeks				.86
Mean (SD)	38.3 (1.45)	38.3 (1.51)	38.3 (1.46)	
Median (IQR)	38.4 (32.9, 41.9)	38.6 (33.7, 40.4)	38.4 (32.9, 41.9)	
Baby's birthweight in kg				.91
Mean (SD)	2.7 (0.41)	2.7 (0.36)	2.7 (0.40)	
Median (IQR)	2.7 (1.6, 4.3)	2.7 (2.0, 3.8)	2.7 (1.6, 4.3)	
Male sex of baby, n (%)	166 (45.7)	73 (68.9)	239 (51.0)	<.001 ^a
BTAT score, n (%)				
2	n/a	1 (0.9)	1 (0.9)	n/a
3	n/a	1 (0.9)	1 (0.9)	n/a
4	n/a	7 (6.3)	7 (6.3)	n/a
5	n/a	24 (21.4)	24 (21.4)	n/a
6	n/a	46 (41.1)	46 (41.1)	n/a
7	n/a	31 (27.7)	31 (27.7)	n/a
8	366 (100)	n/a	366 (100)	n/a
Abbreviations: BTAT, Bristol Tongue Assessment Tool; n/a, not applicable.				
^a P < .05.				

(82.3% vs 73.5%, 95% CI for difference: −7.4% to 25%; $P = .25$).

To further assess the relationship between ankyloglossia and cessation of EBF, we performed multivariable logistic regression. Other variables examined in the model included pain with breastfeeding reported at the 2- to 4-week phone call, concern regarding milk supply reported at the 2- to 4-week phone call, number of previous births, method of delivery, and skin-to-skin contact in the delivery room. The only variable that was associated with a lower odds

of EBF6 was maternal concern regarding milk supply reported at the 2- to 4-week phone call (odds ratio [OR] 0.22, 95% CI: 0.083–0.58; $P = .003$; Table 2).

To examine whether ankyloglossia was associated with differential weight gain for infants who breastfed, we next compared weight gain velocity between groups. Again, there was no difference in weight gain velocity between exclusively breastfed infants in the 2 cohorts at the 3-month (No-TT 26.3 g/d vs TT 26 g/d, 95% CI for difference: −2.63 to 3.14; $P = .86$) and 6-month (19.3 g/d vs 20.4 g/d,

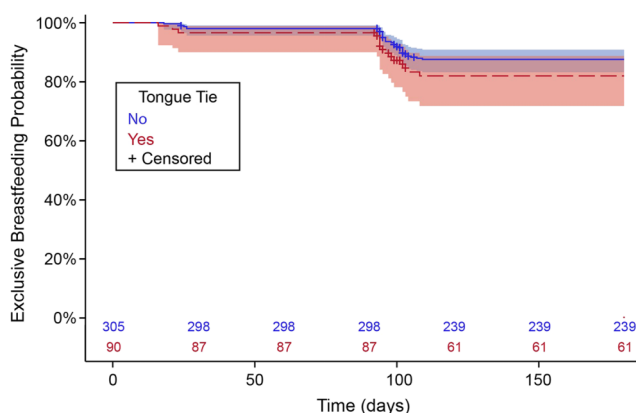


Figure 1.
Exclusive Breastfeeding Maintenance Probability

95% CI for difference: -4.3 to 2.08 ; $P = .49$) follow-up phone calls (Figure 2). Both groups exhibited an expected drop in growth velocity between the 0- to 3-month and 3- to 6-month measures (-7.8 g/d vs -6.4 g/d, 95% CI for difference: -6.8 to 3.9 ; $P = .59$).

Evaluating the Impact of Ankyloglossia on Maternal Concerns

Because mothers of children with ankyloglossia report maternal pain with breastfeeding and concerns regarding milk supply as reasons to undergo frenotomy,¹⁴ we also evaluated rates of each of these concerns in both groups. At the 2- to 4-week follow-up, there was no statistical difference between the No-TT and TT cohorts for maternal pain with breastfeeding (7% vs 11.1%, 95% CI for difference: -13.6% to 5% ; $P = .3$); however, when looking only at those who were exclusively breastfeeding, the difference was slightly greater between the 2 groups, although still not statistically significant (6% vs 12.2%, 95% CI for difference: -16.1% to 3.5% ; $P = .14$). Interestingly, of those in the TT cohort who were not exclusively breastfeeding at the 2- to 4-week follow-up (4 out of 54), none of them reported pain with breastfeeding. At the 3-month follow-up, was

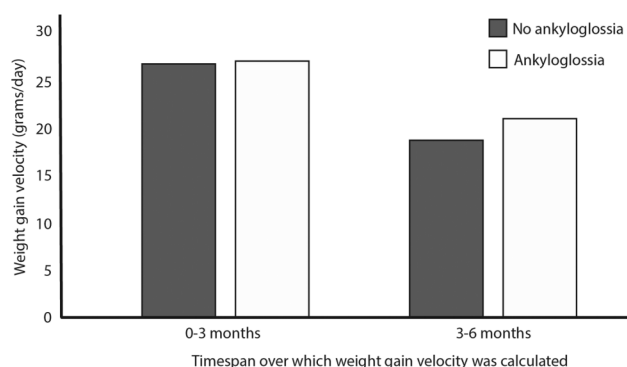


Figure 2.
Weight Gain Velocity

there no difference between the No-TT and TT cohorts for maternal pain with breastfeeding among all mothers who were breastfeeding to any extent (3.1% vs 1.4%, 95% CI for difference: -1.7% to 5.2% ; $P = .43$), as well as among those who were exclusively breastfeeding (1% vs 1.9%, 95% CI for difference: -4.7% to 3% ; $P = .62$).

Given the clinically significant but not statistically significant difference in pain at first follow-up between the 2 cohorts, we performed multivariable regression to evaluate for correlation between ankyloglossia and pain with breastfeeding at the 2- to 4-week follow-up. Other variables examined in the model included mother's age, number of previous births, method of delivery, and skin-to-skin contact in the delivery room. Ankyloglossia was not associated with pain with breastfeeding in our cohort (OR 1.35, 95% CI: 0.45–4.1). Furthermore, concerns regarding low milk supply at neither the 2- to 4-week follow-up nor the 3-month follow-up varied between the No-TT and TT cohorts (2–4 week: 13.8%, 95% CI: 8.8%–18.7% vs 8.9%, 95% CI: 1.5%–16.4%; 3-month: 12.5%, 95% CI: 8.4%–16.6% vs 14.7%, 95% CI: 6.7%–22.7%).

Qualitative Findings and Mixed-Methods Analysis

A total of 36 mothers were interviewed. The data are presented in 4 broad categories, with corresponding themes that emerged from the data. These include the following: (1) infant-related factors; (2) maternal factors; (3) family-related factors; and (4) system-related factors. Major themes, codes, and representative quotations for each category are presented in Table 3.

Given the lack of difference between groups in previously noted areas of concern, we followed the thread of early cessation of EBF to explore potential reasons for early cessation across the TT and No-TT cohorts. Specifically, we explored responses to the question “What kinds of challenges may impact your breastfeeding?” for the 18 mothers who had early cessation. Investigator probes included working outside the house, household demands, poor milk production, and lack of breastfeeding counseling. In both

Effect	OR	95% CI	P Value
Concern regarding milk supply reported at 2- to 4-wk phone call	0.22	(0.08–0.56)	.003 ^a
Pain with breastfeeding	0.86	(0.24–3.09)	.81
Method of delivery	1.73	(0.76–3.96)	.2
Mother's age	1.00	(0.90–1.11)	.98
Number of live births	1.04	(0.68–1.59)	.87
Prematurity	0.71	(0.25–2.05)	.53
Skin-to-skin contact at birth	1.22	(0.54–2.75)	.63

Abbreviations: EBF, exclusive breastfeeding; OR, odds ratio.
^a $P < .05$.

TABLE 3. Qualitative Interview Results						
Themes	Associated Codes	+TT/+EBF6	+TT/−EBF6	−TT/+EBF6	−TT/−EBF6	Quotations
Infant-related factors	Latch issues	0/9 (0%)	1/9 (11%)	0/9 (0%)	1/9 (11%)	+TT/−EBF6: “He is unable to keep the nipple inside his mouth. Many times, he will cry when I breastfeed him... Madam, I have enough milk, but I don't know why my baby is unable to drink the milk properly. <i>My milk is flowing but he is not drinking...he makes some sounds while drinking my milk and he catches the nipple for some time, and later he starts crying.</i> ” −TT/−EBF6: “There is no proper latching... whenever I move him close to the breast, he pulls off the nipple frequently, he cannot suck... <i>I feel my nipples are sunken, and that might be the reason he is unable to suck the milk.</i> ”
	Fussiness with feeding	0/9 (0%)	1/9 (11%)	0/9 (0%)	1/9 (11%)	+TT/−EBF6: “I try to breastfeed them, [and] they will cry and pull off the nipple...”
Maternal factors	Insufficient milk production/perceived infant hunger	0/9 (0%)	8/9 (89%)	0/9 (0%)	9/9 (100%)	+TT/−EBF6: “After my delivery I was not able to make milk for 4–5 days, I informed it to doctor then she said, ‘No problem even if you are unable to make milk just try breastfeeding your baby, after few days milk will be produced, need not to worry about this, even a drop of your milk gives much strength to your baby...’ <i>Then I asked doctor to prescribe some formula milk because [my] baby was crying with hunger, [and] I was unable to see him like that.</i> ” −TT/−EBF6: “We didn't [go to a clinic], <i>we only decided to feed because there was no sufficient milk, baby was crying for more milk, so we started feeding the buffalo milk.</i> ”
	Back to work	0/9 (0%)	0/9 (0%)	1/9 (11%)	0/9 (0%)	−TT/+EBF6: “Sometimes I take a break and go home to feed the baby, my home is just 2 km away from my office... <i>I will also go to home at least 3 times by taking breaks, even my staff support me.</i> ” +TT/−EBF6: “ <i>I have to look after the baby, I don't go out much, so I don't know anything. I'm not very educated [so] no one will give any job...I'm also not interested.</i> ”
	Maternal pain/nipple issues	1/9 (11%)	1/9 (11%)	1/9 (11%)	1/9 (11%)	−TT/+EBF6: “I do not have any pain now, but [the first] <i>2 months I had pain beside the nipples.</i> ” +TT/+EBF6: “Now I don't have any pain madam, but <i>after delivery until 2–3 months definitely we will have pain.</i> ”
	Mother's disappointment/sadness	0/9 (0%)	3/9 (33%)	0/9 (0%)	5/9 (56%)	+TT/−EBF6: “I feel bad sometimes but what to do madam, for this baby there is no milk, <i>nothing in my hands now, what is written on fate that will happen.</i> ” −TT/−EBF6: “ <i>Sometimes I feel very sad but my mother-in-law tells me you are also breastfeeding and bottle feeding her so she will be healthy no problem in that.</i> ” +TT/+EBF6: “I'm happy madam, because after marriage I [had] not conceived until 4 years. I waited many years to have kids, now I have 3 kids, 3 kids are healthy, I'm also healthy. I'm happy madam.”
Family-related factors	Appropriate family advice	9/9 (100%)	6/9 (67%)	9/9 (100%)	8/9 (89%)	−TT/+EBF6: “Everyone will say to continue the feeding until 2 years.”
	Inappropriate family advice	0/9 (0%)	3/9 (33%)	1/9 (11%)	2/9 (22%)	+TT/−EBF6: “Everyone said I'm not making enough milk so it's not good for [the] baby to [drink] 2 types of milk, so my relatives suggested don't give your milk...if you give 2 types of milk, [the] baby may face some problems, they told.” −TT/−EBF6: “My mother said there are many women who are not breastfeeding the baby...if you get the enough milk you can breastfeed. You are not getting what you will do for that? Don't roam around [to] the doctors.”

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TABLE 3. Qualitative Interview Results (Continued)						
Themes	Associated Codes	+TT/+EBF6	+TT/−EBF6	−TT/+EBF6	−TT/−EBF6	Quotations
	Joint family support	9/9 (100%)	7/9 (78%)	7/9 (78%)	8/9 (89%)	+TT/+EBF6: "Ours is a joint family. My mother-in-law is there, we are 3 co-sisters, I'm the last one, so if [I] have any doubts I will ask them. Yes, as I told you <i>we are 4 ladies in the house, everyone is there to take care of my child, my husband is also very supportive, my in-laws will also take care of me and my baby.</i> "
System-related factors	Appropriate physician support for breastfeeding (of those who received advice from a physician)	4/4 (100%)	3/9 (33%)	5/5 (100%)	3/9 (33%)	+TT/−EBF6: "After my delivery I was not able to make milk for 4–5 days. I informed it to the doctor, and she said, <i>No problem, even if you are unable to make milk just try breastfeeding your baby, after few days milk will be produced. You need not worry about this, even a drop of your milk gives much strength to your baby.</i> "
	Physician recommended formula	N/A	6/9 (67%)	N/A	7/9 (78%)	−TT/−EBF6: "Last time when we visited, <i>the doctor told me to use this formula milk for me and my baby...even after consuming the formula milk, I don't have the sufficient milk to feed baby.</i> "
	Appropriate grassroots health care worker breastfeeding support (pre-/postnatal)	3/9 (33%)	2/9 (22%)	3/9 (33%)	1/9 (11%)	+TT/+EBF6: " <i>They will conduct meetings [and] they will tell how to take care of [and feed] the baby, what food to give after 6 months, they will check weight. They will give food also; when I was pregnant, they gave me eggs, milk, rice, curry, curd, dal. Every day they will give vegetable curry, but on Wednesday they give egg curry. Now I'm getting eggs and milk, they are giving balamrutham [vitamins for baby].</i> "
Abbreviations: EBF, exclusive breastfeeding; TT, tongue-tie.						

the TT and No-TT cohorts who had early cessation, all mothers were housewives and did not work outside the home. With regards to taking care of household demands, 6 out of 9 participants from the TT cohort and 8 out of 9 from the No-TT cohort had significant support from either the mother or mother-in-law, living in a multigenerational household ("joint family"). Inadequate supply was a driving reason for early cessation for nearly all participants, with 8 out of 9 in the TT cohort and 9 out of 9 in the No-TT cohort reporting inadequate milk supply. In the TT cohort, the 1 mother who did not report issues with milk supply stated "He is unable to keep the nipple inside his mouth. Many times, he will cry when I breastfeed him...Madam, I have enough milk, but I don't know why my baby is unable to drink the milk properly. My milk is flowing but he is not drinking."

Only 2 out of 9 participants in each cohort reported issues with infant feeding such as difficulty latching or fussiness while feeding. Only 1 out of 9 participants in each cohort noted nipple pain with feeding. A total of 6 out of 9 participants in the TT cohort and 7 out of 9 in the No-TT cohort reported being recommended formula by an outpatient doctor, without breastfeeding counseling, when they presented with concerns after discharge. With regards to breastfeeding advice given from the government community health workers (Accredited Social Health Activist or rural child center, ie, Anganwadi, workers), 2 out of 9 in the TT cohort and 1 out of 9 in the No-TT cohort reported receiving counseling from these government-provided

health care resources. Interestingly, for those without early cessation, one-third did receive breastfeeding education from these services, whether in prenatal classes or postnatally during routine follow-up visits. When asked who they turn to first for breastfeeding advice, 5 out of 9 in the TT cohort and 6 out of 9 in the No-TT cohort stated that they turn to a female figure in their lives, including their mother, sister, grandmother, or mother-in-law. With the help of these female figures, some women reported accepting the inability to breastfeed as their fate: "My mother said there are many women who are not breastfeeding the baby. If you get enough milk you can breastfeed, [but] you are not getting [enough]...Don't roam around [to different] doctors...she told, so that's the reason I didn't visit other doctors. I prayed to many gods but still it is like that...if it was in my hands I can do something, now also baby is healthy with the formula milk, I don't feel sad anymore."; "...but my mother-in-law tells me you are also breastfeeding and bottle feeding her so she will be healthy...no problem in that."

Overall, reasons for early cessation appeared similar in both groups and were primarily related to inadequate supply and seemingly insufficient education surrounding breastfeeding.

DISCUSSION

In this study, we found that ankyloglossia was not a primary factor in maintenance of breastfeeding or in infant growth velocity for our study population. These findings suggest

that when caring for mother–infant dyads with breastfeeding difficulties, a multitude of potential causes should be explored, regardless of the presence or absence of ankyloglossia. Perhaps more importantly, findings inform the interpretation of existing ankyloglossia studies without a control group, as well as how to design high-quality studies on ankyloglossia moving forward. Although there is clearly benefit of frenotomy for breastfeeding difficulties in infants with symptomatic ankyloglossia,¹⁵ without the inclusion of control groups for comparison, it is unclear whether the same improvement may have been seen over time without frenotomy. This possibility is exemplified by nipple pain in our population, a symptom that is commonly believed to be fixed following frenotomy, but that was common in mothers of babies both with and without ankyloglossia and that improved with time in both cohorts. In fact, nipple pain is common among breastfeeding mothers overall and improves to mild levels by days 7 to 18 postpartum.^{25,26} Our data suggest that symptom improvement with time only, even in the setting of ankyloglossia, should be considered when interpreting post-frenotomy data.

When focus is placed only on ankyloglossia, a host of other causes for early breastfeeding cessation may be missed. Mothers in our study discussed concern for poor supply as a frequent reason for early cessation, both in quantitative and qualitative exploration. They also reported that formula was frequently recommended by a physician without further evaluation of causes of breastfeeding difficulty. When multidisciplinary evaluation, including a clinician with expertise in infant feeding and counseling, are provided to patients evaluated for ankyloglossia and breastfeeding difficulties, rates of frenotomy decrease.^{27–29} This is not to say that no infants need frenotomy; it is possible that some dyads in our cohort may have avoided early cessation had the infant undergone frenotomy. However, given the complex multifactorial nature of breastfeeding, our data suggest that evaluating mothers and infants as a dyad and not only looking for the presence of ankyloglossia is the key to both appropriately choosing candidates for frenotomy and improving EBF rates.

If we shift focus away from whether ankyloglossia is present, a feasible target to support breastfeeding mothers would be to empower mothers and those who take care of mothers through breastfeeding education. In our study, physicians were often the ones who recommended formula for mothers who were having breastfeeding difficulty. Although mothers had frequent touchpoints with community health workers, many reported that breastfeeding counseling was not part of the routine support offered by the workers. By training health care workers regarding how to support and educate breastfeeding mothers, as well as regarding when to refer to feeding experts, including those who manage ankyloglossia when other techniques fail, early cessation may decrease simply from improved self-efficacy.

Importantly, maternal breastfeeding self-efficacy is the one of the key factors in EBF maintenance.^{30–32} In addition, although health care workers are key to supporting breastfeeding mothers who are having difficulty, in our population, family members, especially female members, were the *initial* source of advice. Consistent with findings from other qualitative studies evaluating stakeholders' perspectives and perceived barriers to breastfeeding,^{33,34} our data support incorporation of breastfeeding education to family members caring for the mother, as well as into the routine care provided by community health workers.

Although our study sheds light on the impact of ankyloglossia and potential future study design, it is not without limitations. First, the BTAT, like all published tools currently used to assess the severity of ankyloglossia, has not been validated, as pointed out in the recent American Academy of Pediatrics Clinical Report.²⁵ This tool was chosen for practical purposes; its ease of use and accompanying assessment time make it a commonly used tool in otolaryngologic clinics. Second, few of our infants had severe ankyloglossia; therefore, the entire spectrum of the condition was not equally represented. However, given that there is more uncertainty surrounding treating less-severe ankyloglossia, our findings are important in the literature because they demonstrate that many of these dyads successfully breastfeed without intervention. Third, we had moderate loss-to-follow-up for the primary outcome of EBF and high loss-to-follow-up for the secondary outcome of infant weight gain velocity, which may have been biased toward those with more difficulties. However, we still had a reasonable number of participants relative to prior studies with shorter-term follow-up.²⁰ Fourth, we relied on mothers' reports because of difficulty with time and cost associated with travel. It is possible that they gave inaccurate reports to appear more favorable to the members of the research team, as is the case with social desirability bias. Fortunately, moderate accuracy has been noted between actual practice and maternal report of infant and young child feeding in India.³⁵ Additional studies and public health efforts with greater resources should incorporate more objective measurements recorded by health care workers as part of the assessment of effective breastfeeding. Fifth, our study design did not control for the myriad factors that can impact breastfeeding, including anatomic differences such as maternal breast and nipple shape/size and infant craniofacial structure. Therefore, we recognize that these findings cannot be extrapolated to every infant with ankyloglossia. Finally, our results may not be generalizable to other populations because our cohort included mostly non-working mothers in a demographic and culture in which breastfeeding is the norm and expectation. In societies in which issues such as a mother's autonomy or working obligations are considered a higher priority, nuclear families are more common, or pain is viewed differently, the

decision to breastfeed may be thought of more as an individual decision and not the assumed norm. In these cases, rates of EBF may be more heavily influenced by the presence of ankyloglossia because of its impact on breastfeeding self-efficacy.

CONCLUSIONS

Although ankyloglossia may impact breastfeeding for some mother–infant dyads, our findings indicate that other factors play a bigger role in successful breastfeeding, whether a TT is present. Future research should include randomized-controlled trials with both early frenotomy vs strong breastfeeding support, as well as further observational studies across various cultures. In addition, more objective data to help determine who is an appropriate candidate for

frenotomy can aid feeding teams in deciding who would benefit from intervention.

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ABBREVIATIONS

EBF: exclusive breastfeeding
TT: tongue-tie
OR: odds ratio

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