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Diagnosis of ankyloglossia in newborns: is there any difference related to the screening method?

Diagnóstico de anquiloglossia em recém- nascidos: existe diferença em função do instrumento de avaliação?

ABSTRACT

Purpose: To diagnose ankyloglossia in newborns and compare two lingual frenulum assessment instruments. **Methods:** This cross-sectional study was carried out in Recife, Pernambuco, Brazil, in 2018, with 147 mothers/newborns aged up to 30 days. The Bristol Tongue Assessment Tool and the Lingual Frenulum Evaluation Protocol for Infants were the instruments used. Sociodemographic data were also recorded. The two ankyloglossia diagnostic methods were compared using the McNemar test, obtaining the kappa agreement value and the confidence interval. **Results:** Ankyloglossia was present in 4.8% when diagnosed with the Bristol Tongue Assessment Tool, and in 17.0% with the Tongue-Tie Test. Regarding sex, 53.1% of the newborns were males and 46.9% were females; however, there was no association between ankyloglossia and the newborn's sex in either of the assessment methods. **Conclusion:** The ankyloglossia diagnosis in newborns varied depending on the assessment instrument used.

RESUMO

Objetivo: Diagnosticar a anquiloglossia em recém-nascidos, comparando dois instrumentos de avaliação do frênulo lingual. **Método:** Tratou-se de um estudo transversal, realizado em Recife, PE, Brasil no ano de 2018, com 147 mães/recém-nascidos com idade de até 30 dias de vida. Foram utilizados o Instrumento *Bristol Tongue Assessment Tool* (BTAT) e o Protocolo de Avaliação do Frênulo da Língua para Bebês (“Teste da Linguinha”). Dados sociodemográficos também foram anotados. Para a comparação entre os dois métodos de diagnóstico da anquiloglossia, foi utilizado o teste de McNemar e foram obtidos o valor da concordância de Kappa e o respectivo intervalo de confiança. **Resultados:** A presença de anquiloglossia foi de 4,8%, quando diagnosticada por meio do BTAT, e de 17,0%, quando utilizado o “Teste da Linguinha”. Com relação ao sexo, 53,1% dos recém-nascidos eram do sexo masculino e 46,9% do sexo feminino; contudo, não houve associação entre a anquiloglossia e o sexo do recém-nascido nos dois métodos de avaliação. **Conclusão:** O diagnóstico da anquiloglossia em recém-nascidos variou em função do instrumento de avaliação utilizado.

Study conducted at Centro Integrado de Saúde Amaury de Medeiros – CISAM - Recife (PE), Brasil.

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INTRODUCTION

Ankyloglossia is a condition that may occur and be verified at birth. When present, it limits the normal tongue movement and may hinder breastfeeding. For years, such a change was not considered a barrier to begin and continue breastfeeding. Maybe for this very reason, no early diagnosis and treatment strategy was introduced into the maternity routine^(1,2).

However, an increasing search for help in the milk banks due to latch difficulties, nipple pain, the excessive fatigue when the newborn with ankyloglossia is sucking, and, consequently, the mother's low milk production led to an important questioning on the part of this study's researchers about what the impact of ankyloglossia on breastfeeding would be⁽¹⁻⁵⁾.

Changes in the lingual frenulum have been associated with difficulties to begin breastfeeding and with early weaning. However, the studies lack standardized instruments to diagnose ankyloglossia and have different definitions of ankyloglossia, which sets up a barrier to more trustworthy scientific evidence, leading to limited results⁽⁶⁻⁸⁾.

The ankyloglossia prevalence rates range from 0.52% to 21% – which may have been underestimated in some studies that did not use a standardized assessment instrument^(1,3,8-14). Thus, ankyloglossia with limited symptomatology was sometimes not diagnosed^(6,7). Having a standardized and unified protocol is seen as essential to the early diagnosis and treatment of this condition. Nevertheless, no consensus has yet been reached about a single protocol to be used in its diagnosis^(1,3,4,8-14).

Considering the importance of breastfeeding to the newborn's growth and development and the possibility of ankyloglossia interfering with the breastfeeding process, federal law no. 13.002 was passed in Brazil in June 2014. It makes it obligatory for all maternities in Brazil to conduct early ankyloglossia diagnosis. The Brazilian Ministry of Health suggests that the change be diagnosed with the Bristol Tongue Assessment Tool (BTAT)^(11,14). However, such a preference is not backed by scientific evidence, and few studies compare this instrument with other validated ones^(15,16).

Hence, this paper aimed to diagnose ankyloglossia in newborns, comparing two lingual frenulum assessment instruments.

METHOD

This study was approved by the Research Ethics Committee of the *Universidade de Pernambuco* (University of Pernambuco), Brazil, in compliance with resolution no. 466/12 of the National Health Council, under Certificate of Presentation for Ethical Consideration no. 73673117.6.0000.5207 and evaluation report no. 2.414.893; it also complied with the principles of the declaration of Helsinki. All the mothers signed the informed consent form (ICF).

This is an observational, cross-sectional study developed at a public state maternity hospital (Amaury de Medeiros Health Center), in Recife, Pernambuco, Brazil, in 2018. The sample size

was defined based on the estimated population of 1,110 newborns, considering the average births per month at the maternity, for 3 months; on the estimated prevalence of 8.8% (a percentage found in a pilot study, not yet published); 95% confidence interval; 5% maximum acceptable error, a predetermined effect of 1.2, considering a 20% loss. The minimum sample was set at 124 mother-baby dyads. The final sample (147 dyads) was larger than what had been calculated because some mothers asked to participate in the research and have their child's lingual frenulum assessed.

The data were collected by a calibrated researcher trained to administer the two protocols – she was named Assessor 1 (A1). The researcher's training consisted of theoretical-practical courses that qualified her to use the protocols. She was calibrated by the maternity's clinical speech-language-hearing therapist, considered the gold standard – she was named Assessor 2 (A2). The inter-examiner calibration compared the anatomic-functional assessments conducted by A1 and A2 in 25 newborns (test-retest) before this study. These were not considered for the final sample.

The inter-examiner agreement analysis compared the anatomic-functional assessments conducted by A1 and A2 in 30 of the 147 newborns, having compared the data obtained in the assessment of 20% of the sample (test-retest). Hence, one out of every five newborns were reassessed at different moments, ensuring the confidentiality of the result between the examiners. Thus, the inter-examiner degree of agreement was 0.99 for the BTAT and 0.90 for the Tongue-Tie Test.

The intra-examiner agreement analysis compared the data obtained in the anatomic-functional assessment and reassessment of 20% of the sample (test-retest) conducted by A2 in a randomly selected group of subjects. The data were reassessed 15 to 30 days after the first assessment to avoid the memory effect. The intra-examiner degree of agreement was 1.00 for the BTAT and 0.99 for the Tongue-Tie Test. Cohen's kappa coefficient was used, reaching intra- and inter-examiners kappa > 0.90 in both instruments used, thus ensuring reliability.

The eligibility criteria encompassed being full-term born children, aged 1 to 30 days, whose mothers had no post-childbirth complications, staying in the hospital ward, and without syndromes, craniofacial malformation, microcephaly, cleft lip, cleft palate, or both. Newborns that were not submitted to both lingual frenulum assessment protocols were excluded from the study.

Newborns up to 30 days old were included because ankyloglossia must be diagnosed early to avoid breastfeeding difficulties and the possibility of weight loss and weaning.

The instrument used in the research were the BTAT⁽¹⁷⁾ and the Lingual Frenulum Evaluation Protocol for Infants (Tongue-Tie Test)⁽¹⁸⁾. Sociodemographic data, such as the newborns' age in days at the examination, newborn's sex, mother's age, mother's schooling level, and monthly family income, were also collected.

The BTAT considers four aspects⁽¹⁷⁾: (A) tongue tip appearance – it can have the shape of a heart (score 0), have a slight cleft in the apex (score 1), or be rounded (score 2); (B) frenulum fixed to the lower alveolar ridge – it can be fixed to the upper part of the gum (score 0), the inner part of the gum (score 1), or in the middle of the floor of the mouth (score 2); (C) tongue lift when crying with an open mouth – minimal lift (score 0),

only the edges of the tongue are lifted toward the hard palate (score 1), or full tongue lifted toward the hard palate (score 2); and (D) tongue protrusion over the gum – the tip of the tongue can be behind the gum (score 0), over the gum (score 1), or extending over the lower lip (score 2). Each assessed item scores from zero to two and the result is obtained after summing the scores. A total score of zero to three is considered ankyloglossia, whereas four to eight is considered normal.

The Lingual Frenulum Evaluation Protocol for Infants (Tongue-Tie Test)⁽¹⁸⁾ has been validated in two versions. One of them considers the clinical history, the anatomic-functional assessment, and the non-nutritive and nutritive sucking assessment, to be applied in suspicious cases⁽¹⁹⁾. The other one, named screening, can be used in newborns before hospital discharge⁽²⁰⁾.

The final screening scores are obtained from summing all the items: (A) lip posture at rest – they can be closed (score 0), ajar (score 1), or open (score 1); (B) tongue positioning tendency when lifting it or crying – it can be lifted (score 0), halfway up (score 0), halfway up with lifted edges (score 2), or tip of the tongue low with lifted edges (score 2); (C) the shape of the tip of the tongue when lifted as they cry – it can be rounded (score 0), with a slight cleft on the apex (score 2), or shaped like a heart (score 3); (D) lingual frenulum: (D1) thickness – it can be thin (score 0) or thick (score 2); (D2) frenulum fixation on the ventral face of the tongue – it can be in the middle third (score 0), between the middle third and the apex (score 2), or at the apex (score 3); and (D3) fixation on the floor of the mouth as seen from the sublingual caruncles (score 0) or from the lower alveolar ridge (score 1). The scores range from zero to 12 – zero to four indicates a normal result, five to six indicates a suspicious result, and seven to 12 indicates an abnormal lingual frenulum (diagnostic of ankyloglossia)⁽¹⁸⁾.

The data were collected in the maternity ward, at an appropriate moment both to the mothers and newborns. It consisted of using the instruments to interview the mothers and obtain the sociodemographic data, as well as clinically examining the newborns' oral cavity.

The lingual frenulum assessment protocols – BTAT and Tongue-Tie Test – were applied in a random order, defined in a draw. The instruments were applied with an approximately one-minute interval between them.

The researcher conducted the lingual frenulum assessment tests wearing disposable vinyl gloves; each time the gloves were changed, the hands were disinfected with 70% alcohol-based hand sanitizer. The mother was asked to lay the nape of the newborn's neck on the space between her arm and forearm and hold its hands⁽¹⁸⁾. In this study, the newborn's position was standardized in the lingual frenulum examination with both diagnostic instruments. Since the BTAT does not indicate a specific assessment position, the one indicated by the Tongue-Tie Test was used⁽¹⁸⁾.

A specific maneuver was used to lift the newborn's tongue, in which the gloved index fingers were introduced under the tongue through the lateral margins to lift it and properly examine the lingual frenulum⁽²¹⁾. When manipulating the newborns' oral cavity, they tended to cry, which made it possible to assess the positioning of the tongue and the shape of the tip of the tongue.

As for the newborns that did not cry during the examination, these items were observed with the lifting maneuver.

The BTAT was published without describing how to assess tongue protrusion and elevation when the newborn is crying⁽¹⁷⁾. Both items require the examiner's familiarity with normal movements to identify them when they are abnormal⁽²²⁾. Thus, to assess tongue protrusion, the newborns were stimulated to suck with the researcher's gloved index finger on the tongue dorsum, slightly touching the hard palate. Then, the researcher's finger was gently directed toward the lower lip. This made the newborn's tongue follow the researcher's finger as if looking for the mother's nipple, making it possible to assess the tongue protrusion. The tongue elevation was assessed while the newborn was crying. If the newborn did not cry in the assessment, the tongue elevation maneuver used was the same as the Tongue-Tie Test. The BTAT does not consider the newborn's tongue positioned in the median line when crying; since this is considered a normal position in newborns⁽²³⁾, the score was considered as that of tongue lifted when crying.

The data were entered into Excel spreadsheets, using IBM SPSS version 23 for the statistical calculations. Descriptive analyses of the absolute and percentage frequencies were conducted for the categorical variables, as well as of mean, standard deviation, and median, for the age. The 5% significance level was used in the statistical tests. The two ankyloglossia diagnostic instruments were compared with the McNemar test. Ankyloglossia diagnosis was associated with the newborn's sex using Pearson's chi-squared test and Fisher's exact test.

RESULTS

Considering their profile, most of the mothers were 20 to 29 years old, had a monthly family income of up to one minimum wage, and had attended up to middle school. Their mean age was 26.8 years – the youngest was 15 years old and the oldest, 44 years old. As for the newborns, most were up to 3 days old and males. It was also verified that the earliest examination was made on the first day of life, while the latest was made on the 17th day of life (Table 1).

The resulting score of the examinations revealed an occurrence of 4.8% of diagnosed ankyloglossia with the BTAT, whereas, with the Tongue-Tie Test, it was of 17% – when the 14 (9.5%) suspicious cases (whose score was five and six) were considered normal for statistical analysis (Table 2) – and of 26.5% – when these cases were considered abnormal (Table 3).

A significant difference was verified between the two lingual frenulum assessment instruments ($p < 0.001$) in both statistical analyses (Tables 2 and 3).

When the suspicious cases were considered normal in the Tongue-Tie Test, only five examinations (3.4%) coincided with ankyloglossia diagnosis (Table 2). On the other hand, when the suspicious cases were considered abnormal, seven (4.8%) coincided with ankyloglossia diagnosis (Table 3).

There was an 85% agreement between the two instruments used to diagnose ankyloglossia.

Comparing the two diagnostic instruments, 13 (52%) out of the 25 cases of ankyloglossia diagnosed with the Tongue-Tie Test were

Table 1. Sociodemographic characteristics of the mothers and profile of the newborns

Variable	n	(%)
Sociodemographic profile		
Age group		
15 to 19	21	14.3
20 to 29	76	51.7
30 or older	50	34.0
Mother's schooling level		
Middle school	84	57.1
High school	58	39.5
Higher education	5	3.4
Monthly family income (minimum wages)		
Up to 1	107	72.8
Between 1 and 2	28	19.0
More than 2	12	8.2
Newborn's profile		
Days of life		
1 to 3	102	69.4
4 to 7	32	21.8
More than 8	13	8.8
Sex		
Males	78	53.1
Females	69	46.9
Total	147	100.0

n= sample number

males, while 12 were females (48%) when the suspicious cases were considered normal for the statistical analysis. However, when the suspicious cases were considered abnormal for the statistical analysis, 23 (59%) out of the 39 cases of ankyloglossia diagnosed with the Tongue-Tie Test were males, while 16 were females (41%). As for the occurrence of ankyloglossia considered with the BTAT, 71.4% were females and 28.6% were males. Comparing the cases of ankyloglossia identified with both instruments, 80% were females and 20% were males. Despite the different results found when using the two instruments, there was no association between ankyloglossia and the newborn's sex with either of the two assessment methods used (Tables 4 and 5).

DISCUSSION

Breastfeeding is one of the main health promotion practices, especially in less developed countries, which have low socioeconomic and human development indices. As a consequence, inadequate feeding and inefficient basic hygiene are still great causes of child mortality, particularly in the first year of life⁽¹⁵⁾.

According to the second Breastfeeding Prevalence Survey in the Brazilian Capitals and Federal District, the prevalence of newborns' exclusive breastfeeding in Recife was 38.3%, below the Brazilian average (41%)⁽²⁴⁾.

Table 2. Ankyloglossia diagnosis according to the two assessment instruments, considering the suspicious cases as normal in the Tongue-Tie Test

Bristol	Tongue-Tie Test				Total	p-value	
	With ankyloglossia		Without ankyloglossia				
	n	%	n	%			
With ankyloglossia	5	3.4	2	1.4	7	4.8	p < 0.001*
Without ankyloglossia	20	13.6	120	81.6	140	95.2	
Total	25	17.0	122	83.0	147	100.0	

McNemar test; *Statistical significance; p-value = significance probability.

Table 3. Ankyloglossia diagnosis according to the two assessment instruments, considering the suspicious cases as abnormal in the Tongue-Tie Test

Bristol	Tongue-Tie Test				Total		p-value
	With ankyloglossia		Without ankyloglossia		n	%	
	n	%	n	%			
With ankyloglossia	7	4.8	-	-	7	4.8	p < 0.001*
Without ankyloglossia	32	21.8	108	73.5	140	95.2	
Total	39	26.5	108	73.5	147	100.0	

McNemar test; *Statistical significance

Table 4. Ankyloglossia diagnosis screened with the Lingual Frenulum Evaluation Protocol for Infants – Tongue-Tie Test, according to the newborn's (NB) sex

NB sex	Lingual frenulum diagnosis						p-value	OR (95% CI)
	With ankyloglossia		Without ankyloglossia		Total			
	n	%	n	%	n	%		
Males	13	16.7	65	83.3	78	100.0	p = 0.907	1.00
Females	12	17.4	57	82.6	69	100.0		
Total Group	25	17.0	122	83.0	147	100.0		

Pearson's chi-squared test

Table 5. Ankyloglossia diagnosis with the BTAT, according to the newborn's (NB) sex

NB sex	Lingual frenulum diagnosis						p-value	OR (95% CI)
	With ankyloglossia		Without ankyloglossia		Total			
	n	%	n	%	n	%		
Males	2	2.6	76	97.4	78	100.0	p = 0.254	**
Females	5	7.2	64	92.8	69	100.0		
Total Group	7	4.8	140	95.2	147	100.0		

Fisher's Exact test; **It could not be established because the occurrences had a very low frequency; OR = Odds Ratio.

Some authors studied the factors for early weaning, considering those related both to the mother (such as low schooling and socioeconomic level) and to the newborn (such as incorrect latch onto the breast and presence of ankyloglossia)^(6-8,25). The importance of an early ankyloglossia diagnosis is thus emphasized as a means to avoid early weaning – especially in this highly vulnerable population to maintain breastfeeding, as most of them had a monthly family income of up to one minimum wage and more than half of them attended only up to middle school.

Correct ankyloglossia diagnoses require a single, validated, specific, functional, objective protocol, easy to be used by the various health professionals, so the results found can be standardized⁽³⁾.

In the present study, a statistically significant difference was observed between the two instruments, with a lower prevalence of ankyloglossia diagnosed with the BTAT.

All participating newborns were submitted to early ankyloglossia diagnosis, before hospital discharge, and most of them were examined in the first three days of life – as recommended by the Ministry of Health⁽²⁵⁾. The lingual frenulum assessment protocols were applied in a random order, defined in a draw, to avoid information, record, and data analysis biases. Early ankyloglossia diagnosis aims to, depending on each case, support breastfeeding or refer the newborn to surgery to release the lingual frenulum, to avoid weaning⁽¹⁴⁾.

Studies indicate a higher prevalence of ankyloglossia in males^(1,12,26,27). However, in this study, as also evidenced in the study by Lima et al.⁽²⁸⁾, there was no significant association between the prevalence of ankyloglossia and the newborn's sex.

A study conducted in Canada revealed that the ankyloglossia incidence rates increased from 6.86 per 1,000 live births in 2002 to 22.6 per 1,000 live births in 2014 – when the maternities began examining the frenulum⁽²⁶⁾. A study carried out in Spain revealed that the estimated ankyloglossia prevalence in the region was two to three times lower than what was found after diagnosing it with a standardized instrument (Hazelbaker)⁽¹⁵⁾. A similar result was found in a study conducted in Thailand after using a standardized diagnostic protocol (Kotlow's assessment)⁽⁸⁾.

Therefore, although no instrument is considered the gold standard to diagnose this condition, these data suggest that ankyloglossia was being underreported. Hence, using validated instruments along with neonatal lingual frenulum screening increases the diagnostic capacity and leads to better treatment.

Concerning the instruments used in this study, the BTAT is practical, objective, and easy to use⁽¹⁷⁾. It proposes quick diagnosis

and screening criteria for severe ankyloglossia, and the Ministry of Health considers it an appropriate protocol for neonatal screening⁽²⁵⁾. On the other hand, this instrument did not finish all the stages in the validation process, according to international norms. As for its validation for use in Brazil, the translation and transcultural adaptation stages were not carried out⁽²⁹⁾.

It was observed in this study that the Bristol instrument has a serious limitation regarding the tongue protrusion criterion. Besides being a difficult item to assess, as the newborn will not always protrude the tongue at the examination, many newborns with ankyloglossia can protrude the tongue as far as the lip or gum and still have limited movement amplitude. However, scoring this item increases the total above three, indicating a normal result. It must be said that tongue protrusion is not considered important to orofacial functions, whereas its elevation is important to most of these functions. Also, as the lingual frenulum is in the ventral face of the tongue, the tongue must be lifted to correctly inspect it and diagnose ankyloglossia.

A point worth highlighting is that the most severe case of ankyloglossia diagnosed with the Tongue-Tie Test scored 10 (severe cases are those that score above seven) but this very case was not diagnosed as ankyloglossia with the BTAT, in which it scored five (severe cases are those that score below three). Hence, it is suggested that the BTAT needs a revision.

The prevalence found with the Tongue-Tie Test (17%) was near the one found in another study conducted in Brazil (21%), which had a sample of 100 newborns⁽²⁰⁾. Considering only the suspicious cases (scores five and six), the results were identical in both studies (9.5%).

A differential between the two instruments assessed in this study is the definition of suspicious scores, which are only considered in the Tongue-Tie Test. The suspicious cases are reassessed when they are 30 days old to consider their breastfeeding difficulties and then interpret the results and refer for treatment.

In this regard, considering the suspicious cases as normal with the Tongue-Tie Test revealed that only five examinations coincided with ankyloglossia diagnosis with both instruments. On the other hand, when the suspicious cases were considered abnormal, all the cases of ankyloglossia diagnosed with the BTAT were likewise diagnosed with the Tongue-Tie Test. Thus, the prevalence of ankyloglossia was higher when the suspicious cases were considered abnormal.

A Brazilian study, whose authors used the two assessment instruments, verified that the ankyloglossia diagnosis coincided in 3.11%. They also concluded that both instruments are effective to correlate ankyloglossia with breastfeeding difficulties⁽²⁹⁾.

Nonetheless, according to the authors of the technical-scientific evaluation report of the *Instituto de Saúde da Criança de São Paulo* (Child Health Institute of São Paulo), the association between newborns with ankyloglossia and breastfeeding difficulties cannot be evidenced with the BTAT (which is recommended by the Ministry of Health for neonatal screenings) because this instrument does not have reliable results⁽¹¹⁾.

The main limitations of this study are related to the fact that a great part of the newborns was asleep or sleepy at the examination, which is expected due to their few days of life. However, this may have interfered with some assessments, such as that of tongue protrusion and elevation movements when crying. To diminish any biases, the babies were woken up and stimulated to suck, with the researcher's gloved index finger on the tongue dorsum and gently touching the hard palate, before assessing the scores. Another limitation is that only one calibrated researcher conducted the assessments, with agreement analysis with other assessors in 20% of the sample.

These data reinforce the importance of studying the association between ankyloglossia and breastfeeding, with an adequate diagnosis of the condition with instruments not only validated but also effective. Longitudinal studies relating ankyloglossia to the correct latch onto the mother's nipple and continuing breastfeeding are needed, as well as further clinical studies comparing the two assessment instruments to point out the most effective one in neonatal screenings.

CONCLUSION

The ankyloglossia diagnosis in newborns varied depending on the assessment instrument used. Ankyloglossia was detected less often with the BTAT than with the Lingual Frenulum Evaluation Protocol for Infants (Tongue-Tie Test).

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Author contributions

MRBAF and VAM conceived and designed the study; MRBAF and TCBL performed the data collection; MRBAF analyzed the data; VAM and KAB contributed to the analysis; MRBAF and TCBL generated the database; MRBAF and KAB composed the illustrations; MRBAF and TCBL generated the database; MRBAF, VAM and KAB wrote the paper.