

## Case reports

# Clinical and electromyographic evaluation pre and post lingual frenotomy of an infant with ankyloglossia: A case report

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## ABSTRACT

The goal is to present a clinical case of a baby diagnosed with ankyloglossia evaluated clinically and by surface electromyography, during bottle sucking before and after 15, 30 and 60 days of lingual frenotomy. Based on the anatomofunctional evaluation of the Tongue Frenulum Assessment Protocol in Babies, interference of the lingual frenulum was identified. The tongue, during sleep, was positioned on the floor of the mouth. The initial electromyographic evaluation of the suprahyoid muscles, during bottle feeding, showed asymmetry in root mean square (RMS) values, in microvolts ( $\mu V$ ), between the right and left suprahyoid muscles. The evaluation was repeated after 15, 30 and 60 days after lingual frenotomy. After 15 days, appropriate lip and tongue posture, coordinated sucking movements, and absence of noises or choking were observed. After 30 days, symmetry was observed between the suprahyoid muscles, which was maintained 60 days after the surgical procedure. After lingual frenotomy, the adequacy of the position of lips and tongue, during rest and sleep, was reestablished within 15 days, and after 30 days of the procedure, there was adequacy of the tongue posture, during crying, and symmetry of the electromyographic activity of the suprahyoid muscles, during sucking. The results were maintained until the end of the study.

**Keywords:** Ankyloglossia; Lingual Frenulum; Suction; Electromyography

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## INTRODUCTION

The infant lingual frenulum is a dynamic structure formed by a fold in the fascia of the floor of the mouth<sup>1</sup>. Recent studies report that the anatomical variations of the tongue frenulum occur because a portion of tissue that did not undergo apoptosis, during embryonic development, remains on the sublingual surface of the tongue. This condition is called ankyloglossia<sup>2-5</sup>.

Ankyloglossia is a congenital anomaly that can restrict tongue movements<sup>2,3</sup>. Ankyloglossia changes the usual position of the tongue, as it makes it difficult to attach it to the hard palate, impacting jaw development, which can lead to breathing disorders during sleep<sup>4</sup>. The lips are also changed<sup>4</sup>, with a tendency to lack of sealing. These factors negatively impact the correct craniofacial growth and development as well as chewing, speaking and sucking functions<sup>4,6,7</sup>.

Due to difficulty in sucking, ankyloglossia may be responsible for discontinuing exclusive breastfeeding, as it makes milk extraction difficult, interferes with weight gain, and generates complaints from the mother, such as nipple fissure, pain during breastfeeding, mastitis and signs of colic and reflux in children<sup>5,7-9</sup>.

When correctly diagnosed, ankyloglossia can be treated through lingual frenotomy<sup>6,8,10,11</sup>. However, the findings after frenotomy are still controversial. One study reported that frenotomy reduced mothers' breast pain and symptoms of gastroesophageal reflux<sup>7</sup>. Another study, using surface electromyography, identified differences in the electromyographic activity of suprahyoid muscles in babies with ankyloglossia, characterized by different locations of attachment of the frenulum on both the tongue and the floor of the mouth, but no research was found using the surface electromyography (EMGs) after frenotomy<sup>12</sup>.

Therefore, as findings after lingual frenotomy are still scarce, this study aimed at presenting a clinical case of a baby diagnosed with ankyloglossia who was evaluated clinically and by surface electromyography, during bottle sucking before and after 15, 30 and 60 days of lingual frenotomy.

## CASE REPORT

The study was approved by the Research Ethics Committee of the Hermínio Ometto Foundation, under No. 5.460.945 and CAEE No. 59292122.8.0000.53850 Foundation, Brazil. Those responsible for the baby were duly informed about the procedures carried out in the study and signed the Informed Consent Form.

This is a clinical case of a male baby, full-term pregnancy, natural birth and without perinatal complications, clinically evaluated at three months of age using the Tongue Frenulum Assessment Protocol in Babies<sup>13</sup>, and through EMGs of the right and left suprahyoid muscles during three minutes of bottle sucking<sup>12</sup>.

According to the mother's report, initially there was difficulty latching on. The baby released the nipple easily, made noises during sucking and showed pauses and signs of tiredness. The feeding time was, on average, 18 minutes. Thus, she started using a bottle, but the difficulty in feeding the baby persisted. Uncoordinated movements were identified with leakage of milk, choking, long pauses, regurgitation and noise during swallowing.

The structures of the stomatognathic system and the suction function were clinically evaluated by a speech therapist specialized in Orofacial Motricity through the application of the Tongue Frenulum Assessment Protocol in Babies<sup>13</sup>, validated for application in Brazil<sup>14</sup>.

## Application of the Tongue Frenulum Assessment Protocol in Babies

The anatomofunctional assessment - items 1, 2 and 3 - identified posture of lips parted at rest (score 2), tendency for the tongue to be lowered during crying (score 2) and shape of the tongue during crying with a slight crack at the apex (score two). The sum of items 1, 2 and 3 was 6, identified as the worst result.

For item 4, a frenulum of possible visualization was observed, thin in thickness (score 0), fixation on the sublingual surface between the middle third and the apex (score 2) and, on the floor of the mouth, visible from the lingual caruncles (score 0).

The sum of items 1, 2, 3 and 4 totaled 8. Interference of the lingual frenulum is considered in scores equal to or greater than 7<sup>14</sup>.

After the diagnosis of ankyloglossia, frenotomy was performed by a dental surgeon, specialist in oral and maxillofacial surgery, using topical anesthetic, scissors and a tentacannula.

## Electromyographic Assessment

After the clinical evaluation, the surface electromyographic evaluation was performed.

The electromyographic activity was recorded using New Miotool 400® (Miotec) equipment and Miograph software, with a sampling frequency of 2,000 Hz, a low-pass filter of 20 Hz and a high-pass filter of 500

Hz, neonatal-sized disposable electrodes Maxicor® (Pinhais, PR, Brazil), foamed with solid gel, hypoallergenic adhesive, stainless steel button and AgCl cotter pin. Two electrodes were used, with a distance of 20 mm between them, for each collection point.

The skin in the region of the suprahyoid muscles and the styloid process of the ulna was cleaned with gauze and 70% alcohol.

The electrodes were attached to the region of the suprahyoid muscles from the midline below the chin in a postero-anterior direction on the right and left sides. The reference electrode was positioned on the styloid process of the ulna on the right arm. Baby and mother were positioned comfortably, so that the electrodes had little movement interference.

The electromyographic signal was collected for three minutes in the right suprahyoid (SHD) and left suprahyoid (SHE) muscles during breastfeeding using a MAM® bottle and 1 MAM® teat (MAM Babyartikel GesmbH, Vienna, Austria) Slow Flow. The mother was instructed to schedule the feeding so that the baby would not be fed for at least an hour.

For data analysis, a total period of three minutes collected during bottle sucking was used. Root mean

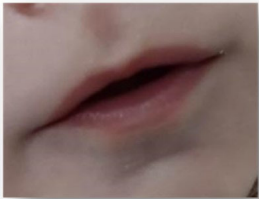
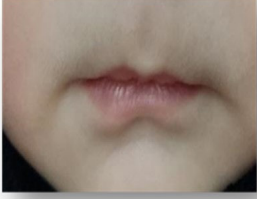
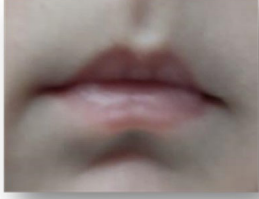
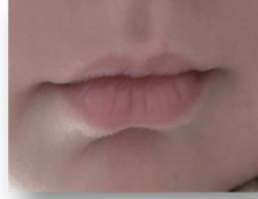
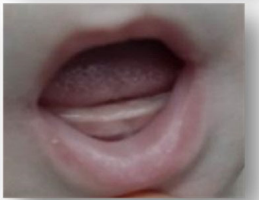
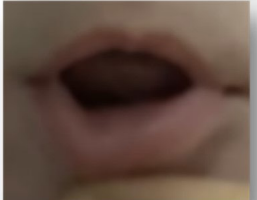




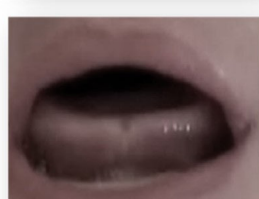
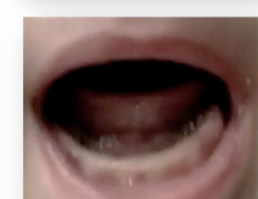
square (RMS) values were calculated. The RMS is a fundamental measure of the magnitude of an electromyographic signal in the time domain (amplitude), expressed in microvolts ( $\mu\text{V}$ )<sup>9</sup>, and is considered the “gold standard”<sup>15</sup>. The data were not normalized in order not to mask possible asymmetries.

The application of the tongue frenulum assessment protocol in babies and the EMG recordings were performed before and after 15, 30 and 60 days of frenotomy.

## RESULTS

Fifteen days after frenotomy, the lips were occluded and the tongue was elevated during sleep. The baby continued to have a tendency to position the tongue in the midline with elevation of the sides during crying. There was an increase in the interval between bottle feedings to more than two hours. The noises during breastfeeding decreased and the choking stopped.

In the reassessment after 30 and 60 days after frenotomy, the baby maintained the posture of occluded lips at rest and the tongue elevated during sleep. During crying, the tongue became positioned in the midline (Figure 1).

<i>Clinical evaluation</i>				
	Before frenotomy	After 15 days of frenotomy	After 30 days of frenotomy	After 60 days of frenotomy
Lip posture at rest				
Tongue position while sleeping				
Tongue position while crying				

**Figure 1.** Images collected during clinical evaluation before, after 15, 30 and 60 days of frenotomy

After lingual frenotomy, an adequate sucking rhythm and coordination between sucking/swallowing/breathing were observed during bottle feeding, with no milk leakage, choking and long pauses.

The initial EMG evaluation of the right and left suprahyoid muscles during three minutes of bottle feeding showed asymmetry between the muscles evaluated. The RMS value was 25.32 microvolts ( $\mu V$ ) for the right suprahyoid muscles, and 49.44  $\mu V$  for the left suprahyoid muscles.

Fifteen days after lingual frenotomy, a decrease in asymmetry was observed. The left suprahyoid muscles showed a value of 39.18  $\mu V$  and the right suprahyoid muscles 24.15  $\mu V$ . Thirty days after lingual frenotomy, asymmetry was observed between the right suprahyoid (32.72  $\mu V$ ) and left suprahyoid (34.72  $\mu V$ ) muscles. The symmetry was maintained in the evaluation after 60 days, with the value for the left suprahyoid muscles being 44.11  $\mu V$  and the right suprahyoid muscles being 40.02  $\mu V$  (Chart 1).

**Chart 1.** Values in RMS (Root Means Square) obtained through electromyographic evaluation before frenotomy and after 15, 30 and 60 days

Muscles	Before	After 15	After 30	After 60
Right suprahyoid	25.32 $\mu V$	24.15 $\mu V$	32.72 $\mu V$	40.02 $\mu V$
Left suprahyoid	49.44 $\mu V$	39.18 $\mu V$	34.72 $\mu V$	44.11 $\mu V$

Caption:  $\mu V$  = microvolts

## DISCUSSION

The Tongue Frenulum Assessment Protocol in Babies<sup>13</sup>, validated for use in Brazil<sup>14</sup>, was applied to evaluate the interference of the frenulum in tongue movements and proved to be sensitive for identifying changes occurring before and after lingual frenotomy.

By observing data from the clinical examination 15 days after frenotomy, it was possible to notice an increase in the interval between feedings, a decrease in clicking sounds and noises during swallowing, findings that corroborate the study published by Martinelli et al<sup>10</sup>. The tongue actively participates in sucking and is essential for the adequate extraction of breast milk. The high tip of the tongue facilitates compression of the nipple to the base and, when lowered, the nipple expands to the transition region between the hard and soft palate, causing a vacuum and allowing milk to flow into the soft palate, thus ensuring greater effectiveness in the dynamics of sucking and extracting milk<sup>12</sup>. In this way, lingual frenotomy increases the number of suction and reduces the duration of pauses between suction<sup>10</sup>, allowing a recovery of adequate suction patterns<sup>16</sup>.

The tongue must be attached to the hard palate at rest and the presence of ankyloglossia leads the tongue to remain low in the oral cavity at rest<sup>14</sup>. In this clinical case, it was found that, 15 days after the procedure and until the end of the study, these signs changed, with the adequate positioning of the tongue at rest, attached to the hard palate during sleep, and

occluded lips<sup>6</sup>. The tongue plays an important role in the development of craniofacial structures. At rest, the light and constant pressure of the tongue against the hard palate, counterbalanced by the pressure provided by adequate lip sealing, serves as a guide for maxillary growth. Ankyloglossia makes it difficult to attach the tongue to the hard palate, a factor responsible for the correct craniofacial growth and development, and can cause adaptations in chewing, speaking and sucking functions<sup>6,7,12</sup>.

However, during crying, the tongue continued to tend to remain in the midline with elevation of the sides 15 days after frenotomy. In babies without changes to the lingual frenulum, the tongue tends to be in the midline or elevated during crying. Thus, the finding indicates limitation of the superior longitudinal muscle, whose primary function is dorsiflexion of the tip of the tongue, which allows the anterior part of the tongue to be positioned in the palatal rugae at rest, on the palate for swallowing and ensures elevation from the tip of the tongue to produce sounds that involve this characteristic<sup>17</sup>. The expected change was identified at 30 days and remained until the end of the study.

Case monitoring was also carried out through EMGs. During suction, the suprahyoid muscles, composed of the digastric, geniohyoid and stylohyoid muscles, can be monitored using EMGs, which allows changes in suction patterns to be monitored over time<sup>12</sup>.

It was possible to observe that, before frenotomy and after 15 days, the electrical activity of the right and

left suprahyoid muscles still remained asymmetric, showing the lack of balance between the muscles when performing suction and, possibly, compensations in the attempt to perform the function. More symmetrical values in electrical activity were observed at 30 days, possibly characterizing greater synchronicity and competence between the muscles analyzed after frenotomy, remaining so until the end of monitoring. Another study evaluated the electrical activity in the same muscles involved in the baby's sucking and concluded that the electrical activity was greater in children who had the lingual frenulum inserted in the middle third of the tongue and visible from the sublingual caruncle compared to those with the lingual frenulum fixed to the apex of the tongue and visible from the inferior alveolar ridge<sup>12</sup>. Throughout this study, it was possible to observe that there was an increase in electrical activity, with the EMG signal amplitude values observed at 60 days being similar to those found in babies who had the lingual frenulum inserted in the middle third of the tongue and visible from the sublingual caruncle<sup>12</sup>. Therefore, EMGs proved to be effective for diagnosing changes in the lingual frenulum, whose fixation points raise doubts regarding the restriction of tongue mobility<sup>12</sup>.

After frenotomy, and with the application of the Tongue Frenulum Assessment Protocol in Babies<sup>14</sup>, it was observed that there was no frenulum interference in tongue movements and remission of family complaints related to bottle sucking, although there was no possibility of resumption of natural breastfeeding. In this case, the interference of the frenulum in tongue movements contributed to early weaning<sup>12</sup>, as ankyloglossia is related to impairments in breastfeeding<sup>18</sup>. The main problems identified in cases of altered lingual frenulum in relation to breastfeeding are difficulties in latching on, nipple pain and fissures, prolonged breastfeeding time, reduced milk intake by the baby, weight loss, dehydration and growth deficiency. These factors can make it difficult to continue breastfeeding with consequences for the baby's health and, subsequently, for the development of chewing, speaking<sup>12</sup> and breathing<sup>19</sup> functions.

The use of a standardized clinical assessment instrument in this case study made it possible to analyze and compare results after frenotomy. The diagnosis of ankyloglossia is important and must be carried out early, still in the maternity ward<sup>20-22</sup>, to encourage breastfeeding, especially during the period recommended by the World Health Organization (WHO).

This case report showed that ankyloglossia had the clinical outcome of early interruption of exclusive breastfeeding<sup>18</sup>.

## CONCLUSION

After lingual frenotomy, the adequacy of the position of lips and tongue, during rest and sleep, occurred within 15 days, while the adequacy of the tongue position, during crying and sucking, occurred 30 days after the procedure, with improvement in the symmetry of the electromyographic activity values collected in the suprahyoid muscles between the right and left sides.

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#### Author contributions:

DFFB: Conceptualization; Writing - Original draft.

VVD: Conceptualization, Research; Writing - Original draft.

GBF: Project administration; Supervision; Writing - Review and editing.

#### Data Sharing Statement:

The authors inform that data from this study cannot be shared. The repository of the institution in which the study was conducted, and which was approved by the Research Ethics Committee, is currently being reformulated.