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Association between ankyloglossia and breastfeeding

Associação entre anquiloglossia e amamentação

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Keywords

Breastfeeding
Lingual Frenulum
Newborn
Sucking Behavior
Tongue

ABSTRACT

Purpose: To analyze the association between ankyloglossia and breastfeeding. **Methods:** A cross-sectional study was undertaken on 130 newborn infants in exclusive breastfeeding with Apgar score ≥ 8 within the first five days of life. The research was approved by the Ethics Committee on Human Research. The data collection was performed by the researcher and by three trained speech therapists of the team. The protocols applied were the Neonatal Tongue Screening Test from the Lingual Frenulum Protocol for Infants, the UNICEF Breastfeeding Observation Aid, and the collection of maternal complaints related to the difficulty in breastfeeding was also considered. The data were submitted to statistical analysis – chi-square test and Fisher's exact test, with a significance level of 5%. **Results:** When correlating the data, the statistical analysis revealed an association between ankyloglossia and the items of suckling category of the Breastfeeding Observation Aid. The association between complaint of difficulty in breastfeeding and ankyloglossia was also seen. **Conclusion:** On the first days of life, ankyloglossia is associated with the mother's breastfeeding complaint and with the newborn's sucking difficulty.

Descritores

Aleitamento Materno
Freio Lingual
Recém-Nascido
Comportamento de Sucção
Língua

RESUMO

Objetivo: Verificar associação entre anquiloglossia e amamentação. **Método:** Estudo transversal, realizado em 130 recém-nascidos, em um Hospital Universitário. Foram incluídos recém-nascidos entre um a cinco dias de vida, com Apgar score ≥ 8, em aleitamento materno exclusivo. Participaram da pesquisa apenas recém-nascidos de termo e saudáveis. Esta pesquisa foi aprovada pelo Comitê de Pesquisa com Seres Humanos. A coleta de dados foi realizada pela pesquisadora e por três fonoaudiólogas da equipe, devidamente treinadas e calibradas. Os protocolos aplicados foram: avaliação anatomofuncional do Protocolo de avaliação do frênulo da língua em bebês, o Protocolo de Observação da Mamada da UNICEF e coleta das queixas maternas referentes a dificuldade ou não para amamentar. Os dados obtidos foram submetidos à análise estatística, sendo aplicado o teste Qui-quadrado e teste exato de Fisher, adotando nível de significância de 5%. **Resultados:** Quando correlacionados os dados, a análise estatística demonstrou associação entre anquiloglossia e os itens da categoria de sucção do Protocolo de Observação da Mamada. Encontrou-se também associação entre queixa de dificuldade para amamentar e anquiloglossia. **Conclusão:** Nos primeiros dias de vida, a anquiloglossia está associada com queixa da mãe para amamentar e com a dificuldade de sucção do recém-nascido.

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INTRODUCTION

The advantages of breastfeeding for the puerperal woman and for the newborn's life are widely discussed in the literature[1]. Breast milk prevents childhood obesity[2], the development of atopic diseases[3], in addition to help in the childhood intellectual and cognitive performance[4,5].

According to the World Health Organization (WHO), the exclusive breastfeeding for a 6-month period and the complementary feeding up to two years old or more are key to reach the advantages mentioned above[6]. In this context, besides being conscious of the advantages of breastfeeding for the baby and his mother, every health professional who assists the mother/infant dyad must be aware of the prevention and management of the main problems that can occur during breastfeeding, aiming at the prevention of early weaning.

The causes of early weaning are multifactorial; the literature reports nipple trauma[7], the family's socioeconomic and intellectual issues, lack of specific guidance by health professionals, use of pacifiers and artificial formulas, and the mother's return to work[8,9].

A study conducted by Kent et al.[10] concluded that the most common cause of discontinuity of exclusive breastfeeding is the insufficient offering of milk during the first month, as breastfeeding becomes more efficient between the first and third month of lactation.

Another study followed 1344 mother/baby dyad for six months and concluded that the determining factors for early weaning are limiting the number of feedings at night, the lack of guidance in prenatal care and the presence of nipple trauma[11].

In view of this reality, strategies for household follow-up by means of guidelines and interventions in the breastfeeding process and individual or group counseling of pregnant and puerperal women are carried out inside hospitals and soon after hospital discharge to help in the intercurrences during breastfeeding, with the aim of avoiding early weaning[9,12].

By contrast, in Brazil, despite the improvement in exclusive breastfeeding rates in the first six months of life[13,14], the percentage achieved falls short of that recommended by the WHO[15]. In the period from 1999 to 2008, a study carried out with the objective of checking the current situation of breastfeeding in the Brazilian capitals and the Federal District revealed a prevalence of 41%, with average duration of exclusive breastfeeding of 54.1 days (1.8 months) and average duration of breastfeeding of 341.6 days (11.2 months)[15].

Although many articles have been published in the last 15 years showing the interference of ankyloglossia with breastfeeding[16-24], as well as the importance of the anatomophysiology involved in the coordination of suckling, deglutition, and breathing of newborns for breastfeeding, many factors still need to be evaluated since the duration of breastfeeding lies behind that recommended by the WHO. Most studies report an association between ankyloglossia and maternal complaint of nipple pain for breastfeeding, as well as handling difficulty[16,18-21,24-26]. Thus, the hypothesis of this study was that ankyloglossia, in addition to the above mentioned signs and symptoms, interferes with sucking during breastfeeding.

The objective of the present study was to analyze the association between ankyloglossia and breastfeeding.

METHODS

Study population and design

Initially, the sample calculation was performed considering the significance level of 5% and test power of 90%. The result showed the need for the evaluation of 117 newborns; considering possible sample losses, a sample calculation of 130 infants was made.

Thus, 130 newborns within one to five days of life from the accommodation set mother-newborn (Common Lodging Ward) of the School Hospital of the Federal University of Mato Grosso do Sul (UFMS) were evaluated from June to December 2016. The research was approved by the Ethics Committee on Human Research under n. 1.514.715. All newborn's parents or guardians included in the research were informed of the study's objectives and procedures and invited to sign the Informed Consent Form agreeing to participate in the study.

The inclusion criteria comprised term newborns with Apgar score ≥ 8 and exclusive breastfeeding. Infants with the following conditions were excluded from the study: preterm newborns, newborns with perinatal complications, craniofacial anomalies, neurological diseases, genetic syndromes visible at the time of evaluation and artificial feeding, newborns of HIV positive postpartum period and newborns with unstable clinical conditions. Those descendants from indigenous or quilombolas (descendants of Afro-Brazilian slaves) parents were also excluded.

Procedures

Evaluations of the newborns were performed after 24 hours of birth. Data collection was done by the researcher and by three speech therapists from the UFMS University Hospital team, duly trained by the researcher. For this phase, a pilot study was carried out, with the participation of 14 newborns. At the end of training, the researchers acquired a degree of agreement above 90% in the evaluations.

The Neonatal Tongue Screening Test from the Lingual Frenulum Protocol for Infants (LFPI), developed and validated by Martinelli[27], was applied. The application was filmed with a Sony DSC-HX1 digital camera close to the feeding time.

Initially, the resting lip position was evaluated while the infant was sleeping; the lips could be closed, open or half-opened. Then, with the newborn awake, it was observed if the positioning of the tongue during crying was in the midline, elevated, in the midline with the elevation of the sides or tongue apex down with elevation of the sides. The shape of the tongue apex when raised during crying or elevated maneuvers could be round, with a slight crevice by V-shaped, or heart-shaped. Next, it was observed whether it was possible or not to visualize the lingual frenulum, or to visualize it with the maneuver. The frenulum thickness was classified as thin or thick. The frenulum attachment to the sublingual face of the tongue could be in the middle third, between the middle third and the apex, or in the apex. The frenulum attachment to the mouth floor could be visible

from the sublingual caruncles or the inferior alveolar crest. When the sum of the evaluated items was equal to or less than 4, it was considered normal; between 5 and 6, doubtful; 7 or more, altered, with the lingual frenulum limiting the tongue movements (Figure 1).

The Breastfeeding Observation Aid[28] was applied during breastfeeding, and six categories were evaluated: mother (healthy or ill/depressed, relaxed and comfortable or tense and uncomfortable, signs of bonding between mother and child or no eye contact between mother and child); baby (healthy or

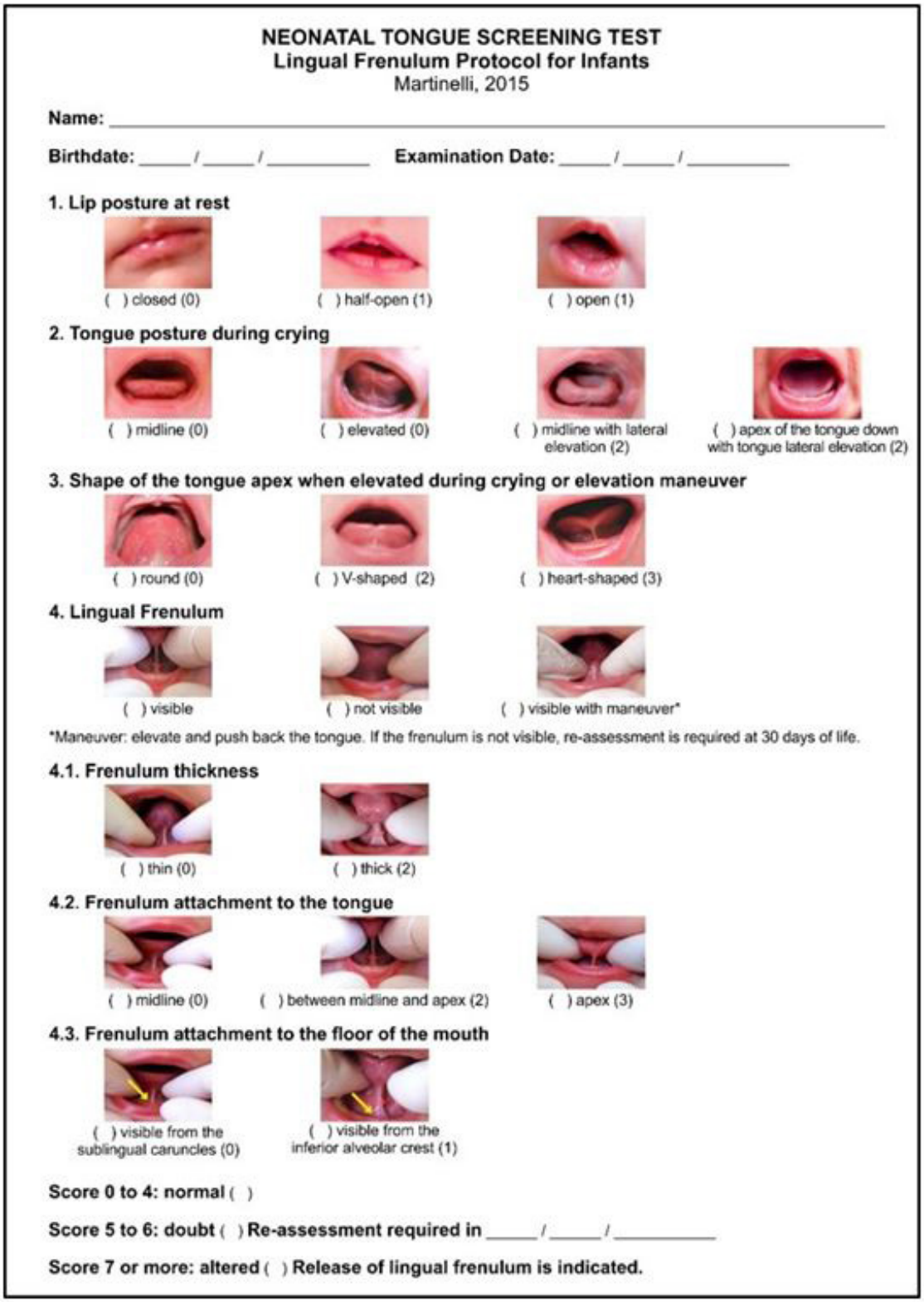

Figure 1. Neonatal Tongue Sreening TEST, Martinelli[27]

sleepy/ill, calm and relaxed or restless/crying, trying to reach or root for the breast if hungry or does not reach or root for the breast); breasts (healthy or looking red/swollen/sore, presence of pain or discomfort, whether the breast was well supported with the fingers away from the nipple or on the areola); baby's position (head and body in line/neck and head twisted to feed, baby held/not held close to mother's body, baby's whole body supported/supported by the head and neck only, baby approaches breast, nose/lower lip or chin to nipple); baby's attachment (more areola seen above baby's top lip or below bottom lip, baby's mouth wide opened/not wide opened, lower lip turned outwards/lips pointing forward or turned in, baby's chin touches breast or not); and suckling (slow deep/rapid shallow sucks with pauses, cheeks round when suckling/cheeks pulled in when suckling, baby releases breast when finished/mother takes baby off the breast, mother notices signs of oxytocin reflex or not). The categories of this protocol indicate favorable and unfavorable behaviors, pointing to normal or difficult beginning of breastfeeding, that is, with or without signs of difficulties in breastfeeding. A single item marked from the signs of possible difficulty is already considered an indication that may jeopardize breastfeeding (Chart 1).

Both the Neonatal Tongue Screening Test from the Lingual Frenulum Protocol for Infants and the Protocol of Breastfeeding Observation Aid were applied by two speech therapists *in loco*. The application was filmed and later analysed by a third speech therapist. The results of the protocols were defined by consensus of the three speech therapists.

The collection was also performed through the response of the mother/guardian for the newborn, with or without complaint of difficulty in breastfeeding the newborn. The mother/guardian answered yes or no to the following question: Are you having difficulty in breastfeeding?

The cases considered altered were discussed by an interdisciplinary team comprised of speech therapists, pediatricians and a maxillofacial surgeon and, after the parents' guidance and consent, the newborns were referred for surgery.

The data were tabulated and submitted to statistical analysis. The chi-square test and Fisher's exact test were used, adopting a significance level of 5%.

RESULTS

Data of newborns and lingual frenulum

The results obtained through the application of the Neonatal Tongue Screening Test from the LFPI revealed that of the 130 newborns, 105 (81%) presented normal frenulum and 25 (19%) altered frenulum, that is to say, with ankyloglossia.

The results obtained through the application of the Breastfeeding Observation Aid are described in Table 1.

Table 2 shows the results of the comparison between the data obtained with the application of the Neonatal Tongue Screening Test from the LFPI and the Breastfeeding Observation Aid.

Of the 130 newborns, 44 did not present ankyloglossia or signs of difficulty in suckling according to the Breastfeeding Observation Aid; however, all 25 newborns detected with

Chart 1. Breastfeeding Observation Aid (WHO, UNICEF)[28]

No signs of difficulties	Signs of possible difficulties
MOTHER:	
( ) looks healthy	( ) looks ill or depressed
( ) relaxed and comfortable	( ) looks tense and uncomfortable
( ) signs of bonding between mother and baby	( ) no mother/baby eye contact
BABY	
( ) looks healthy	( ) looks sleepy or ill
( ) calm and relaxed	( ) is restless or crying
( ) reaches or roots for the breast if hungry	( ) does not reach or root
BREAST	
( ) looks healthy	( ) is red, swollen or sore
( ) no pain or discomfort	( ) breast or nipple painful
( ) well supported with fingers away from nipple	( ) held with fingers on areola
BABY'S POSITION	
( ) baby's head and body in line	( ) neck and head twisted to feed
( ) baby held close to mother's body	( ) baby not held close to mother's body
( ) baby's whole body supported	( ) baby supported by head and neck only
( ) baby approaches breast, nose to nipple	( ) baby approaches breast, lower lip/chin to nipple
BABY'S ATTACHMENT	
( ) more areola seen above baby's top lip	( ) more areola seen below bottom lip
( ) mouth wide opened	( ) mouth not wide opened
( ) lower lip turned outwards	( ) lips pointing forward or turned in
( ) chin touches breast	( ) chin not touching breast
SUCKLING	
( ) slow, deep sucks with pauses	( ) rapid shallow sucks
( ) cheeks round when suckling	( ) cheeks pulled in when suckling
( ) baby releases breast when finished	( ) mother takes baby off the breast
( ) mother notices signs of oxytocin reflex	( ) no signs of oxytocin reflex noticed

ankyloglossia (100%) by the Neonatal Tongue Screening Test from LFPI presented signs of possible suckling difficulties by the Breastfeeding Observation Aid. The statistical analysis showed an association between ankyloglossia and the suckling category of the Breastfeeding Observation Aid, indicating that, in the presence of ankyloglossia, the suck was faster/shallower, with cheeks pulled in when suckling, as well as with the mother taking the baby off the breast, with no signs of oxytocin reflex noticed by the mother ($p < 0.001$*).

Of the 130 mothers interviewed according to the methodology established by this research, 93 (72%) reported no difficulties in breastfeeding and 37 (28%) did so. When comparing the results of the Neonatal Tongue Screening Test from the LFPI with complaints of difficulty in breastfeeding reported by mothers, of the 37 mothers who reported difficulties, 12 (32%) had newborns with ankyloglossia. Statistical analysis revealed an association between ankyloglossia and complaint of difficulty in breastfeeding reported by the mother ($p = 0.016$) (Table 3).

DISCUSSION

In addition to the mothers' answers about the difficulty in breastfeeding, the present study combined two specific protocols, one to diagnose the presence of ankyloglossia and another to diagnose possible difficulties in breastfeeding.

When comparing the data, the statistical analysis showed that ankyloglossia in newborns is associated with difficulty in suckling, i.e. the probability of newborns present signs of difficulty in sucking was 36.07 times greater for babies with ankyloglossia than for babies without ankyloglossia, corroborating with the study by Martinelli et al.[18] who reports that babies with ankyloglossia have a lower number of suctions, as well as a longer pause time between suction groups. A study conducted by ultrasonography reported the importance of tongue movements to remove milk from the nipples during breastfeeding. It was observed that in order to extract the milk from the nipple, the tongue rises towards the palate to create the vacuum and when

Table 1. Absolute and relative frequencies for the category variables of the UNICEF Breastfeeding Observation Aid[28]

Category	With no signs of difficulty	Possible signs of difficulty
Mother	111 (85%)	19 (15%)
Baby	106 (82%)	24 (18%)
Breast	56 (43%)	74 (57%)
Baby's position	85 (65%)	45 (35%)
Breast attachment	44 (34%)	86 (66%)
Suckling	44 (34%)	86 (66%)

Table 2. Comparison between the data of the Neonatal Tongue Screening Test from the LFPI and the Breastfeeding Observation Aid[28] using the chi-square and Fisher's exact tests

Breastfeeding Observation Aid		Without ankyloglossia		With ankyloglossia		Value-p	O.R.	C.I.-95%
Mother (General)	No signals of difficulties	91	82%	20	18%	0.396	1	-
	Signals of possible difficulties	14	74%	5	26%		1.63	(0.53; 5.03)
Baby (General)	No signals of difficulties	85	80%	21	20%	1.000	1	-
	Signals of possible difficulties	20	83%	4	17%		0.74	(0.28; 2.69)
Breast	No signals of difficulties	43	77%	13	23%	0.316	1	-
	Signals of possible difficulties	62	84%	12	16%		0.64	(0.27; 1.54)
Baby's Position	No signals of difficulties	69	81%	16	19%	0.871	1	-
	Signals of possible difficulties	36	80%	9	20%		1.08	(0.43; 2.68)
Baby's Attachment	No signals of difficulties	36	82%	8	18%	0.828	1	-
	Signals of possible difficulties	69	80%	17	20%		1.11	(0.44; 2.82)
Suckling	No signals of difficulties	44	100%	0	0%	<0.001	1	-
	Signals of possible difficulties	61	71%	25	29%		36.06	(2.19; 622.4)

Caption: O.R. = Odds Ratio; C. I. = Confidence interval

Table 3. Comparison between the data of the Neonatal Tongue Screening Test from the LFPI with and without complaints of difficulty in breastfeeding reported by mothers, using the chi-square and Fisher's exact tests

Variable	Without ankyloglossia		With ankyloglossia		Value-p	O.R.	I.C-95%
Complaint without difficulty	80	86%	13	14%	0.016	1	-
Complaint with difficulty	25	68%	12	32%		2.95	(1.2;7.3)

Caption: O.R. = Odds Ratio; I.C. = Confidence Interval

the tongue lowers, the vacuum increases, so the nipple expands and the milk flows into the oral cavity[29]. The ultrasonography has also revealed that babies with ankyloglossia present stronger compression of the nipple by the tongue resulting in pain[19], and reducing the effectiveness of milk sucking during breastfeeding by limiting the tongue movements provided by ankyloglossia[28].

A significant association between ankyloglossia and the difficulty in breastfeeding reported by the mother was seen (p = 0.016*), and the statistical analysis showed that the probability of mothers of newborns with ankyloglossia report a complaint of difficulty in breastfeeding is 2.95 times greater than the mothers of newborns without ankyloglossia, confirming the results obtained in several studies[16,20,21,25]. A broad question was chosen, with a simple answer, only to verify the presence or absence of difficulty in breastfeeding, since any difficulty can lead to early weaning.

These findings corroborate the literature that reports that ankyloglossia may contribute to the early weaning since the difficulty in suckling can cause discomfort, pain and nipple injury in the mothers; and lower intake of milk in ml for 24 hours in the infants, thus being necessary to be breastfed more frequently[16,20,22-24,26]. In addition, it is also stated in studies that the mothers of infants with ankyloglossia reported distress and tension caused by the lack of explanation and advice from health professionals about the consequences of this change in breastfeeding[25].

Thus, this study led to the association of ankyloglossia and the newborns' greater difficulties in initiating the breastfeeding by means of the Breastfeeding Observation Aid and the complaint of difficulty referred by the mother.

However, the limitations of this research should be pondered in view of the reduced number of newborns with ankyloglossia, which allows considering the results found only for the population concerned.

More studies that associate difficulties in the coordination of suckling, swallowing and breathing, and the anatomo-functional alterations in clinically stable newborns are necessary, since the duration percentage of exclusive breastfeeding in Brazil is below that recommended by the WHO.

CONCLUSION

In the first days of life, ankyloglossia is associated with the mother's complaint and with the newborn's suckling difficulty. This may be a risk factor for the successful breastfeeding.

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Author contributions

SMAC had primary responsibility for protocol development, patient screening, enrolment, outcome assessment, preliminary data analysis and writing the manuscript; DBP and RLCM participated in the development of the protocol and analytical framework for the study and contributed to the writing of the manuscript.