Tongue-tie: incidence and outcomes in breastfeeding after lingual frenotomy in 2333 newborns

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Aim: *To determine the prevalence of ankyloglossia in newborns with breastfeeding problems and to assess the effectiveness of frenotomy in the maintenance of exclusive breastfeeding at 1 month, 3 months and 6 months in newborns at an Andorran Hospital.* ***Study design:*** *A descriptive, cross-sectional, population-based, retrospective study of newborns over a 5-year period (2016–2020) was performed. Nine medical history variables (presence or absence of ankyloglossia and type of frenulum, surgical intervention or not, first degree hereditary component, gender, Rh and blood group, type of breastfeeding, causes of cessation and duration of breastfeeding) related to perinatal and feeding history were collected confidentially and anonymously. The Coryllos classification was used for the diagnosis of ankyloglossia. Descriptive analysis of the data, Chi-square test and prevalence ratios were calculated.* ***Results:*** *A total of 2333 newborns were included in the study (50.02% males and 49.98% females). The prevalence of ankyloglossia was 7.84% (n = 183). Of the infants examined, 136 underwent lingual frenotomy. The number of infants who maintained exclusive breastfeeding, both surgically and non-surgically treated, was no statistically significant differences at 1 month (p = 0.65), 3 months (p = 0.61) and 6 months (p = 0.49).* ***Conclusions:*** *Lingual frenotomy was only performed on patients with ankyloglossia associated with ineffective suction that causes BF difficulties. The realization or not of frenotomy was not a determining factor for the maintenance of breastfeeding at 1 month, 3 months and 6 months. On the contrary, it was a determining factor for the prolongation of mixed feeding. Ankyloglossia related to breastfeeding difficulties should be treated by a multidisciplinary team.*

Keywords: *Ankyloglossia, Newborns, Frenotomy, Breastfeeding*

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INTRODUCTION

The lingual frenulum is a dynamic structure often referred to as a "cord" or "submucosal band" of connective tissue[1]. This mucosa extends from the ventral surface of the tongue, in the midline, to the floor of the mouth, securing its movements. Ankyloglossia is defined as a limitation of the possibilities of protrusion and elevation of the tongue tip due to the shortness of the frenulum and/or genioglossus muscles[2].

The prevalence of ankyloglossia in infants varies from 4 to 11%, depending on the study population and the criteria used to define it[3–6]. It is more frequent on males, in a proportion male:female of 1.5:1 to 2.6:1[3]. Although most cases of ankyloglossia are sporadic, it can be associated with some syndromes, such as Ehlers-Danlos, Ellis-Van Creveld, Pierre Robin, digital facial gold, infantile hypertrophic pyloric stenosis or X-linked cleft palate[7,8]. Cases of autosomal dominant and recessive inheritance have been documented, finding newborns with mostly first-degree family history[9].

The tongue influences the growth and development of the

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oral cavity and also functional processes such as chewing, sucking, swallowing and speech[9]. Due to its high impact, a good diagnosis of ankyloglossia is imperative to help early correction and thus promote a better prognosis. Diagnostic criteria for ankyloglossia are controversial due to the lack of a universally accepted definition and objective diagnostic criteria. The most widely used is the Hazelbaker descriptive assessment tool (Assessment Tool for Lingual Frenulum Function (HATLFF))[10,11]; however, this took is not validated. Coryllos and Kotlow made an anatomical classification of the sublingual frenulum, dividing it into four types identifiable by palpation[12–14]. Depending on where the frenulum is anchored to the tongue, a distinction is made between anterior frenulum, which accounts for 75% of ankyloglossia, and posterior frenulum, which is more difficult to evaluate with the naked eye and requires manual inspection[8].

The clinical presentation of symptomatic ankyloglossia in the infant includes painful breastfeeding, inability to maintain an effective latch and, consequently, poor weight gain[13]. Most infants with short lingual frenulum can breastfeed without difficulty[3]; however, breastfeeding problems are reported more frequently among infants with ankyloglossia than with no[3,5]. There is a disparity of opinion as to whether it should be treated conservatively or surgically. Some authors advocate a multidisciplinary approach with breastfeeding (BF) sessions together with myofunctional therapy before performing the surgical technique of frenotomy and, if necessary, to prevent adhesions[7,11,13].

Taking into account the benefits of BF[15], the World Health Organization (WHO) in association with the United Nations Foundation (UNICEF) advises exclusive BF until 6 months of age, it is important for the health professional to address any condition that may be detrimental to BF. Early detection is important to prevent BF problems. Therefore, the purpose of this study was to determine the prevalence of ankyloglossia, to describe the prevalence of ankyloglossia in newborns with BF problems and to assess the effectiveness of lingual frenotomy in the maintenance of exclusive BF at 1 month, 3 months and 6 months of life in newborns at the only Andorran Hospital, Europe.

MATERIALS AND METHOD

Study design and population

This descriptive, cross-sectional, population-based retrospective study was evaluated and approved by the Clinical Research Ethics Committee of the Universitat Internacional de Catalunya, Barcelona (ODP-ECL-2020-01) and registered in ClinicalTrials.gov (Identifier: NCT04703946). It was conducted at the "Hospital Nostra Senyora de Meritxell" (the only hospital in Andorra, Europe), evaluating the prevalence of ankyloglossia, surgical technique of frenotomy and its relationship with the duration of exclusive BF at 30 days, 3 and 6 months in all newborns in this country from January 2016 to December 2020.

For the estimation of the prevalence of ankyloglossia, assuming it to be less than 15%, with a 95% confidence interval (CI) and a precision of 1.5% and no loss to follow-up, a necessary sample of at least 2177 individuals was determined.

All patients who met the following inclusion criteria were selected: patients born at the "Hospital Nostra Senyora de Meritxell" during the established period and of both genders. We excluded all those newborns who were transferred for complex neonatal pathology to reference hospitals in neighboring countries and those with missing medical records.

Procedure

The sociodemographic data were collected through the review of the computerized clinical histories of the patients, confidentially and anonymously, being recorded in a database to finally correlate them and check the existence or not, of any association with the studied parameters. All the parents or guardians of the newborns signed an informed consent form at the time of birth, authorizing that the clinical-health information included in the clinical history and in their clinical course could be used later in the health, teaching and/or research fields.

The variables collected for the study were the following: presence or absence of ankyloglossia and type of frenulum according to the Coryllos classification[12] (Fig. 1), surgical intervention or not, first degree hereditary component, gender, Rh and blood group, type of breastfeeding as defined by the World Health Organization[1], causes of cessation and duration of breastfeeding.

All patients followed the same puerperium control protocol from delivery until 6 months of age. The Nursing Service recorded the type of initial breastfeeding and the gender of the newborn in the clinical history. The Pediatrics Service, composed of 5 calibrated pediatricians, performed an assessment at the time of birth by means of a complete physical examination of the newborn, including examination of the oral cavity and maxillofacial region, diagnosing ankyloglossia according to the Coryllos classification[12] and recording the existence of a family history of ankyloglossia. Furthermore, during the first 72 hours after birth, the Maternal and Infant Unit, made up of pediatricians and nurses, was responsible for reevaluating the morphological and functional aspect of the lingual frenulum, offering support measures and reinforcement of the BF technique (the existence or not of difficulties with BF was evaluated). In cases in which these support measures failed and difficulties with BF persisted, the pediatrician referred the infant to the Pediatric Surgery Unit to evaluate the performance of lingual frenotomy within a maximum of 7 days, after detailed information and informed consent by the parents. The main reason for performing the surgical intervention of frenotomy was the combination of a diagnosis of ankyloglossia according to the Coryllos classification and ineffective sucking causing difficulties with BF.

Surgical intervention was performed using 3 drops of a gluco-saline solution (0.9 mL sodium chloride and 5% glucose) applied topically in the oral cavity by a disposable 2 mL syringe, prior to placing a grooved catheter and 12 cm Metzenbaum dissecting scissors. A horizontal incision and subsequent section of the mandibular insertion fibers was performed for surgical release of the lingual frenulum. After the surgical procedure, ambulatory controls were carried out at home by the midwives of the nursing service to evaluate the evolu-

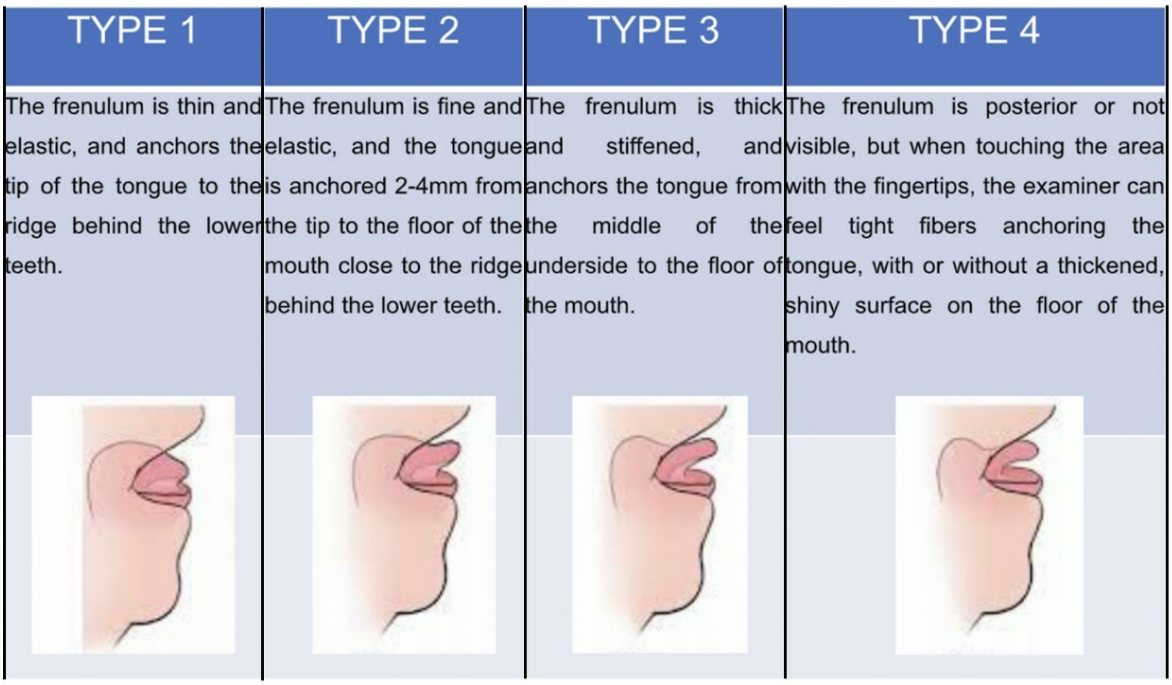

Figure 1: Anatomical classification of the lingual frenulum used in the present research study.

tion of BF and to record the maintenance of BF after surgical treatment at 30 days, 3 months and 6 months. The reasons for cessation BF were also recorded: health problems of the child or the mother, breast pain, mastitis, maternal decision and/or Coronavirus disease 2019 (covid-19).

To minimize inter-examiner variability, an inter-examiner calibration of the 5 pediatricians was performed prior to the start of data collection for the diagnostic of presence or absence of ankyloglossia and type of frenulum. A randomly selected group of 20 neonates, not belonging to the sample, was examined. Each pediatrician examined these patients and recorded them on a card. Subsequently, the concordance between examiners was checked with the percentage of concordance and the Kappa test (0.9). A concordance percentage of 90% was obtained, being an almost complete concordance. Thus, the diagnostic criteria used throughout the sample collection period were considered valid.

Statistical analysis

Statistical analysis was performed with the R Version 4.1.1 (R Foundation for Statistical Computing, Core Team, Vienna, Austria). Means and standard deviations, together with percentages, were used to describe each of the variables. To compare prevalences the ratio was calculated with a confidence interval and a chi-square test. The confidence level was 95% and the *p*-values under 0.05 were statistically significant.

RESULTS

A total of 2333 newborns were included, of whom 183 met diagnostic criteria for ankyloglossia. The overall prevalence of ankyloglossia was 7.84%. Of the 183 patients with short lingual frenulum, 95 were male (51.95%) and 88 were female (48.05%) with a 2:1.8 ratio. In relation to the characterization of the sample and the demographic characteristics according to sex, the presence or absence of ankyloglossia, hereditary component, ABO blood group and RH, are described in Table 1.

Regarding the ABO blood group variable of 2333 newborns, the larger group was A with 1233 newborns (52.85%), of which 7.5% (n = 92) presented ankyloglossia. No significant differences (p-value $\geq$ 0.05) were found between the different ABO blood groups and the fact of presenting or not short lingual frenulum. Regarding Rh, the most frequent was Rh positive (86.71%; 2023/2333); of which, 7.5% (n = 151) presented ankyloglossia. The remainder of the present study sample (n = 310 infants) were Rh negative (13.9%); of which, 10.3% (n = 32) had ankyloglossia. No statistically significant differences were found, but there was some tendency for being Rh negative and presenting short lingual frenulum to be significant (p = 0.081).

Of the newborns with ankyloglossia, 22.4% (n = 41) had a family history, most of them being first-degree relatives: parents or siblings (Table 1).

Of the 183 newborns with ankyloglossia, lingual frenotomy was performed in 136 subjects with ankyloglossia associated with BF difficulties; of which, according to the Coryllos anatomical classification [12], 13 patients (9.5%) had a type I frenulum, 114 patients (83.9%) had a type II frenulum, 8 patients (5.9%) had a type III frenulum and, finally, 1 patient (0.7%) had a type IV frenulum. In 47 patients (25.68%), of the 183 infants with ankyloglossia, lingual frenotomy was not necessary because they followed the measures of support and reinforcement of the BF technique established by the Maternal

Table 1: Demographic profile and prevalence of ankyloglossia in the study population.

		N	Global % (95% CI)	N	Ankyloglossia % (95% CI)	*p*-Value
Global		2333		183	7.84 (6.80; 9.03)	
Gender						
	Women	1166	49.98	88	7.5 (6.13; 9.25)	
	Man	1167	50.02	95	8.1 (6.67; 9.90)	0.594 NS
Rh						
	Positive	2023	86.71	151	7.5 (6.37; 8.72)	
	Negative	310	13.29	32	10.3 (7.27; 14.39)	0.081 NS
Blood Type						
	A	1233	52.85	92	7.5 (6.09; 9.11)	
	B	200	8.57	17	8.5 (5.18; 13.48)	
	AB	99	4.24	11	11.1 (5.95; 19.41)	
	O	801	34.33	63	7.9 (6.14; 10.01)	0.608 NS
Type of frenulum (Ankyloglossia Newborns)						
	Anterior	Type I		17	9.3	
	(n = 170)	Type II		153	83.6	
	Posterior	Type III		11	6	
	(n = 13)	Type IV		2	1.1	
Hereditary component (Ankyloglossia Newborns)				41	22.4	

NS = Nonsignificant value ($p > 0.05$); CI = confidence interval.

and Infant Unit of the Hospital, overcoming the difficulties in breastfeeding.

Of the 2150 infants who were not diagnosed with ankyloglossia, 1611 (74.93%) received exclusive BF at the beginning, 397 (18.47%) used exclusive bottle-feeding and 142 (6.6%) mixed feeding (breastfeeding and bottle-feeding). Table 2 provides the results regarding the evolution of the 1611 infants who received exclusive BF and who were not diagnosed with ankyloglossia, at 1 month, 3 months and 6 months of life, showing a 21.5% decrease in exclusive BF at 1 month of life.

Table 2: Persistence of exclusive breastfeeding at 1, 3 and 6 months of life in the group of newborns without ankyloglossia.

	Method of Feeding	N (%)
1 month		
	By exclusive bottle-feeding	119 (7.39%)
	By mixed feeding	227 (14.09%)
	By exclusive breastfeeding	1265 (78.52%)
3 months		
	By exclusive bottle-feeding	137 (8.5%)
	By mixed feeding	259 (16.08%)
	By exclusive breastfeeding	1215 (75.42%)
6 months		
	By exclusive bottle-feeding	144 (8.94%)
	By Bottle and Breast	262 (16.26%)
	By exclusive breastfeeding	1205 (74.80%)

Of the 136 infants who underwent frenotomy, 120 (88.24%) received exclusive BF at baseline, 14 (10.29%) were exclusively bottle-feeding and 2 (1.47%) were mixed feeding. On the other hand, of the 47 patients who were diagnosed with ankyloglossia but did not undergo surgical intervention, 39 (82.97%) received exclusive BF at baseline, 7 (14.89%) used exclusive bottle-feeding and 1 (2.13%) mixed feeding. Despite the surgical intervention, there was a 26.7% decrease in exclusive BF at 1 month of life among the patients who underwent frenotomy. Table 3 provides data on the evolution of exclusive BF at 1 month, 3 months and 6 months of life in neonates with ankyloglossia, both in the surgical intervention group (frenotomy) and in the non-surgical intervention group. The persistence of exclusive BF in the frenotomy group was similar to that of the non-surgical group, both at 1 month (73.3% *vs.* 76.92%), 3 months (67.5% *vs.* 71.8%), and 6 months (65.84% *vs.* 71.8%). However, statistically significant differences were found with respect to mixed feeding. The percentage of mixed feeding increased in the surgical intervention group with respect to the non-intervention group at one month ($p = 0.05$), 3 months ($p = 0.016$) and 6 months of life ($p = 0.016$). Likewise, statistically significant differences were also found with regard to exclusive bottle-feeding. This type of feeding increased in percentage in the group that did not undergo surgery with respect to those who underwent surgery at one month ($p = 0.033$), 3 months ($p = 0.013$) and 6 months ($p = 0.037$).

Finally, Table 4 describes the main reasons for exclusive BF cessation before 6 months of life in the 458 newborns (19.63% of the total sample) in whom this type of feeding was initially recorded, regardless of whether or not they had been diagnosed with ankyloglossia.

Table 3: Persistence of exclusive breastfeeding at 1 month of life, 3 months and 6 months in the intervention and non-surgical intervention groups of patients diagnosed with ankyloglossia.

		Method of Feeding		X2 test		
		Intervention N (%)	Non Intervention N (%)	PR	(95% CI)	*p* Value
1 month						
	By exclusive bottle-feeding	6 (5.0%)	6 (15.39%)	0.32	(0.11 to 0.95)	0.033*
	By mixed feeding	26 (21.7%)	3 (7.69%)	2.82	(0.90 to 8.80)	0.05*
	By exclusive breastfeeding	88 (73.3%)	30 (76.92%)	0.95	(0.78 to 1.17)	0.656 NS
3 months						
	By exclusive bottle-feeding	8 (6.67%)	8 (20.51%)	0.32	(0.13 to 0.81)	0.013*
	By mixed feeding	31 (25.83%)	3 (7.69%)	3.36	(1.09 to 10.38)	0.016*
	By exclusive breastfeeding	81 (67.5%)	28 (71.8%)	0.94	(0.75 to 1.19)	0.616 NS
6 months						
	By exclusive bottle-feeding	10 (8.33%)	8 (20.51%)	0.41	(0.17 to 0.96)	0.037*
	By mixed feeding	31 (25.83%)	3 (7.69%)	3.36	(1.09 to 10.38)	0.016*
	By exclusive breastfeeding	79 (65.84%)	28 (71.8%)	0.92	(0.72 to 1.16)	0.491NS

**Statistically significant ($p < 0.05$). NS = Nonsignificant value ($p > 0.05$); CI = confidence interval; PR = prevalence ratio.*

Table 4: Main reasons for cessation of exclusive breastfeeding in the study sample.

Reasons for early weaning	N	%
Mother's decision	207	45.20%
Child health problems (low weight gain)	122	26.64%
Breast pain	43	9.39%
Still hungry after feedings	42	9.17%
Mastitis	29	6.33%
Mother health problems	12	2.62%
By Covid-19	2	0.44%
Others	1	0.21%
Total	458	

DISCUSSION

The prevalence of ankyloglossia in infants is variable in the literature[5,7,11], probably due, among other causes, to a lack of consensus regarding its definition. Prevalence varies from 4.2 to 10.7% in newborns. The prevalence of ankyloglossia in our study group was 7.84%, a value similar to that obtained by others authors[3–5]. However, Ferrés-Amat *et al.*[13] obtained a prevalence of 15% of ankyloglossia in the population of Barcelona, Spain, who attended a pediatric hospital, a value that is 2 to 3 times higher than that obtained by Messner *et al.*[3], who found a prevalence of 4.8%. This high value in the Barcelona study, compared to that published in other studies[3–5], is most likely due to the fact that these were newborns whose families attended a specialized BF service that dealt with problems derived from BF.

As reported in the literature, males seem to be more affected than females[3,5,16,17], although Walsh and Tunkel[18] observed an inverse relationship. In our study we also found a slight difference (not statistically significant) between males and females in children with ankyloglossia, with a ratio of 2:1.8.

Furthermore, although most cases of ankyloglossia are sporadic, a genetic etiology with a greater male predilection is suspected because it is linked to an X-linked transmission pattern[3,19]. In multiple studies[20–22] positive family history was observed in 10 to 53% of the subjects. In our work group, 22.4% of the newborns with short lingual frenulum had a family history, most of them being first-degree relatives (parents or siblings). Therefore, there is some evidence that genetic components could be involved in the transmission and manifestation of ankyloglossia. However, it is difficult to define the inheritance of a condition being its diagnosis merely retrospective and based exclusively on parental reports.

One of the main unresolved problems of ankyloglossia is its classification and diagnosis. There is great controversy due to the lack of a universally accepted definition and diagnosis and the lack of consensus on the relationship between the symptomatology presented by nursing mothers and the degree of ankyloglossia in infants. In our study, we used the Coryllos classification[12] because allows identifying by palpation type III and IV short frenulum that can go unnoticed to the visualization[13] and is also easy to apply. It focuses on the type of frenulum, but does not address functionality or criteria for ankyloglossia. The diagnosis of ankyloglossia in newborns requires a combination of functional and anatomical criteria[20]. Haham *et al.*[23] found no statistical correlation between the type of Coryllos lingual frenulum and the presence or absence of problems with BF. In the present study, the pediatricians of the Hospital of Andorra used the Coryllos classification[12] as a diagnostic tool and criterion, associating the limitation of lingual mobility with difficulties in sucking and ability to maintain latching to the breast to decide whether surgery should be performed.

Regarding the study of the AB0 blood group variable, no association was found with the presence or absence of ankyloglossia in neonates. However, a certain tendency has been observed between being Rh negative and presenting ankyloglossia, being a relevant but not conclusive result. It should not be forgotten that the expression of these group antigens is genetically determined[24].

It is common for health care professionals to advise treating problems resulting from ankyloglossia and BF with the sur-

gical technique of frenotomy[25]. Clearly defining the indications and diagnostic criteria is essential to limit overtreatment and, at the same time, to ensure that infants who would benefit from surgical intervention are treated in a timely manner. Emond *et al.*[26] concluded that when a diagnosis of short lingual frenulum associated with breastfeeding problems (pain, lack of latch-on) does not improve with conservative treatment (positioning techniques, nipple shields, myofunctional speech therapy), lingual frenotomy should be performed as soon as possible to reduce the likelihood of the mother discontinuing breastfeeding.

The studies by Emond *et al.*[26] and Dollberg *et al.*[27] have not answered the clinically relevant question of whether frenotomy, in infants presenting ankyloglossia with feeding difficulty, results in long-term BF success and resolution of maternal pain. In our study, lingual frenotomy did not result in statistically significant differences in the maintenance of exclusive BF compared to untreated infants. Statistically significant differences were obtained in the maintenance of mixed feeding at 1, 3 and 6 months of life, in favor of those treated surgically with respect to those not treated. Statistically significant differences were also found in relation to exclusive bottle-feeding, which increased at 1, 3 and 6 months, in favor of those treated surgically over those not treated. These results indicate that lingual frenotomy can provide improvements in BF for the baby and mother, achieving effective sucking and reducing maternal pain, but it cannot be affirmed that it is the only treatment. Surgical treatment of the lingual frenulum can improve the tongue movements necessary for effective sucking, but the help of different specialists is also advised to stimulate sucking as a complement to the surgical intervention. Mother's motivation to breastfeed and the support of a specialized team with which she has access play a decisive role in resolving the difficulties and maintaining exclusive breastfeeding or mixed feeding.

Ankyloglossia alone is not an indication for surgical intervention; other functional factors should be assessed to determine whether a short lingual frenulum is or will be a cause of breastfeeding failure. Emond *et al.*[26] and Dollberg *et al.*[27] found no objective changes after frenotomy, but showed a steady reduction in maternal nipple pain. However, one clinical trial reported that lingual frenotomy objectively improved BF[26]. Because of the benefits of BF, the World Health Organization (WHO) in association with the United Nations Foundation (UNICEF) advises exclusive BF until 6 months of age. After this period, children should continue BF associated with complementary feeding until the age of two years or more[28]. The literature reports that there is a growing tendency to cease exclusive BF before 4 months of age, despite the fact that most infants leave maternity wards being breastfed[26,27,29]; data that agree with those of the present study in which a 21.5% decrease in exclusive BF at 1 month of life was observed in infants not diagnosed with ankyloglossia and 26.7% at 1 month of life among patients who underwent lingual frenotomy. Previous studies[26,27,29] have indicated that early weaning is a common practice in most nursing mothers. The causes of early weaning are diverse. Among the causes reported by mothers for discontinuing BF, the most common are: mastitis, milk production deficit, failure to gain weight, and return to work[30]. In our study, the most frequent cause of cessation was maternal decision not due to any objective factor, followed by low weight gain and breast pain. Vega *et al.*[30] reported that of the interventions that promote BF, previous educational programs were the most effective, hence the importance of improving BF promotion and education, especially in the prenatal stage.

Undoubtedly, it is essential to unify and objectify the diagnostic criteria for ankyloglossia, as well as its typification, based on morphological and functional criteria, in order to finally establish multidisciplinary circuits and protocols to define the correct and effective treatment of short lingual frenulum and, also, to maintain and ensure BF.

CONCLUSIONS

• Lingual frenotomy was only performed on patients with ankyloglossia associated with ineffective suction that causes BF difficulties.

• Whether or not lingual frenotomy was performed was not statistically significant for the maintenance of exclusive BF at 1 month, 3 months and 6 months in patients diagnosed with ankyloglossia. On the other hand, it was statistically significant in the prolongation of mixed feeding.

• Finally, to highlight the importance of working with a multidisciplinary team and the need for a correct and early diagnosis of ankyloglossia to solve the problems of breastfeeding.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

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