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ORIGINAL ARTICLE

Prevalence of neonatal ankyloglossia in a tertiary care hospital in Spain: a transversal cross-sectional study

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Abstract

Ankyloglossia, or tongue-tie, is a congenital anomaly in which a short lingual frenulum or a highly attached genioglossus muscle restricts tongue movement. The reported prevalence of neonatal ankyloglossia varies between less than 1 and 12.1% depending upon the study population and criteria used to define and grade ankyloglossia. Our hypothesis was that ankyloglossia had a higher prevalence among our newborn population than previously reported. We conducted an observational, transversal cross-sectional study which included all neonates born in our center between January 1 and December 31, 2018, and actively assessed for tongue-tie. We considered "clinically significant" or "symptomatic" ankyloglossia using the Hazelbaker tool for appearance and function when the mother experienced nipple pain or bruises, or when the neonate had difficulty latching onto the breast. A total of 1392 neonates were born at our center in 2018. Tongue-tie was identified in 645 infants (46.3%), of which 453 were symptomatic (70.2%). Thus, clinically significant ankyloglossia was present in 32.5% of the neonates born in 2018. Their distribution according to Coryllos's types were as follows: 45 type 1 (7.0%), 230 type 2 (35.6%), 321 type 3 (49.8%), and 42 type 4 (6.5%).

Conclusion: The prevalence of symptomatic ankyloglossia in our population is higher (32.5%) than studies have reported to date. Actively assessing for tongue-tie increases its diagnosis.

What is Known:
- *There are four types of tongue-tie according to Coryllos (1, 2, 3, and 4), of which the two anterior types (1 and 2) are the most apparent and easy to diagnose.*
- *The reported prevalence of ankyloglossia generally varies from < 1 to 12.1%, although some recent studies report a higher prevalence.*

What is New:
- *We found a prevalence of neonatal ankyloglossia of 46.3%, of which 70.2% was symptomatic (clinically significant ankyloglossia was present in 32.5% of the neonates born in 2018 at our hospital).*
- *Actively assessing for ankyloglossia and posterior tongue-ties, which are likely more often undiagnosed, increases its diagnosis.*

Keywords Ankyloglossia · Tongue-tie · Neonate · Prevalence · Breastfeeding problems

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Abbreviations

IBCLC International Board of Lactation Consultant Examiners

Introduction

The lingual frenulum is a submucosal band of connective tissue or a membrane that inserts on the tip of the tongue or along its undersurface [1–3]. Ankyloglossia, or tongue-tie, is a congenital anomaly in which a short lingual frenulum or a highly attached genioglossus muscle restricts tongue movement [4, 5]. It may present as an abnormally short frenulum that inserts at or near the tip of the tongue, as difficulty lifting the tongue to the upper dental alveolus, as an inability to protrude the tongue past the lower central incisors, as an impaired side-to-side lingual movement, or as a notched or heart-shaped tongue when it is protruded [1, 3, 6, 7]. There are four types of tongue-tie defined by Coryllos depending on the site of insertion (Fig. 1) [8]. Posterior ankyloglossia refers to when a short, thickened, or even submucosal frenulum is attached at the middle or the posterior part of the undersurface of the tongue and can restrict tongue mobility [9].

The reported prevalence of neonatal ankyloglossia varies between less than 1 and 12.1% depending upon the study population and criteria used to define and grade ankyloglossia [1, 3, 6, 10–14]. Our study hypothesis was that ankyloglossia had a higher prevalence among our newborns than previously reported. Our objectives were to characterize the population of neonates born at our hospital within the year 2018 in relation to the presence of ankyloglossia, determine the prevalence of ankyloglossia, characterize the most frequent types of tongue-tie, and determine whether they were clinically significant or not.

Patients and methods

We conducted an observational, transversal cross-sectional study of the neonates born at a tertiary care hospital in Barcelona, Spain, which experiences approximately 1400 births per year and services an area of population of approximately 400,000 people. We included all the neonates born in our center between January 1 and December 31, 2018, to make this a population-based study. At least one of three staff neonatologists evaluated the presence of tongue-tie in the neonates and graded it based on Coryllos's criteria (see Fig. 1) as part of the routine neonatal evaluation, both during the first neonatal exam and before discharge. Apart from the prominence of the frenulum, we also assessed its impact on tongue movement and on breastfeeding following the Hazelbaker tool [15] (see Fig. 2). We considered tongue-tie to be symptomatic if it scored 8 points or less in appearance and/or 11 points or less in function according to Hazelbaker, or if the mother experienced nipple pain or bruises, or the neonate had problems latching onto the breast after an International Board Certified Lactation Consultant (IBCLC) examiner assessed for and corrected other reasons for maternal pain such as retrognathia, micrognathia, incorrect positioning, insufficient mouth opening, and latching onto the nipple only. In our neonatal unit, we offer intensive lactation support to all the mothers and have two IBCLC nurses on our staff. Our Ethics Committee approved the study.

Study variables were gender, gestational age, birth weight, weight at discharge, birth mode, prematurity, presence of ankyloglossia, type of tongue-tie according to Coryllos, clinically significant ankyloglossia (nipple pain or problems latching onto the breast, Hazelbaker score), and feeding choice at birth (breastfeeding alone, bottle-feeding, or mixed). We included the Hazelbaker tool (appearance and function) of a representative sample of 125 of the included newborns who underwent a frenotomy (we started registering the scores obtained using the Hazelbaker tool in our database in September 2018).

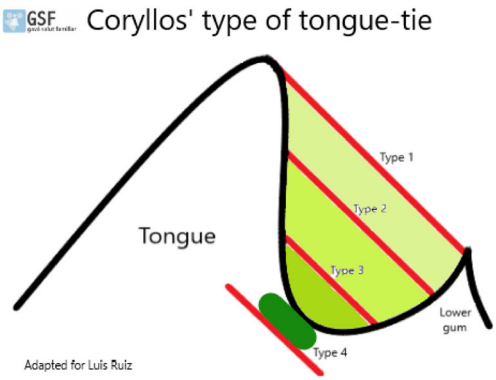
Statistical analyses Quantitative variables (gestational age, birth weight, weight at discharge), which were normally distributed, were described using the mean and standard deviation. Categorical variables (gender, birth mode, prematurity, presence of ankyloglossia, clinically significant ankyloglossia, feeding choice at birth) were expressed as frequencies and percentages. We compared the population of tongue-tied neonates with non-tongue-tied neonates (controls) to verify that the two samples were homogeneous in terms of gestational age, birth weight, and sex. Qualitative variables were compared with Pearson's chi-square or Fisher's exact test as appropriate and quantitative variables with Student's *t* test when two groups were compared and with ANOVA when four groups were compared. Significance was set at the $p < 0.05$ level. To perform statistical analyses, we used STATA version 15.1 (StataCorp, College Station, TX, USA).

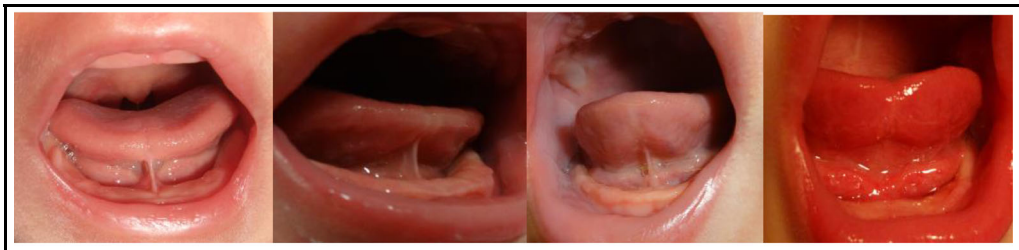
Results

A total of 1392 neonates were born at our center in 2018. We had 950 normal vaginal births (68.2%), 131 instrument-assisted births (9.4%), 311 cesareans (22.3%), and 133 preterm neonates (9.5%). The mean ± standard deviation (SD) birth weight was 3195 ± 535 g with a mean ± SD gestational age of 39 $^{1/7}$ ± 2 $^{0/7}$ weeks. Weight at discharge was 3032 ± 487 g. Weight loss was globally between 4.67 and 5.43%. Excluding neonates with birth weight < 2000 g, mean weight loss was 5.28%, and excluding preterm babies, mean weight loss was 5.21%. The mean ± SD maternal age was 31.1 ± 6.2

Types of ankyloglossia according to Coryllos [8].

- Type 1: Insertion of the frenulum to the tip of the tongue.
- Type 2: Insertion of the frenulum slightly (two to four mm) behind the tip of the tongue.
- Type 3: Thickened frenulum attached to the mid-tongue and the middle of the floor of the mouth, usually tighter and less elastic.
- Type 4: Thick, shiny and very inelastic submucosal frenulum that restricts movement at the base of the tongue.





Type 1 Type 2 Type 3 Type 4

GSF

Types 1 and 2, considered "classical" tongue-tie, are the most common and obvious tongue-ties. Types 3 and 4 are less common, and since they are more difficult to visualize, they are the most likely to go untreated. Photographs taken by Dr. Luis Ruiz-Guzmán after obtaining written permission from the infants' parents.

Fig. 1 Types of ankyloglossia according to Coryllos [8]. Type 1: insertion of the frenulum to the tip of the tongue. Type 2: insertion of the frenulum slightly (2 to 4 mm) behind the tip of the tongue. Type 3: thickened frenulum attached to the mid-tongue and the middle of the floor of the mouth, usually tighter and less elastic. Type 4: thick, shiny, and very inelastic submucosal frenulum that restricts movement at the base of the tongue. Types 1 and 2, considered "classical" tongue-tie, are the most common and obvious tongue-ties. Types 3 and 4 are less common, and since they are more difficult to visualize, they are the most likely to go untreated. Photographs taken by Dr. Luis Ruiz-Guzmán after obtaining written permission from the infants' parents

years. Tongue-tie was found in 645 infants (46.3%). Of them, 453 were symptomatic (70.2%) (Fig. 3). Thus, clinically significant ankyloglossia was present in 32.5% of all neonates born in 2018 at our hospital. Their distributions according to Coryllos's types were as follows: 45 type 1 (7.0%), 230 type 2 (35.6%), 321 type 3 (49.8%), and 42 type 4 (6.5%). The mean Hazelbaker score from the representative sample of our tongue-tied infants (n = 125) was 5.8 points (SD 1.7, range 1–9) for appearance and 7.8 points (SD 1.8, range 1–13) for function. The male-to-female ratio was 1.4:1. There were no statistical differences between tongue-tied and non-tongue-tied infants in terms of birth weight and weight at discharge, birth mode, and prematurity rate. However, as Table 1 shows, there were significant differences in gender, maternal age, gestational age, and feeding choice at birth. More mothers of tongue-tied infants decided to breastfeed at birth (89.9 vs

Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLLF)			
Appearance items	Score	Function items	Score
Appearance of tongue when lifted		Lateralization	
Round or square	2	Complete	2
Slight cleft in tip apparent	1	Body of tongue but not tongue tip	1
Heart-shaped	0	None	0
Elasticity of frenulum		Lift of tongue	
Very elastic (excellent)	2	Tip to mid-mouth	2
Moderately elastic	1	Only edges to mid-mouth	1
Little or no elasticity	0	Tip stays at alveolar ridge or rises to mid-mouth only with jaw closure	0
Length of lingual frenulum when tongue lifted		Extension of tongue	
More than 1 cm or embedded in tongue	2	Tip over lower lip	2
1 cm	1	Tip over lower gum only	1
Less than 1 cm	0	Neither of above, or anterior or midtongue humps	0
Attachment of lingual frenulum to tongue		Spread of anterior tongue	
Posterior to tip		Complete	2
At tip		Moderate or partial	1
Notched tip		Little or none	0
Attachment of lingual frenulum to inferior alveolar ridge		Cupping	
Attached to floor of mouth or well below ridge	2	Entire edge, firm cup	2
Attached just below ridge	1	Side edges only, moderate cup	1
Attached at ridge	0	Poor or no cup	0
Total appearance score		Peristalsis	
Function items score ▪ 14: perfect score (regardless of *Appearance item* score) ▪ 11: acceptable, if *Appearance item* score is 10 ▪ <11: function impaired Frenotomy should be considered if management fails. Frenotomy necessary if *Appearance item* score is <8.		Complete, anterior to posterior (originates at the tip)	2
		Partial: originating posterior to tip	1
		None or reverse	0
		Snapback	
		None	2
		Periodic	1
		Frequent or with each suck	0
		Total function score	

Fig. 2 Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLLF) [15]

84.2%, $p = 0.002$; Table 1), whereas more mothers of non-tongue-tied infants chose formula (11.6 vs 6.2%).

Table 2 presents the demographic characteristics of the ankyloglossia group depending on the type of tongue-tie. There were no differences in terms of maternal age, gestational age, birth mode, or prematurity rate between the four groups. We found differences among the male-to-female ratio in different Coryllos's types: higher in types 1 and 2 (2:1 in type 1 and 1.7:1 in type 2, $p = 0.000$). There were differences in weight loss at discharge depending on the type of tongue-tie. Type 4 tongue-tied patients presented the lowest weight loss ($p = 0.026$). The results presented in Table 2 are global; therefore, they include preterm babies who may have been discharged later than 72 h post birth. We performed a subanalysis excluding neonates born prior to 35 weeks of gestation and those with a birth weight < 2000 g, because in those cases, hospital stay was longer and those patients were discharged when already gaining weight. Excluding neonates with birth weight < 2000 g, weight loss was 5.28%, and excluding preterm babies, weight loss was 5.21%, which is similar to the global results. Clinically significant ankyloglossia was more frequent in types 1 and 2 (93.2 and 83.4%, respectively) compared with posterior tongue-ties ($p = 0.000$).

Discussion

The prevalence of ankyloglossia is difficult to estimate because a unique definition with an objective grading system does not exist [1, 3, 6, 10–14, 16, 17]. Some authors have

Table 1 Demographic characteristics of the ankyloglossia group and non-tongue-tied neonates

	Tongue-tied infants $n = 645$ (%)	Non-tongue-tied infants $n = 747$ (%)	p value
Male newborn	377 (58.4)	350 (46.8)	0.000[a]
Ratio ♂:♀	1.4:1	1.1:1	
Maternal age (mean, SD) (years)	31.4 (6.1)	31.1 (6.2)	0.047[b]
Gestational age (mean, SD) (range) (weeks)	$39^{0/7}$ ($2^{2/7}$) $31^{3/7}$–$42^{1/7}$	$39^{1/7}$ ($2^{0/7}$) $31^{3/7}$–$42^{1/7}$	0.036[b]
Prematurity	65 (10.1)	68 (9.1)	0.827[a]
Feeding choice at birth			0.002[a]
Breastfed	580 (89.9)	629 (84.2)	
Breastfed and bottle-fed	25 (3.9)	31 (4.1)	
Bottle-fed	40 (6.2)	87 (11.6)	
Clinically significant ankyloglossia	453 (70.2 of the total, 74.9% of breastfed infants)	Not assessable	-

[a] Pearson's chi-square

[b] Student's t test

published opinions without previous field research. Even though most authors report a prevalence from less than 1 to 12.1%, some report a much higher prevalence: 14.9% according to Schlatter and 22.5% according to Martinelli. Haham found that 99.5% of the newborns had a visible sublingual frenulum, although only 3.5% of them had a symptomatic tongue-tie and required a frenotomy [18–20]. González-Jiménez, a Spanish author, recently reported a prevalence of ankyloglossia of 12.1%, even though in a subarea of his study, the prevalence reached 50% [14]. One of our co-authors, whose population is very similar to ours, found a prevalence of ankyloglossia in children from 0 to 14 years of 31% and stated that this condition was often undiagnosed [21, 22]. Still, we found a higher prevalence among our study population (46.3%). Since the diagnosis of symptomatic ankyloglossia in our study required that the neonate be breastfed, 74.9% (453 of 605) of the neonates who were exclusively or mixed breastfed exhibited symptomatic ankyloglossia. Despite not being specified in the literature, we believe that studies often only refer to Coryllos's types 1 and 2 ankyloglossia (easily visualized at simple inspection) and do not consider or take into account types 3 and 4, which are less evident during simple evaluation and may require palpation of the undersurface of the tongue. If only types 1 and 2 were considered, we would have obtained a prevalence of 3.2% (type 1) or 19.7% (types 1 and 2 combined), which is more consistent with the literature. The most frequent tongue-tie in our population was type 3 (49.8%), which is consistent with the findings of another author from our area, Pastor-Vera, who found that type 3 accounted for 57.4% of ankyloglossia [16]. González Jiménez hypothesized that this was the reason for the generally lower prevalence reported as well as regional differences found within his own study [14]. Ghaheri also found a higher prevalence of posterior than anterior tongue-ties (78 vs 21%) which was in line with our results [23].

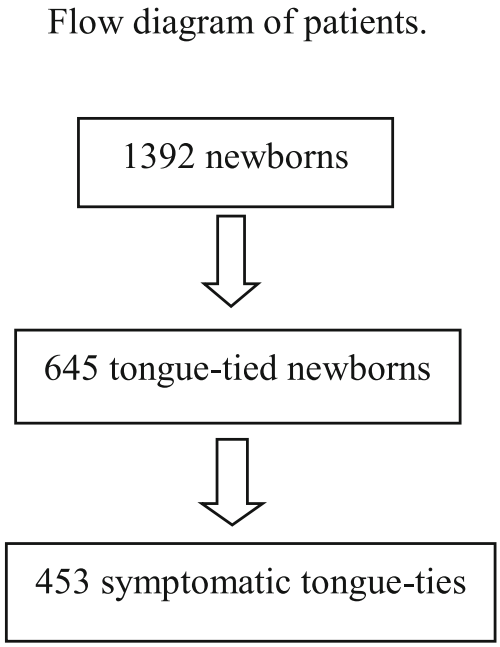


Fig. 3 Flow diagram of patients

We found that symptomatic ankyloglossia was significantly more frequent in anterior (types 1 and 2) than in posterior (types 3 and 4) tongue-ties. Among anterior tongue-ties, it was more frequent in type 1 than in 2, and among posterior tongue-ties, in type 3 than in 4. We hypothesize that because the lingual frenulum inserts on the tip of the tongue or close to it in types 1 and 2, this may restrict tongue movements more than posterior tongue-ties, thus making them more symptomatic.

Table 2 Demographic characteristics of the ankyloglossia group depending on the type of tongue-tie

	Type 1 (n = 45) (7.0%)	Type 2 (n = 230) (35.6%)	Type 3 (n = 321) (49.8%)	Type 4 (n = 42) (6.5%)	p value
Male newborn	30 (66.7)	145 (63.0)	178 (55.5)	24 (57.1)	0.000[a]
Ratio ♂:♀	2:1	1.7:1	1.2:1	1.3:1	
Birth weight (g) (mean, SD)	3165 (523)	3132 (537)	3192 (543)	3342 (560)	0.117[b]
Weight at discharge (mean, SD) (% of weight loss)*	2999 (483) (5.24)	2962 (483) (5.43)	3020 (493) (5.39)	3186 (468) (4.67)	0.026[b]
Prematurity	3 (6.7)	28 (12.2)	32 (9.9)	2 (4.8)	0.396[a]
Feeding choice at birth					0.019[a]
Breastfed	39 (86.7)	205 (89.1)	292 (91.0)	39 (92.9)	
Breastfed and bottle-fed	1 (2.2)	11 (4.8)	13 (4.0)	0 (0)	
Bottle-fed	5 (11.1)	14 (6.1)	16 (5.0)	3 (7.1)	
Clinically significant ankyloglossia	41 (93.2)	191 (83.4)	196 (61.4)	17 (40.5)	0.000[a]

Type of tongue-tie: data missing from 7 patients (1.4%)

*Global results (see text for details)

[a] Pearson's chi-square

[b] ANOVA

In most series, the frequency of tongue-tie is higher among boys, probably due to genetic factors, with a male-to-female ratio of 1.5:1 to 2.6:1 [1, 3, 11–14, 16, 22, 23]. We found a male-to-female ratio of 1.4:1 (375 out of 645, 58%), which is slightly lower than reported but as frequent as reported by other authors if we only considered anterior ankyloglossia (types 1 and 2). There were differences between tongue-tied infants and controls in terms of maternal age (31.4 ± 6.1 vs 31.1 ± 6.2 years) and gestational age ($39^{0/7} \pm 2^{2/7}$ vs $39^{1/7} \pm 2^{0/7}$), although these differences are not clinically relevant. In our study, ankyloglossia was not more common among preterm than term neonates. We observed a global low weight loss among our newborns, and only 39 neonates (2.8%) lost 10% of birth weight or more. In our center, newborns receive hospital discharge at 48 h after birth if born vaginally and at 72 h after birth if born via cesarean. There were no differences in weight loss between tongue-tied infants and controls. We believe that ankyloglossia does not affect weight loss at discharge because discharge is relatively early in the neonate's life. Since ankyloglossia is diagnosed through evaluation and report of maternal discomfort, affected neonates do not have enough time to exhibit failure to thrive during their hospital stay. We found differences in the feeding choice at birth among tongue-tied vs non tongue-tied infants (p = 0.002): more mothers of tongue-tied neonates decided to breastfeed at birth compared with mothers of controls (89.9 vs 84.2%), and less mothers of tongue-tied infants chose formula compared with mothers of non-tongue-tied infants (6.2 vs 11.6%).

We acknowledge that our study has limitations. First, we focused our efforts into identifying symptomatic ankyloglossia in breastfed neonates and did not study the possible effects of ankyloglossia on bottle-fed infants. Even though most authors agree that tongue-tie does not interfere with bottle-feeding, some found that it may indeed affect the infant's ability to suck from a bottle as well [1, 3, 11–13, 17]. Second, we did not objectively evaluate latching onto the breast in all the study patients. Even though we based our assessment of the breastfeeding dyads on the Hazelbaker tool, we only recorded Hazelbaker scores for the last 125 neonates included. We have since adopted its universal use and currently record this information for all neonates diagnosed with a tongue-tie. Third, we considered symptomatic tongue-tie if the mother experienced nipple pain or the infant had problems latching onto the breast. In addition, we acknowledge that there are several other reasons for maternal nipple pain apart from ankyloglossia. Nevertheless, IBCLC nurses assessed all mother/infant dyads during breastfeeding and provided lactation support which helped solve issues such as incorrect positioning.

Conclusions

The prevalence of symptomatic ankyloglossia in our population is higher than studies have reported to date. Actively assessing for tongue-tie increases its diagnosis.

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Authors' Contribution Dr. SM and Dr. MP designed the study, collected data, analyzed the results, drafted the initial manuscript, and approved the final manuscript as submitted.

Dr. LR instructed Dr. SM in distinguishing the different types of tongue-tie, provided the photographs included in this manuscript and created the figure, helped analyze our results, helped draft the initial manuscript, and approved the final manuscript as submitted.

Dr. XD performed all the statistical analyses and helped the rest interpret them. He helped write the "Patients and methods" part and approved the final manuscript as submitted.

Dr. ML helped conceptualize and design the study, helped draft the initial manuscript, and approved the final manuscript as submitted.

Compliance with ethical statements

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval This article does not contain any studies with human participants or animals performed by any of the authors. Our hospital Ethics Committee approved this study.

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