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Position of lips and tongue in rest in newborns with and without ankyloglossia

Posição de lábios e língua no repouso em recém-nascidos com e sem anquiloglossia

Keywords

Lip
Tongue
Lingual Frenulum
Newborn
Ankyloglossia

Descritores

Lábio
Língua
Freio Lingual
Recém-Nascido
Anquiloglossia

ABSTRACT

Purpose: Verify the position of lips and tongue at rest in newborns with and without ankyloglossia. **Methods:** Cross-sectional study, carried out with 130 newborns in University Hospital. Data collection was performed by the researcher and speech-language pathologists from the Hospital. Information on gestational age, sex, weight, height and days of life was collected. The position of the lips and tongue at rest was evaluated through visual inspection with the newborns asleep. After the newborns were awakened, Neonatal Screening of the validated Protocol for the evaluation of the lingual frenulum for infants was performed to detect the alteration of the lingual frenulum. The data obtained were described and submitted to statistical analysis using the Chi-Square test to verify the association between the position of the lips with the tongue and to compare the position of the lips and tongue with and without ankyloglossia. The Mann-Whitney test was used to verify the behavior of the variables that differed between newborns with and without ankyloglossia. The significance level of 5% was adopted. **Results:** When comparing the data, a significant difference was found between: weight and height with and without ankyloglossia; position of lips and tongue. An association between the position of the tongue and lips with and without ankyloglossia was also found. **Conclusion:** Newborns without alteration of the lingual frenulum have a tendency to remain with their lips closed and their tongue elevated during rest and newborns with ankyloglossia have a tendency to keep their lips parted and their tongue low during rest.

RESUMO

Objetivo: Verificar a posição de lábios e língua no repouso em recém-nascidos com e sem anquiloglossia. **Método:** Estudo transversal, realizado com 130 recém-nascidos em um Hospital Universitário. A coleta de dados foi realizada pela pesquisadora e por fonoaudiólogas do Hospital. Foi coletado informações sobre idade gestacional, sexo, peso, altura, dias de vida dos recém-nascidos. Foi avaliada a posição dos lábios e língua no repouso, com os recém-nascidos adormecidos, por inspeção visual. Após os recém-nascidos serem despertados, foi realizada Triagem Neonatal do Protocolo validado de avaliação do frênulo da língua em bebês, para detecção da alteração do frênulo lingual. Os dados obtidos foram descritos e submetidos à análise estatística através do teste Qui-Quadrado, para verificar a associação entre posição dos lábios com a língua e para comparar posição de lábios e língua com e sem anquiloglossia. O teste de Mann-Whitney foi utilizado para verificar o comportamento das variáveis que diferiam entre recém-nascidos com e sem anquiloglossia. Foi adotado o nível de significância de 5%. **Resultados:** Houve diferença significativa entre: peso e altura com e sem anquiloglossia; entre posição de lábios e língua. Encontrou-se também associação entre posição de língua e de lábios com e sem anquiloglossia. **Conclusão:** Os recém-nascidos sem alteração de frênulo lingual têm a tendência de permanecerem com os lábios fechados e a língua elevada durante o repouso, e recém-nascidos com anquiloglossia, têm a tendência de manterem os lábios entreabertos e a língua baixa durante o repouso.

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INTRODUCTION

Early childhood comprises the period from birth to the child's six years of age⁽¹⁾. These years are marked by intense development processes that will be the foundation for the establishment of the future competences and skills of the adult. With regard to the stomatognathic system, there are transient phases of accommodation of the structures of hard tissues (bones) and soft tissues (muscles, tendons, nerves, among others)⁽²⁾ during this period. According to Carruth and Skinner, until six months old, the tongue, the lips and the jaw move together as a unit⁽³⁾, beginning to dissociate their movements only between the sixth and ninth months of age⁽⁴⁻⁶⁾.

The harmonic relationship of the orofacial components provides adequate performance of breathing, sucking, chewing, swallowing and speaking functions, in addition to the correct posture of the jaw, tongue and lips⁽⁷⁾. In the first six months, the predominantly nasal breathing pattern allows the organization of the oral posture with the proper support of the tongue on the palate and passive lip sealing, being essential for the myofunctional growth and balance⁽²⁾.

The position of the tongue and lips at rest has been investigated over the years in individuals of different age groups⁽⁷⁻¹²⁾ with the aim of verifying its impact on the orofacial development. However, only one study was carried out with babies⁽¹³⁾. This study concluded that the position of the tongue in babies without alteration of the lingual frenulum tended to remain elevated at rest, while in babies with ankyloglossia the tongue tended to remain low in the oral cavity during rest⁽¹³⁾.

In the first days of life, the lips and tongue are important for the newborn's vital functions, as they have a direct relation with breastfeeding and breathing. During breastfeeding, the lips participate in the anterior sealing around the nipple-areola complex and the movements of the tongue help in the extraction of the milk⁽¹⁴⁾.

Due to the anatomy of the oral cavity, the baby's breathing is predominantly nasal, with occlusion of the lips and the tongue positioned on the transverse palatine folds⁽¹⁵⁾. The literature posits that the posture of parted lips and tongue position on the floor of the mouth are associated with oral breathing⁽¹⁶⁾.

In this context, the posture of closed lips at rest is extremely important for the orofacial development, as it helps to keep the jaw elevated and the tongue well positioned in the oral cavity, being easily observed in babies, especially during sleep.

Some studies report that ankyloglossia makes it difficult the tongue coupling on the hard palate, as well as favoring the position of half-open lips at rest^(13,17,18), impacting in the maxillary expansion, which can lead to sleep-disordered breathing⁽¹⁹⁻²¹⁾. These changes occur early in life, since orofacial growth is particularly fast in the first two years of life⁽¹⁹⁾. According to Meredith⁽²²⁾, the craniofacial skeleton of a white American corresponds to 60% of the adult head size at birth; at six months old, to 80% of it; and, at three years, to 90%.

It is worth mentioning the lack of studies describing the position of the tongue and lips at rest, both in newborns without alteration of the lingual frenulum and in newborns with ankyloglossia, since the vast majority of the current publication is based on reports of experts and not in observational studies.

Thus, considering the importance of the closed lips and tongue coupling to the hard palate during rest for orofacial growth, as well as the impact of ankyloglossia on tongue movement, it's important to conduct studies to understand the relation between those variables.

Based on what was exposed, the hypothesis of this study was that the ankyloglossia interferes both in the rest position of the lips and in the rest position of the tongue in babies.

Thus, the aim of the present study was to verify the position of lips and tongue at rest in newborns with and without ankyloglossia.

METHODS

This was a cross-sectional study, carried out with 130 newborns from Setor de Alojamento Conjunto (Joint Housing Sector) of a University Hospital, approved by the Research Ethics Committee with human beings at School Hospital of the Federal University of Mato Grosso do Sul (UFMS) under number 1,514,715, and all the responsible people/mothers were informed about the procedures and signed the Free and Informed Consent Form.

Initially, the sample calculation was performed considering a 5% significance level, 90% test power and average effect size based on the hypotheses of association between the variables of the study. The result showed the need for the evaluation of 117 newborns. Considering the possible losses of the sample, the sample calculation of 130 newborns was reached.

The study included newborns from one to five days of age, on exclusive breastfeeding and having an APGAR value greater than or equal to eight at birth in the fifth minute of life. The study excluded indigenous and quilombola populations, preterm newborns, newborns with perinatal complications, presence of craniofacial anomalies, neurological diseases, genetic syndromes visible at the time of assessment and artificial feeding, newborns of mothers with seropositive for human immunodeficiency virus (HIV) and with unstable clinical conditions, according to data from medical records.

Data collection was performed by the researcher and three speech-language pathologists from the University Hospital team, properly trained by the researcher. For this phase, training was carried out with 14 newborns that were included in the final sample. At the end of the training, the researchers achieved a degree of agreement above 90% in the evaluations, considering simple percent agreement calculation.

Information on gestational age, sex, weight and height at birth and days of life of the 130 newborns was collected from the medical records data. The mean and its standard deviation (SD) were used to describe the variables on gestational age, weight, and height at birth and the days of life of the newborn.

Firstly, the position of the lips and tongue at rest was evaluated with the newborns asleep. The position of the lips was verified through visual inspection, and the newborns could have presented closed, parted or open lips, at rest (Figure 1).

The assessment of the tongue position at rest was carried out in three moments, with an interval of five minutes between them, with the newborns still asleep, using the technique described by Martinelli⁽¹³⁾. Thus, the evaluators positioned themselves

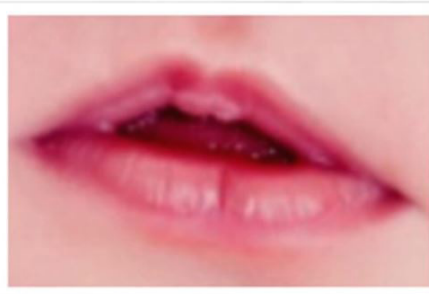
closed lips**half-open lips****open lips**

Figure 1. Position of the lips at rest (the images were authorized and provided by the authors of the screening of the protocol of evaluation of the lingual frenulum in babies⁽²³⁾)

in front of the newborns, opening the mouth of each of them, supporting the gloved thumb in the chin region to lower the mandible; concomitantly, with the index fingers, they raised the upper lip, allowing the visualization of the tongue position at rest, which could be elevated (the anterior third of the tongue in contact with the palatine wrinkles), low or elevated, but without the maintenance of the elevation when the mouth opening maneuver is performed (Figure 2).

To detect the alteration of the lingual frenulum, the Neonatal Screening of the validated protocol for the assessment of the lingual frenulum in babies was applied with the babies awake and close to the time of feeding⁽²³⁾, when the item corresponding to the anatomofunctional finding should be marked. The posture of the lips at rest was observed again, which could be closed, parted or open; the tendency of tongue positioning during crying, which could be in the midline, high line, in the midline with elevation of the sides or with the low point and elevation of the sides was also observed; the shape of the tongue tip when raised during crying or lifting maneuver, which could be rounded, with a slight slit or heart shape was also object of observation. Through the elevation of the lateral margins of the tongue with the gloved right and left index fingers of the evaluator, it was observed the possibility of visualizing the frenulum or not visualizing it; It was possible to visualize it, its thickness (thin or thick) was verified, as well as its fixation on the tongue (in the middle third, between the middle third and the apex or at the apex) and on the floor of the mouth (visible from the sublingual caruncles or from the lower alveolar crest). When the sum of the above described evaluated items was equal to or less than 4, the frenulum was considered normal; between 5 and 6, doubtful frenulum; 7 or more, it was considered altered, with the lingual frenulum limiting the movements of the tongue, characterizing ankyloglossia.

The data obtained were entered into an electronic spreadsheet and submitted to statistical analysis, using the R program (version 3.4.3). The mean was used to describe the variables on gestational age, weight and height at birth and the days of life of the newborn. To check the association between the position of the lips and the position of the tongue at rest, the Chi-Square test was used. The Chi-Square test was also used to compare the position of lips and tongue at rest in babies with and without ankyloglossia. The Mann-Whitney test was used to

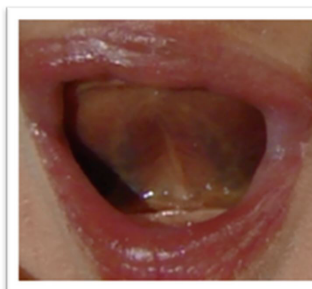
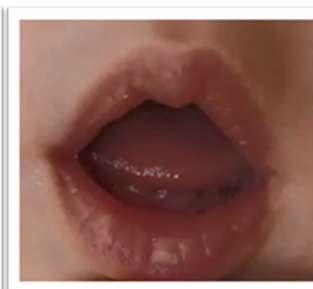
elevated tongue**low tongue**

Figure 2. Position of the tongue at rest (These images do not belong to the protocol – they belong to the authors of this article)

verify whether the behavior of the variables differed between newborns with and without ankyloglossia. The significance level of 5% was adopted for all tests.

RESULTS

Of the 130 newborns evaluated, 69 (53%) were female and 61 (47%) were male. Of these, 105 (81%) had a normal lingual frenulum and 25 (19%) were diagnosed with ankyloglossia. Of the newborns with normal frenulum, 59 (56%) were female, and 46 (44%) were male; of newborns with ankyloglossia, 15 (60%) were male and 10 (40%) female. The statistical analysis showed that there was no statistically significant difference between the sexes ($p = 0.122$).

The average gestational age of the 130 newborns was 39.22 weeks ($SD=1,31$). The average gestational age of newborns with normal lingual frenulum was 39.21 weeks and of those with ankyloglossia was 39.28 weeks. There was no statistically significant difference between them ($p = 0.580$).

The average weight found for the 130 newborns was 3.29 kg ($SD= 0,43$); the average weight of newborns with normal lingual frenulum was 3.10 kg and of those with ankyloglossia was 3.26 kg. There was a statistically significant difference between newborns with and without ankyloglossia ($p <0.05$).

The average height at birth of the 130 newborns was 49.22 cm ($SD=2,06$); the average height of newborns with a normal lingual frenulum was 49.15 cm and, of those with ankyloglossia, it

Table 1. Association between position of lips and tongue at rest of the 130 babies assessed

Variables		Closed Lips		Open/Half-open Lips		Value de p
Tongue Position	Elevated	89	92.7%	7	7.3%	<0.001*
	Low	23	67.6%	11	32.4%	

Chi-square test; *Statistical significance

Table 2. Position of the tongue and lips at rest in babies with and without ankyloglossia

Variable	Normal frenulum		Ankyloglossia		Value of p
	Frequency	Percentage	Frequency	Percentage	
Elevated tongue at rest	86	82.0%	10	40%	< 0.001*
Low tongue at rest	19	18.0%	15	60%	
Closed lips	97	93.0%	15	60%	< 0.001*
Half-open or open lips	8	7.0%	10	40%	

Chi-square Test; *Statistical significance

was 49.55 cm. There was a statistically significant difference ($p < 0.05$) when the height at birth of newborns with and without ankyloglossia was compared.

The average days of life of the 130 newborns was 1.68 days ($SD=1,02$). In newborns with normal lingual frenulum, the average was 1.76 days and, in those with ankyloglossia, it was 1.36 days. There was no statistically significant difference when comparing newborns with and without ankyloglossia ($p = 0.156$).

The results showed a significant association between position of lips and tongue at rest ($p < 0.001$), showing that newborns who presented position of closed lips tended to maintain the position of the tongue elevated at rest, while babies who presented position of parted lips tended to have a low tongue position in the oral cavity (Table 1).

A significant association was also found between the rest position of tongue and lips with ankyloglossia ($p < 0.001$), showing that the babies with this congenital malformation tend to have low tongue position and parted lips at rest; and the babies with normal lingual frenulum tend to stay with the tongues elevated and lips closed at rest (Table 2).

DISCUSSION

This study was conducted with the purpose of verifying the position of lips and tongue at rest in newborns with and without ankyloglossia.

Although this study is not intended to determine prevalence, 19% of the sample was diagnosed with ankyloglossia, being within the range found in the literature, which indicates a variation in the prevalence of ankyloglossia between 4.8%⁽²⁴⁾ and 37%⁽²⁵⁾. This variation can be related to the different definitions and methodologies used to carry out the studies. A Brazilian study, using the same protocol for assessing the lingual frenulum, found a prevalence close to the one found in this study (21%)⁽²³⁾.

When comparing data on sex, gestational age and days of life of newborns with normal frenulum and ankyloglossia, no statistically significant differences were found, showing that the samples were homogeneous in relation to these variables.

Regarding weight and height, statistically significant differences between newborns with normal lingual frenulum and those with ankyloglossia were found. No studies were found that considered these variables for comparison. However, a possible explanation for this finding may be the fact that the majority of newborns

diagnosed with ankyloglossia were male. Siqueira et al.⁽²⁶⁾ reported that the measurements of weight and height in full-term male newborns are greater than those of female ones.

In this study, an association between the position of lips and tongue at rest of the 130 evaluated babies was found, showing that newborns who presented position of closed lips maintained the position of the tongue elevated at rest, while babies who presented position of parted lips had a low rest tongue position. A possible explanation for these findings may be the fact that, until six months old, the tongue, lips and jaw move as a unit⁽²²⁾, not dissociating their movements. Therefore, if the tongue remains low in the oral cavity, the jaw lowers, causing the lips to part. Santos-Neto et al.⁽²⁷⁾ point out that the position of parted lips and the tongue on the floor of the mouth at rest are associated with oral breathing.

Thus, the hypothesis that ankyloglossia interferes with the position of lips and tongue at rest was confirmed in the present study, corroborating the findings of Martinelli et al.^(13,18), who showed, in two different studies, the position of the lips and the position of the tongue at rest in babies with and without ankyloglossia, respectively, but did not correlate both positions with each other. Huang et al.⁽¹⁹⁾, also reported that the low tongue position caused by ankyloglossia interferes with orofacial growth, particularly affecting the development of the maxilla, leading to abnormal development of the hard palate (high and narrow) and, secondarily, to oral breathing during sleep. Haraké et al.⁽²⁸⁾ refer that the resting position of the tongue, coupled to the hard palate, is a fundamental requirement for correct speech and swallowing. The findings of this study showed that ankyloglossia prevents the tongue from coupling to the palate and favors that the lips remain slightly parted at rest, which may interfere with the proper performance of orofacial functions.

The results showed that the diagnosis and treatment of ankyloglossia must be performed early so as not to interfere with the position of the tongue and lips at rest, and, consequently, in the orofacial growth, since the anatomical characteristics of the lingual frenulum do not change over time⁽²⁹⁾ and its histological constitution does not allow it to be stretched through exercises⁽³⁰⁾.

This study made it possible to associate the position of the tongue and lips in newborns with or without ankyloglossia. However, the limitations of the present research must be considered given the reduced number of the sample, which allows considering the results found only for the population concerned.

Future studies, with the objective of verifying whether the position of the tongue and lips at rest, in newborns diagnosed with ankyloglossia, changes after the performance of lingual frenectomy, can contribute to prove the importance of both diagnosis and early intervention in of the treatment of this congenital malformation.

CONCLUSION

In the first days of life, clinically stable newborns without alteration of the lingual frenulum tend to keep the lips closed and elevated tongue during rest. And newborns with ankyloglossia tend to keep their lips parted and low tongue in the oral cavity during rest.

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Author contributions

SMAC idealization of the study, collection, analysis and interpretation of data and writing of the article; RLCM and DBP as supervisor, the idealization of the study, analysis, data interpretation and writing of the article.