INTRODUCTION

A small fold of mucous membrane, called lingual frenulum, connects the underside of the tongue to the floor of the mouth. The lingual frenulum effects the movement of the tongue. When the lingual frenulum cells don’t undergo apoptosis completely during the embryologic development, the residual tissue may restrain the movements of the tongue1.

The absence of agreement on the criteria used for evaluation and anatomical classification of the lingual frenulum may be the cause of the variation in the reported incidence rates of ankyloglossia which is between 0.88% and 12.8%2-5.

Only one protocol was identified to evaluate the frenulum in infants up to six months of age. This protocol was proposed in 1993. It includes several items regarding the appearance of the frenulum, and proposes the evaluation of the movements of the tongue through the stimulation of reflex, and non-nutritive sucking6. However, authors reported limitations of the protocol in the identification of children with tongue-tie, who also present breastfeeding difficulties.

In 2012, Martinelli et al8 proposed an experimental protocol from the literature review considering the anatomic variations of lingual frenulum and the functions of sucking and swallowing. This protocol represented a great advance for diagnosing
the interference of the lingual frenulum with the movements of the tongue during breastfeeding. However, the administration of the protocol was long and complex. From the experimental protocol model, a new protocol was designed.

Breastfeeding is directly related to the functions of sucking and swallowing, which depend on the movements of the tongue. Any alteration can result in function impairment and breastfeeding difficulties.

Considering those facts, a new lingual frenulum protocol for infants was designed. The protocol correlates anatomic and functional aspects. That may contribute to an accurate diagnosis and treatment avoiding early weaning and/or poor weight gain. The aim of this study is to identify the anatomical characteristics of the lingual frenulum interfering with the functions of sucking and swallowing in full-term infants in order to re-design the experimental protocol proposed by Martinelli et al.

**METHOD**

This was a cross-sectional study with a sample of a 100 subjects. The re-designed protocol was administered to 44 female and 56 male. All subjects were born at the same hospital in the countryside area of Sao Paulo between September/2011 and April/2012. Full-term healthy infants, independently from type of parturition, who were being breastfed, were included in the research. Prematurity, perinatal complications, craniofacial anomalies, neurological diseases, genetic syndromes visible at the moment of assessment and artificial feeding were the exclusion criteria.

The evaluation was performed when the infants were 30 days old, considering that at this time they were adapted to breastfeeding.

The experimental lingual frenulum protocol for infants proposed by Martinelli et al was administered to the infants by one SLPs at breastfeeding time. All steps of the protocol were recorded and photographed. The information collected was sent to two expert SLPs, who evaluated and discussed the cases based on the recordings and photographs.

The data collected by the specialists were subjected to statistical analysis using the Pearson’s chi-squared test, followed by Fisher’s exact test for qualitative variables. The quantitative data were submitted to analysis of variance (ANOVA) followed by Tukey’s test. For statistical analysis the significance level was 5% (p< 0.005).

This study was approved by the Ethics Committee of the Faculty of Odontology of Bauru, University of Sao Paulo under number 113/2011.

**RESULTS**

From the 100 infants who were evaluated, 29 had posterior frenulum; therefore, lingual frenulum was not visible. The protocol was fully administered to 71 infants. 55 infants were normal and 16 infants (22.54%) had anatomical characteristics that restrained the movements of the tongue during nutritive sucking. From the 16 subjects, 3 were female and 13 were male. Statistical analysis demonstrated that there is a relationship between the anatomical characteristics of the lingual frenulum and gender as well as between the anatomical characteristics of the lingual frenulum and the nutritive and non-nutritive sucking (Tables 1 and 2).

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**Table 1 – Results from Person’s chi-squared test in relation to clinical history data, anatomical and functional evaluations**

<table>
<thead>
<tr>
<th>Comparison</th>
<th>P-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>gender x frenulum attachment to the floor of the mouth</td>
<td>0.005</td>
</tr>
<tr>
<td>Tongue posture during crying x time between feeding</td>
<td>0.010</td>
</tr>
<tr>
<td>Tongue shape when elevated x fatigue during feeding</td>
<td>0.002</td>
</tr>
<tr>
<td>Frenulum attachment to the tongue x tongue movement during nutritive sucking</td>
<td>0.044</td>
</tr>
</tbody>
</table>
It was possible to identify that the following items of the protocol contribute to detection of interference of lingual frenulum with the movements of the tongue: tongue posture during crying, tongue shape when elevated, frenulum attachment to the tongue and to the floor of the mouth, time between feedings, fatigue during feeding, tongue movement during non-nutritive sucking and time of pause between groups of suction.

Based on the clinical experience by administering the experimental protocol to 100 infants, mothers’ complaints and the statistical analysis results, the protocol was re-designed.

In history, new items were included: “sucks a little and sleeps”, “slips off nipple” and “chews nipple”.

In the anatomo-functional evaluation, the item “midline with lateral elevation” was included in “tongue posture during crying”. In item 4, a maneuver for lingual frenulum visualization with directions was included.

Scores were assigned to the 16 infants with lingual frenulum alteration. Results are shown in Table 3.

Scores indicating alterations were included based on the results of the re-designed lingual frenulum protocol with scores administered to 16 infants. Partial and total scores were defined.

In history, when the sum of items is equal or more than 4, the interference of frenulum with tongue movements may be considered. This reference was obtained from the items “time between feedings – 1 hour or less” (2) – and “chews nipple” (2).

In the anatomo-functional evaluation, when the sum of items 1, 2 and 3 is equal or more than 4, the interference of frenulum with tongue movements may be considered. This reference was obtained from item 2 (tongue posture during crying): down or midline with lateral elevation (2) and from item 3 (shape of tongue apex when elevated during crying): v-shaped (2). When the sum of item 4 is equal or more than 3, the interference of frenulum with tongue movements may be considered. This reference was obtained from item 4.1 (frenulum thickness): thick (2) and from item 4.3 (frenulum attachment to the floor of the mouth): visible from the crest (1).

In part II, when the sum of non-nutritive sucking and nutritive sucking is equal or more than 2, the interference of frenulum with tongue movements may be considered. This reference was obtained from item 1.1 (tongue movement coordination): inadequate (1) and from item 2.1 (suction rhythm): a few suctions followed by long pauses (1).

When the sum of clinical examination is equal or more than 9, the interference of frenulum with tongue movements may be considered. This reference was obtained from part I, item 2 (tongue posture during crying): down (2) or midline (2); from item 3 (shape of tongue apex when elevated during crying): V-shaped (2); from item 4.1 (frenulum thickness): thick (2); from item 4.3 (frenulum attachment to the floor of the mouth): visible from the crest (1); from part II, items 1.1 (tongue movement): inadequate (1) and 2.1 (suction rhythm): a few suctions followed by long pauses (1).

When the sum of history and clinical examination is equal or more than 13, the interference of frenulum with tongue movements may be considered. This reference was obtained from the following items: in history, affirmative answers for chews nipple (2) and 1 hour or less for time between feedings(2); in the anatomo-functional evaluation, part I, item 2 (tongue posture during crying): down (2) or midline

<table>
<thead>
<tr>
<th>Sucking</th>
<th>Shape of the apex of the tongue during crying</th>
<th>Average</th>
<th>Standard variation</th>
<th>ANOVA p-value</th>
<th>Comparison</th>
<th>Tukey p-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Rhythm ( # of suctions per group)</td>
<td>Round</td>
<td>31,88</td>
<td>20,80</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td></td>
<td>V-shaped</td>
<td>26,45</td>
<td>12,96</td>
<td>0,131</td>
<td>round ≠ v-shaped</td>
<td>*0,023</td>
</tr>
<tr>
<td></td>
<td>Heart-shaped</td>
<td>17,73</td>
<td>10,00</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Pause time</td>
<td>Round</td>
<td>4,78</td>
<td>1,60</td>
<td>-</td>
<td>round ≠ v-shaped</td>
<td>-</td>
</tr>
<tr>
<td></td>
<td>V-shaped</td>
<td>5,96</td>
<td>2,76</td>
<td>0,013</td>
<td>v-shaped ≠ heart-shaped</td>
<td>0,169</td>
</tr>
<tr>
<td></td>
<td>Heart-shaped</td>
<td>6,54</td>
<td>1,56</td>
<td>-</td>
<td>v-shaped ≠ heart-shaped</td>
<td>0,832</td>
</tr>
</tbody>
</table>
with lateral elevation (2); item 3 (shape of tongue apex when elevated during crying): V-shaped (2); item 4.1 (frenulum thickness: thick (2) and item 4.2 (frenulum attachment to the tongue): visible from the crest (1); in part II, item 1.1 (tongue movement): inadequate (1) and item 2.1 (suction rhythm: a few suctions followed by long pauses (1).

Scores 0-8 were considered normal. 25 was considered the worst result, being 8 the worst result for history, 12 for the anatomo-functional evaluation, and 5 for evaluation of non-nutritive and nutritive sucking during breastfeeding.

From the administration of the protocol to 100 infants and the results from the statistical analysis, the lingual frenulum protocol for infants with scores was re-designed (Figures 1, 2, 3 and 4).

Table 3 – Results from the administration of the re-designed protocol with scores to 16 infants with lingual frenulum alteration

<table>
<thead>
<tr>
<th>Infant</th>
<th>Gender</th>
<th>History scores</th>
<th>Anatomo-functional evaluation scores</th>
<th>Non-nutritive and nutritive sucking evaluation scores</th>
<th>Total scores</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td></td>
<td>Items 1 to 3</td>
<td>Item 4</td>
<td></td>
</tr>
<tr>
<td>1</td>
<td>M</td>
<td>5</td>
<td>5</td>
<td>3</td>
<td>5</td>
</tr>
<tr>
<td>2</td>
<td>M</td>
<td>7</td>
<td>6</td>
<td>4</td>
<td>5</td>
</tr>
<tr>
<td>3</td>
<td>M</td>
<td>4</td>
<td>4</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>4</td>
<td>F</td>
<td>5</td>
<td>4</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>5</td>
<td>M</td>
<td>4</td>
<td>4</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>6</td>
<td>M</td>
<td>7</td>
<td>6</td>
<td>3</td>
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<tr>
<td>7</td>
<td>M</td>
<td>6</td>
<td>5</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>8</td>
<td>M</td>
<td>5</td>
<td>5</td>
<td>3</td>
<td>2</td>
</tr>
<tr>
<td>9</td>
<td>M</td>
<td>6</td>
<td>5</td>
<td>3</td>
<td>3</td>
</tr>
<tr>
<td>10</td>
<td>M</td>
<td>4</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>11</td>
<td>M</td>
<td>7</td>
<td>5</td>
<td>4</td>
<td>5</td>
</tr>
<tr>
<td>12</td>
<td>F</td>
<td>6</td>
<td>6</td>
<td>4</td>
<td>5</td>
</tr>
<tr>
<td>13</td>
<td>M</td>
<td>7</td>
<td>3</td>
<td>3</td>
<td>5</td>
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<tr>
<td>14</td>
<td>F</td>
<td>5</td>
<td>5</td>
<td>3</td>
<td>5</td>
</tr>
<tr>
<td>15</td>
<td>M</td>
<td>6</td>
<td>5</td>
<td>3</td>
<td>3</td>
</tr>
<tr>
<td>16</td>
<td>M</td>
<td>6</td>
<td>6</td>
<td>4</td>
<td>5</td>
</tr>
</tbody>
</table>

| Average | 5,62 | 4,75 | 3,25 | 4,18 | 17,81 |
| Standard deviation | 1,08 | 1,12 | 0,44 | 0,91 | 2,63 |
Figure 1 – Frenulum Protocol for infants – Clinical History
Figure 2 – Frenulum Protocol for infants – Clinical Exam Part 1
LINGUAL FRENULUM PROTOCOL WITH SCORES FOR INFANTS
Martinelli, 2013

4. Lingual Frenulum

( ) visible
( ) not visible
( ) visible with maneuver*

If the frenulum is not visible, go to PART II (Non-nutritive sucking and nutritive sucking evaluations)

4.1. Frenulum thickness

( ) thin (0)
( ) thick (2)

4.2. Frenulum attachment to the tongue

( ) midline (0)
( ) between midline and apex (2)
( ) apex (3)

4.3. Frenulum attachment to the floor of the mouth

( ) visible from the caruncles (0)
( ) visible from the crest (1)

*Maneuver: elevate and push back the tongue.
If the frenulum is not visible, the infant must be seen by a SLPs each two months for periodic frenulum evaluation.

Anatomo-functional evaluation total score (item 4): Best result=0 Worst result=6
When the score of item 4 of the anatomo-functional evaluation is equal or greater than 3, the interference of the frenulum with the movements of the tongue may be considered.

Anatomo-functional evaluation total score (items 1, 2, 3 and 4): Best result=0 Worst result=12
When the score of items 1, 2, 3 and 4 of the anatomo-functional evaluation is equal or greater than 7, the interference of the frenulum with the movements of the tongue may be considered.

Figure 3 – Frenulum Protocollo for infants – Clinical Exam Part 2
LINGUAL FRENULUM PROTOCOL WITH SCORES FOR INFANTS
Martinelli, 2013

PART II – EVALUATION OF NON-NUTRITIVE SUCKING AND NUTRITIVE SUCKING

1. Non-nutritive suction (little finger suction wearing gloss)
   1.1. Tongue movement coordination
   ( ) adequate: coordinated movement
   ( ) inadequate: restricted tongue protrusion, uncoordinated movements and late suction start

2. Nutritive sucking during breastfeeding
   (when breastfeeding starts, observe infant sucking during five minutes)
   2.1. Suction Rhythm (observe groups of suction and pauses)
   ( ) several suction in a row followed by short pauses (0)
   ( ) a few suction followed by long pauses (1)
   2.2. Coordination among suction/swallowing/breathing
   ( ) adequate (0) (balance between feeding efficiency and sucking, swallowing and breathing functions without stress)
   ( ) inadequate (1) (cough, choking, dyspnea, regurgitation, hiccup, swallowing noises)

2.3. Nipple chewing
   ( ) no (0)
   ( ) yes (1)

2.4. Clicking during sucking
   ( ) no (0)
   ( ) Yes (1)

Non-nutritive sucking and nutritive sucking total score: Best result= 0 worst= 5
When the score of non-nutritive sucking and nutritive sucking is equal or greater than 2, the interference of the frenulum with the movements of the tongue may be considered.

When the sum of clinical examination is equal or more than 9, the interference of the frenulum with the movements of the tongue may be considered.

HISTORY AND CLINICAL EXAMINATION TOTAL SCORES: Best result= 0 Worst result= 25
When the sum of history and clinical examination is equal or more than 13, the interference of the frenulum with the movements of the tongue may be considered.
DISCUSSION

This study found 22.54% of frenulum alterations in the subjects assessed. Results from previous studies ranged from 4.8% to 12.8%. The higher incidence may be due to the fact that different methodologies were used in previous studies. In this study anatomo-functional aspects and the evaluation of non-nutritive and nutritive sucking during breastfeeding were considered.

From the 16 infants who had frenulum alteration, 13 were male. This finding agrees with previous studies that report the prevalence of lingual frenulum alteration in male infants.

From the 100 infants who were evaluated, 29 had posterior frenulum; therefore, lingual frenulum was not visible. Posterior frenulum is not very addressed in the literature. Surgery indication for posterior frenulum is not recommended. Periodic appointments with an SLPs for evaluation may be the indication for infants with posterior frenulum.

A maneuver consisting of elevating and pushing back the tongue was developed to visualize posterior frenulum. The maneuver was included in the re-designed protocol.

The statistical analysis demonstrated relation between: tongue posture during crying and time between feedings; tongue shape when elevated and fatigue during feeding; suction rhythm/pause time and tongue shape during crying, and lingual frenulum attachment and the tongue movement during non-nutritive sucking. These findings indicate that lingual frenulum alteration restrains the tongue movements during crying and breastfeeding. Although the literature agrees that the infants' feeding cycle is compromised when the lingual frenulum is altered, studies correlating the variables cited were not found.

Data analysis also demonstrated that the worse the shape of the apex of the tongue when elevated during crying, the fewer number of suctions. In infants with V-shaped tongue during crying, pause time was longer. Studies comparing these variables were not found in the literature. These findings may be due to the fact that because the movements of the tongue are restrained, the infants make more effort during breastfeeding, causing muscle fatigue. That may justify the longer pauses for resting.

The re-designed protocol is considered to be an effective tool for diagnosing possible infants' lingual frenulum alteration interfering with breastfeeding. Its administration is non-invasive and presents low risks for infants. The re-designed protocol differs from the protocol published by Hazelbaker, being visual inspection and evaluation of tongue movements and non-nutritive sucking the different items.

In the re-designed protocol scores indicate alterations. In the literature scores are proposed to help professionals to assess the level of alterations as well as to establish assessment patterns for comparing data from different research centers. When the full administration of the protocol is not possible, administering only the anatomo-functional part helps the professional to refer to surgery, due to the partial scores.

The consequences of frenulum alteration and surgery indications are subject of divergence in the literature. Frenotomy is also the subject of divergence, since there are frequent questions about whether to perform surgery or not, when to perform surgery, and who would be the most qualified professional to perform it. An effective protocol may contribute to diagnose possible lingual frenulum alterations and its consequences, and to refer to surgery when necessary. Specific protocols allow planned therapeutic actions, clinical procedure documentation, and evidence-based clinical practice.

Analyzing the relation between the anatomic and functional aspects was the first step to validate the protocol. Validated protocols for infants were not found in the literature. Therefore, the comparison to a gold standard protocol will not be possible. Administration the protocol by other qualified professionals and re-evaluation tests to verify interjudge and intrajudge agreement will be the future steps required for the validation of the protocol.

CONCLUSIONS

Based on the results it was possible to identify that the shape of the tongue during crying and the lingual frenulum attachment are the anatomical characteristics of lingual frenulum that interfere with the tongue movement during the non-nutritive sucking and the suction rhythm during breastfeeding.

From the administration of the experimental protocol it was possible to re-design the lingual frenulum protocol for infants with scores. The new protocol is considered to be an effective tool for assessing and diagnosing the anatomical alterations of the lingual frenulum that may interfere with sucking and swallowing during breastfeeding.
RESUMO

Objetivo: verificar quais características do frênulo da língua influenciam nas funções de succção e deglutição em bebês nascidos a termo, com a finalidade de propor adequações no protocolo proposto por Martinelli et al (2012). Método: foi aplicado o protocolo de avaliação do frênulo da língua em 100 bebês saudáveis nascidos a termo. Os filmes obtidos na avaliação foram analisados por duas fonoaudiólogas especialistas em MO, com experiência em avaliação de frênulo lingual. Foram aplicados os testes Qui-quadrado seguido do teste exato de Fisher, além da análise de variância, considerando os dados qualitativos e quantitativos, respectivamente. Resultados: nos 16 bebês que apresentaram alteração do frênulo lingual, verificou-se a relação entre: a tendência do posicionamento da língua durante o choro e o tempo entre as mamadas; a forma da língua quando elevada e o cansaço para mamar; bem como a fixação do frênulo na língua e o movimento da língua na succção não nutritiva. Pela análise dos dados dos bebês com alteração de frênulo lingual, foi possível definir as características indicativas de alteração, que possibilitaram a adequação do protocolo inicial, e a atribuição de escores. Conclusão: a forma da língua, quando elevada durante o choro, influencia o movimento da língua durante a succção não nutritiva, e o ponto de fixação do frênulo na língua influencia o ritmo da succção durante a amamentação. O novo protocolo com escores é uma ferramenta efetiva para avaliar e diagnosticar alterações anatômicas do frênulo da língua e suas possíveis interferências na amamentação.

DESCRITORES: Freio Lingual; Protocolos Clínicos; Aleitamento Materno; Comportamento de Succção; Deglutição

REFERENCES