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Presence of ankyloglossia and breastfeeding in babies born in Lima, Peru: a longitudinal study

Presença de anquilglossia e amamentação em bebês nascidos em Lima, Peru: um estudo longitudinal

Keywords

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Descritores

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ABSTRACT

Purpose: to evaluate the lingual frenulum and breastfeeding in infants from a maternal-perinatal referral center, as well as to monitor infants with ankyloglossia up to six months of age. **Methods:** a cohort study conducted at the Instituto Nacional Materno Perinatal - Maternidad de Lima in Lima, Peru. The consecutive intentional sample consisted of 304 newborns and their respective mothers, evaluated during December 2017 and January 2018, which were the baseline of the study. A clinical evaluation of the lingual frenulum adapted and the Clinical Evaluation of Breastfeeding Efficacy (CEBE) scale, was performed. **Results:** of the 304 newborns, 15 (4.9%) were considered with altered frenulum, and only 4 (26.7%) presented a low score in CEBE. The mean of the CEBE score was 9.3. (DP=1.35, Min=3, Max=10). Of the follow-up infants, only 2 (13.3%) persisted with breastfeeding difficulties for which frenotomy was indicated. **Conclusion:** This study demonstrates the low prevalence of ankyloglossia in infants, as it does not indicate a trend of difficulty or negative interference in breastfeeding.

RESUMO

Objetivo: avaliar o frênulo lingual e a amamentação em lactentes de um centro de referência materno-perinatal, bem como monitorar lactentes com anquilglossia até seis meses de idade. **Método:** estudo de coorte realizado no Instituto Materno Perinatal em Lima, Peru. A amostra intencional consecutiva foi composta por 304 recém-nascidos e suas respectivas mães, avaliados entre dezembro de 2017 e janeiro de 2018, que foram a linha de base do estudo. Foi realizada uma avaliação clínica do frênulo lingual adaptado e a escala de Avaliação Clínica da Eficácia da Amamentação (ACEA). **Resultados:** dos 304 recém-nascidos, 15 (4,9%) foram considerados com frênulo alterado e apenas 4 (26,7%) apresentaram escore baixo no ACEA. A média do escore ACEA foi de 9,3. (DP = 1,35, Min = 3, Max = 10). Dos lactentes de acompanhamento, apenas 2 (13,3%) persistiram com dificuldades de amamentar para os quais foi indicada frenotomia. **Conclusão:** Este estudo demonstra a baixa prevalência de anquilglossia em lactentes, pois não indica tendência de dificuldade ou interferência negativa no aleitamento materno.

Study conducted at: Instituto Nacional Materno Perinatal - Maternidad de Lima, Peru and at the Programa de Pós-Graduação em Epidemiologia, Faculty of Medicine Universidade Federal do Rio Grande do Sul, Porto Alegre (RS), Brasil and at the Graduate Studies Program in Dentistry, Universidade Federal do Rio Grande do Sul, Porto Alegre, Brasil.

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INTRODUCTION

According to the World Health Organization, the practice of exclusive breastfeeding is recommended until the child is six months old and should be offered as a supplement until the child is two years old⁽¹⁾. Breastfeeding involves several physiological, emotional and social aspects, which contribute to the growth and development of the child, as well as to the construction of the relationship and the bond between the mother and the baby⁽²⁻⁵⁾. Breast milk provides all the nutrients the baby needs during the first few months of life⁽⁴⁾.

Breastfeeding is one of the main focuses of investigation nowadays and fostering breastfeeding has been a priority in health programs and public policies globally⁽⁶⁻¹⁰⁾. This practice is a topic of discussion among professionals and researchers in the health area, especially in cases of ankyloglossia. However, there are still doubts concerning clinical approach in its presence^(11,12). There is a lot of uncertainty concerning the real recommendations, the real harm to the baby and the mother, as well as regarding the necessary procedures⁽¹³⁾. Studies monitoring the natural history of ankyloglossia have not been found in the literature.

Considering that ankyloglossia can possibly result in consequences for the baby such as difficulty in breastfeeding, early and timely identification is important, mainly for the promotion of breastfeeding⁽¹⁴⁻¹⁵⁾. In addition, breastfeeding, on its turn, will promote better growth and development of facial structures, as well as the maturation of the functions that compose the stomatognathic system⁽¹⁶⁾. Moreover, breastfeeding is a possibly predictor of the development of the entire functionality of oral structures during the child's growth.

Ankyloglossia in infants has been associated with failure to growth, breast damage and maternal breast pain, mastitis, engorgement and breast rejection⁽¹⁷⁻²⁰⁾. A systematic review that investigated the effects of ankyloglossia on breastfeeding and speech has revealed that ankyloglossia provides objective and subjective harm in breastfeeding. However, there is a limited number of studies providing quality, non-biased evidence⁽²¹⁾. Even though there are studies investigating these relations in the literature, further research is needed to clarify their findings.

Therefore, further studies are necessary to highlight the factors associated to ankyloglossia and the need for early intervention. The aim of this study was to evaluate babies' lingual frenulum and breastfeeding at a maternal-perinatal reference center in Maternal Perinatal Institute in Lima, Peru, and to monitor infants with ankyloglossia until six months of age.

METHODS

Cohort study conducted at the Instituto Nacional Materno Perinatal - Maternidad de Lima, Peru, institution of greater medical-surgical complexity for maternal and perinatal care in Peru. To calculate the size of the sample, a 50% proportion of the population was used, as the value of ankyloglossia prevalence is not yet consensus in world literature. Since this is the safest estimate, as it corresponds to the larger sample size that may be calculated, we chose to estimate on the basis of this proportion. A 6% error was used and adding

10% for eventual refusals, resulting in a final sample of 275 participants. The intentional consecutive sample was composed by 304 newborns and their respective mothers, evaluated in December 2017 and January 2018, forming the study baseline. High-risk pregnancies, premature or intensive care unit babies with any neurological or craniofacial disorder, as well as mothers who were under 18 years-old were excluded mothers who did not give consent.

A pilot study was carried out for training and standardization of data collection. To identify personal characteristics, a guided questionnaire was used on the study baseline: age (in hours), sex (female, male), and weight of the newborn (in grams); gestational age (in weeks); age (in years) and education of the mother (incomplete elementary school, complete elementary school, incomplete secondary education, complete secondary education, incomplete technical or higher education, undergraduate, graduate), as well as prenatal follow-up (yes, no). To clinically evaluate breastfeeding, the Clinical Evaluation of Breastfeeding Efficacy scale was used (CEBE) (Minimum value = 0 - low efficacy; and maximum = 10 - high efficacy)⁽²²⁾ translated and validated in the Spanish version.

For the evaluation of the lingual frenulum of infants, a list of clinical criteria, carefully selected by the researchers was used based on the Lingual Frenulum Protocol for Infants (LFPI), proposed and developed by Martinelli et al., 2012 in its Brazilian version⁽²³⁾, but adapted by the authors for the Spanish version: (position of the lips at rest, visibility, thickness and lingual frenulum attachment, tongue position during crying, and shape of tongue tip when elevated during crying), which generated a functionality score (Minimum value = 0 - worse functionality and maximum = 12 - best functionality). 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⁽²³⁾. Babies scoring equal or greater than 7 were considered as having alterations in the lingual frenulum and were eligible to follow-up. All interviews and clinical examinations were performed by a speech therapist trained for the study evaluation parameters.

Out of the 304 newborns and their respective mothers, 15 were diagnosed with change to for monthly follow-up, until the sixth month of the child's life, as they presented a score equal or greater than 7, compatible with frenulum change. A minimum of 5 calls to all contacts made available at the time of the guided interview (mother, family and friends) were made to schedule the baby and the mother follow-up at the study referral hospital. At each month, the baby was weighted and breastfeeding was reanalyzed by a qualified pediatric dentist using the same initial study evaluation criteria. Cases of low weight gain and breastfeeding harm were referred to frenotomy of the lingual frenulum by the pediatric dentistry team of the Maternity Hospital (2 cases).

A descriptive analysis through absolute and relative frequency measurements was carried out, and the distribution of variables through medians and standard deviations was

calculated. The SPSS version 21.0 (SPSS Inc., Chicago, IL) was used.

This research was approved by the Ethics in Research Committee of the Instituto Nacional Materno Perinatal - Maternidad de Lima in Lima, Peru under legal opinion Exp. No. 16873. All responsible individuals involved signed the Informed Consent Form.

RESULTS

The total sample consisted of 304 newborns, with a mean birth weight of 3351.7 (DP \pm 654.9) grams, most of them male (55%). Their respective mothers had an average age of 28.2 (DP \pm 6.2) years, most of them with complete secondary education (50.8%) and went to childbirth with an average gestational age of 39.11 (DP \pm 1.14) weeks. There were 15 (4.9%) newborns who were considered altered frenulum cases and 3

(1.0%) cases of neonatal teeth were found. The CEBE score presented an average of 9.3. (DP \pm 1.35; Min=3, Max=10). It is important to emphasize that the evaluations occurred in newborns with only two days of life. Table 1 presents the characterization of the sample.

Considering the total cases of ankyloglossia, in the evaluation carried out in the first hours of life, only 4 (26.7%) presented a low score on CEBE, characterizing inappropriate breastfeeding. In the first month of follow-up, 8 cases were lost, which were living in other cities or had changed telephone even after several efforts to continue. Out of those 8 absent babies, 7 presented maximum score in the CEBE scale (10), and only one presented a score of 5, considered as inappropriate breastfeeding in the study baseline. Among the babies that continued, only 2 (13.3%) persisted in breastfeeding difficulties in the first month of life. The 2 cases that persisted in breastfeeding difficulties were referred to frenotomy (Table 2).

Table 2 describes the characteristics of the monitored babies.

Table 1. Sample characterization. Lima, Peru. 2018. (n=304)

Variable	Values (n=304)
Sex (M/F)	167/137
Birth weight (grams)	3351.7 (SD \pm 654.9)
Gestational age	39,11 (SD \pm 1,14)
Mother's age	28.2 (SD \pm 6.2)
Mother's education	
Incomplete primary education	5 (1.6%)
Complete primary education	17 (5.6%)
Incomplete secondary education	50 (16.4%)
Complete secondary education	154 (50.8%)
Incomplete technical or higher education	28 (9.2%)
Technical or higher (undergraduate or graduate level)	50 (16.4%)
Prenatal	
Yes	288 (94.7%).
No	13 (4.3%).
Missing data	3 (1.0%).
TOTAL LATCH - CEBE	9.30 (SD \pm 1.35; Min=3, Max=10)
L. Lactch Coger	1.78 (SD \pm 0.44; Min=0, Max=2)
A. Audible swallowing Deglución audible	1.84 (SD \pm 0.42; Min=0, Max=2)
T. Type of Nipple Tipo de pezón	1.93 (SD \pm 0.29; Min=0, Max=2)
C. Confort Comodidad, confort (pecho, pezón)	1.95 (SD \pm 0.22; Min=1, Max=2)
H. Hold (positioning) Mantener colocado al pecho	1.80 (SD \pm 0.63; Min=0, Max=2)
Lingual Frenulum (LFPI)	
Normal (equal to or less than 4)	248 (81.6%)
Doubtful (between 5 and 6)	41 (13.5%)
Altered (7 or more)	15 (4.9%)

M=male; F=female. SD=standard deviation. Min=Minimum; Max=Maximum.

Table 2. Description of babies monitored during the first six months of life. Lima, Peru. 2018

Case	Baseline		Month 1			Month 2			Month 3		
	CEBE	Weight	CEBE	Weight	Frenectomy	CEBE	Weight	Frenectomy	CEBE	Weight	Frenectomy
1	Adequate	3300				Adequate	5200	Not	Adequate	6500	No
2	Inadequate	3130	Inadequate	4400	Not	Inadequate	5800	Not			
3	Adequate	3590	Adequate	5800	Not	Adequate	6900	Not			
4	Inadequate	3270	Inadequate	3100	Not	Inadequate	3750	Not	Adequate	5910	Not
5	Adequate	3060							Inadequate	5800	Not
6	Inadequate	3660	Adequate	4600	Not	Adequate	5900	Not	Adequate	7200	Not
7	Inadequate	3880	Adequate	5530	Not				Adequate	8100	Not

Case	Baseline		Month 4			Month 5			Month 6		
	CEBE	Weight	CEBE	Weight	Frenectomy	CEBE	Weight	Frenectomy	CEBE	Weight	Frenectomy
1	Adequate	3300									No
2	Inadequate	3130							Inadequate	Yes	
3	Adequate	3590	Adequate	8700	Not	Adequate	9020	Not	Adequate	Not	
4	Inadequate	3270	Adequate	7540	Not						Not
5	Adequate	3060	Inadequate	6090	Not	Inadequate	9000	Not	Inadequate	Yes	Not
6	Inadequate	3660				Adequate	9000	Not			Not
7	Inadequate	3880				Adequate	8800	Not			Not

Black squares: missing data; CEBE= Clinical Evaluation of Breastfeeding Efficacy.

DISCUSSION

Among the 304 babies, 15 (4.93%) presented ankyloglossia and only 4 (26.7%) of them presented difficulties with breastfeeding in the first hours of life. Seven babies were monitored, and only 2 persisted having difficulties in breastfeeding throughout the months, and were referred to frenotomy; only 1 presented weight below the expected for the age group. Our findings are similar to a study conducted with a population of Brazilian children⁽²⁴⁾.

Even though the prevalence of ankyloglossia is not a consensus in the literature studies that analyze their prevalence are important. This study presents prevalence of 4.93%, a value that is in the range already described by other articles. The prevalence of reported ankyloglossia varies from 3 to 16%. Recent studies in the Brazilian context confirm this prevalence variation ranging from 3.11⁽²⁴⁾ to 19%⁽²⁵⁾. The main reason for this variation is justified by the lack of standardization or clinical criteria accepted to compose the diagnosis. The pediatric dentistry is not trained to evaluate lingual french and alterations in orofacial motricity, because of this unknown subkygnosis. In epidemiological studies investigating problems in the oral mucosa, in general, the prevalence is lower (0.1% to 4.4%) than in other studies that investigate the problem in isolation (4.2% to 10.7%). The prevalence is also higher in studies investigating newborns (1.7% to 10.7%) than in studies investigating children, teenagers, or adults (0.1% to 2.08%)⁽²⁶⁾. Some lighter forms of ankyloglossia can be adapted spontaneously with growth, which would explain this age-related difference. In general, boys seem to be more affected than girls, even though

in some studies a similar proportion or and inverse relationship has been observed⁽²⁷⁾.

Breastfeeding is a highlight in the public policies favoring children globally⁸. Due to the broad incentive of government spheres, campaigns, media, training or professionals, and the recommendation of the World Health Organization, defining that exclusive maternal breastfeeding is recommended until the six months of age and should be offered as a supplement until the two years of age⁽¹⁾, it is suggested that those actions had an impact on the positive result of the efficacy and efficiency in maternal breastfeeding, regardless the presence of altered lingual frenulum. With trained professionals and well-oriented mothers and supported by the family, this practice has been carried out successfully, and such repercussions may be influencing even cases of ankyloglossia, where it is suggested that the mother presented a better management at the time of breastfeeding.

One study analyzing the opinion of experts about the topic found out that approximately 90% of pediatricians and 70% of otorhinolaryngologists believed that ankyloglossia rarely causes feeding difficulties, whereas 69% of breastfeeding consultants believed that it causes difficulties⁽²⁸⁾. In the present study, the majority of mothers whose babies had ankyloglossia had no management difficulties at the time of breastfeeding, demonstrating that this is a practice determined by multiple social, behavioral, cultural and not only anatomical factors.

Randomized clinical trials have shown contradictory results, some demonstrating that there is no objective improvement in breastfeeding but that there is self-efficacy improvement⁽¹⁰⁾ and

others demonstrating that there is immediate improvement in breastfeeding and self-perception of the mother⁽²⁹⁾. Literature on ankyloglossia is outdated⁽¹⁷⁾. However, the natural history and the factors associated to ankyloglossia are not explained in literature^(21,29). One study systematically reviewed the literature about the results in the ankyloglossia treatment and found out that the existing current data are insufficient to evaluate the effect of frenotomy on breastfeeding associated to babies with ankyloglossia⁽²⁹⁾.

The benefits of breastfeeding have already been broadly described in the literature⁽⁷⁻¹⁰⁾. Factors associated to harm in this practice should be thoroughly investigated with high methodological rigor so that appropriate measures are implemented. It is suggested that the surgical procedure may not be always necessary, reinforcing that the orientation about breastfeeding to the mother and the follow-up with the newborn born by the multiprofessional health team may be enough⁽²⁹⁾.

The present study presents a few limitations, one of them is not evaluating the total number of babies born and not doing the follow-up throughout the months to study the natural history of the disease. Still, this is the only study found in the literature that presents ankyloglossia data from Lima, Peru institution of greater medical-surgical complexity for maternal and perinatal care in the country.

CONCLUSION

In conclusion, this study demonstrated the low prevalence of ankyloglossia in babies, as not showing a trend of difficulty or negative interference in maternal breastfeeding. Further longitudinal research is necessary to show the aspects related to ankyloglossia and the recommendations for best treatment.

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Authors' contributions

RSR, BAC and FNH conceived the ideas; RSR, BAC and PB collected the data; RSR, BAC, DDFS, JBH and FNH analysed the data; and RSR, BAC, PB, DDFS, JBH and FNH led the writing and reviewed the final version.