# UNUSUAL CASE OF ANKYLOGLOSSIA RECURRENCE AFTER FRENECTOMY IN A CHILD WITH CEREBRAL PALSY

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**Palavras-chave:** Anquiloglossia. Pessoas com Deficiência. Freio Lingual.

**RESUMO**

**Introdução**: A frenectomia é o tratamento mais indicado para os casos de anquiloglossia em crianças e jovens, sendo a recorrência extremamente rara. **Objetivo**: O objetivo é relatar o caso incomum de um menino de 8 anos, com paralisia cerebral, apresentando recidiva de anquiloglossia dois anos após a realização de frenectomia lingual**. Relato de caso:** Ao exame físico, em consulta de revisão, 2 anos após uma frenectomia lingual observou-se freio lingual levemente elástico, curto e aderido, presença de formato de coração na língua, restrição severa dos movimentos laterais e superiores, além de dificuldades na fala e deglutição. Uma nova frenectomia foi realizada após dois anos da primeira frenectomia , sob anestesia local, restaurando a mobilidade lingual. O paciente foi acompanhado periodicamente junto com a fonoaudiologia e após 2 anos da segunda cirurgia, a inserção firme do freio e a continuidade do aspecto bífido da língua foram novamente verificadas, porém, os movimentos da língua e as funções orais mantiveram-se satisfatórios. O paciente continuará a ser acompanhado periodicamente e, se houver alterações nas funções orais, uma nova intervenção será realizada. **Conclusão:** É fundamental o diagnóstico e tratamento multidisciplinar da anquiloglossia, além da necessidade de acompanhamento clínico periódico para identificar possíveis casos de recidiva, que, embora raros, podem ocorrer.

**Keywords:** Ankyloglossia. Disabled Persons. Lingual Frenum.

**ABSTRACT**

**Introduction:** Frenectomy is the most indicated treatment for ankyloglossia in children and adolescents, with recurrence being extremely rare. **Objective**: Here, we report the unusual case of an 8-year-old boy with cerebral palsy presenting recurrence of ankyloglossia 2 years after undergoing lingual frenectomy. **Case report**: On physicalexamination, in a follow-up, 2 years after a lingual frenectomy, we observed a slightly elastic, short and adhered lingual frenulum and the presence of a heart-shaped tongue, with severe restriction of lateral and superior movements, and difficulties in speech and swallowing. A new frenectomy was performed under local anesthesia, restoring lingual mobility. The patient has been followed-up periodically and has undergone speech therapy; 2 years after the second surgery, firm insertion of the frenulum and the continuity of the bifid aspect of the tongue have been again verified, but the tongue movements and oral functions remain satisfactory. The patient will continue to be followed periodically and a new intervention will be performed if there are changes in oral functions. **Conclusion**: Beyond the need forperiodic clinical follow-up, the diagnosis and multidisciplinary treatment of ankyloglossia are fundamental for identifying possible cases of recurrence, which, although rare, can occur.

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## INTRODUCTION

Ankyloglossia, or tongue-tie, is a congenital anomaly where a short lingual frenulum interferes with normal movement of the tongue, and the insertion of the frenulum may be anterior to the inferior alveolar crest. [1,2] Its prevalence is estimated at less than 1% to 10%, with a male to female ratio of 2.6:1.0 in newborns. [3,4] The wide variation of the prevalence data between studies may be associated with the difficulty in accessing dental services, especially in some populations, and a lack of diagnosis by dentists, leading to possible under-reporting of prevalence. [5]

There are several criteria for diagnosing ankyloglossia, such as the presence of a bifid aspect of the tongue during the protrusion movement (presenting a heart shape), the presence of restrictions in relation to the amplitude of tongue movements, and/or the impossibility of touching the palate with the tip of the tongue. [6, 7] Some treatments, such as clinical monitoring, preservation and frenotomy can be performed until more invasive surgical procedures such as frenectomy can be performed. Frenectomy consists of complete removal of the frenulum and is most commonly performed in children and adolescents because of better understanding and acceptance of the procedure. Frenectomy has a high success rate, with few recurrences postoperatively. [8]

In the present paper, we report an unusual case of an 8-year-old male patient who presented with recurrence of ankyloglossia 2 years after undergoing frenectomy.

## CASE REPORT

An 8-year-old boy with cerebral palsy was referred by his speech-language pathologist with the complaint of difficulty in phonation and deglutition as a result of ankylosis recurrence 2 years after undergoing lingual frenectomy. Analysis of the dental records of the previous surgical procedure verified that a frenectomy had been performed 2 years ago and that the patient had history of collaborative behavior despite mild cognitive deficit. We observed that the patient did not attend the periodic follow-up consultations after the surgical procedure.

Clinical examination verified the poor mobility and bifid aspect (heart-shaped appearance) of the tongue when lifted. We also observed little tongue elasticity, lingual frenulum length > 1 cm, appropriate lateralization movement of the body of the tongue, anterior movement with moderate restriction, and impossibility of tongue tip touching the lingual surface of the maxillary incisors (Figure 1). After evaluation by the bucomaxilofacial surgeon, we decided to perform another frenectomy. The surgery was performed, with the mother's consent, under local anesthesia (2% lidocaine with epinephrine 1:100.000) and with the pediatric dentists' assistance for better behavior control. Resection of the lingual frenulum (complete removal) was performed and a simple catgut 5-0 suture was used to immobilize the tongue. Immediately after the surgery, we observed the re-establishment of tongue mobility (Figure 2).

Seven days after the frenectomy, the patient returned for a clinical follow-up of his postoperative condition, confirming satisfactory healing. Subsequently, he was clinically re-evaluated after 15 days and 1 and 3 months; and in all of these appointments were verified satisfactory clinical evolution of the case. Since then, he has been followed-up every 6 months in partnership with speech therapy. After 2 years of follow-up, we have observed that the tongue is functionally adequate, with satisfactory lateral and anterior movements, where the tip of the tongue can touch the lingual surface of the maxillary incisors. In addition, phonation and deglutition have remained satisfactory. Besides that, firmer insertion of the lingual frenulum has been verified, and the tongue tip still presents the bifid aspect when lifted (Figure 3). However, new surgical intervention is not needed at the moment. The patient will continue to be followed-up, and if new surgical treatment is needed, will be carried out a multidisciplinarily planning.

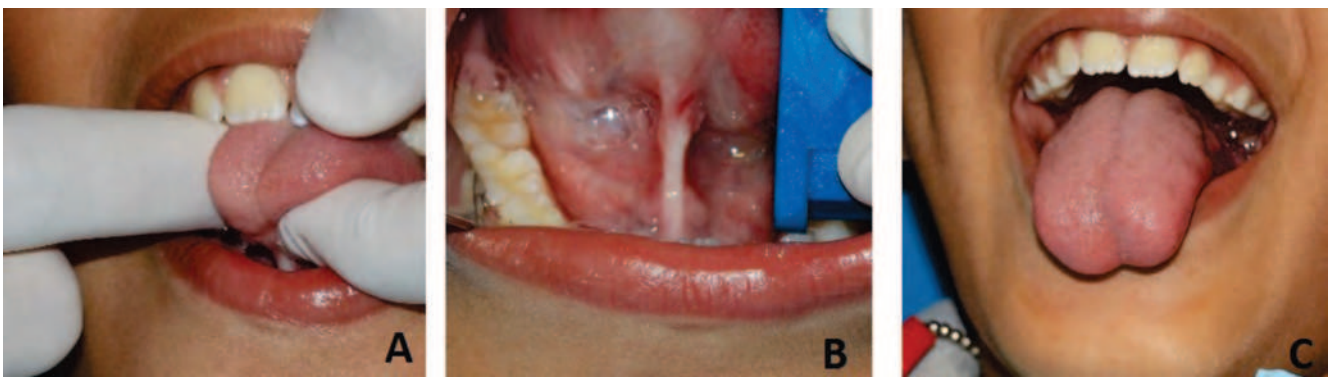



***Figure 1***: *A: Intraoral photography of initial clinical examination showing restriction of the superior movements of the tongue, whose tip cannot rest on the lingual surface of the maxillary incisors. B: A less elastic lingual frenulum inserted from the base of the tongue to the crest of the alveolar ridge. C: The bifid appearance, besides the restricted anterior movements.*

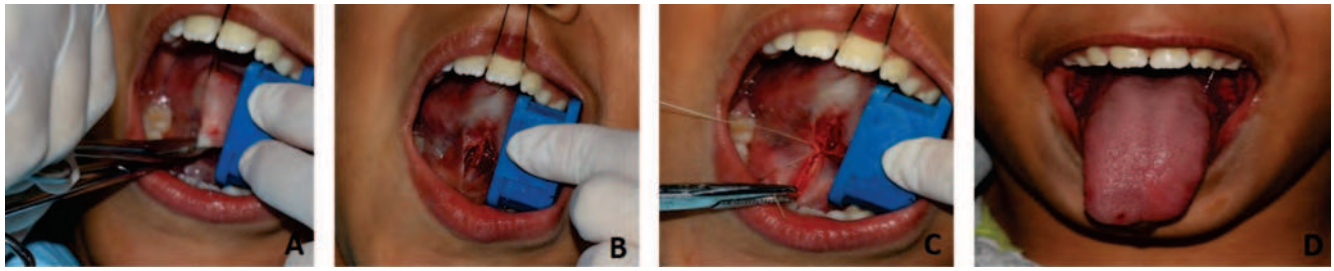



***Figure 2***: *A, B: Images of the surgical procedure, showing debridement and complete resection of the lingual frenulum and release of the tongue movements. C: Local suture performed in the area at the end of the surgery. D: Intraoral photography showing the re-established lingual mobility just after the second frenectomy.*




***Figure 3***: (A) *Firm insertion of the lingual frenulum verified 2 years after the second frenectomy.* (B and C) *besides the maintenance of the bifid aspect of the tongue, lingual movements and oral functions (deglutition and phonation) were maintained satisfactorily.*

## DISCUSSION

Ankyloglossia is a congenital anomaly that causes speech and feeding difficulties and is not correlated with any other specific cause. There are some standards for recognizing and diagnosing ankyloglossia, such as the appearance of the tongue when lifted, frenulum elasticity, lingual frenulum length when the tongue is lifted, lingual frenulum attachment to the tongue, lingual frenulum attachment to the inferior alveolar ridge, lateralization, and tongue lifting. [1,9]

When ankyloglossia is diagnosed, the treatment options include frenotomy and frenectomy. [1,10] In frenotomy, the frenulum is only superficially cut, and it is even performed only with topical anesthesia. It is used most often in babies, as it is less invasive and faster. Frenectomy is a more invasive surgical procedure that requires local anesthesia and consists of complete debridement of the lingual frenulum, whose insertion must be removed from the base of the mandibular bone. Thus, the choice of a less invasive treatment, such as frenotomy, may cause relapses, as the frenulum can have several insertions, while the recurrence of ankyloglossia after frenectomy is rarer. [1,8,10,11]

In the present case, the first-choice treatment was frenectomy, but even then, the patient returned after 2 years with the lingual frenulum well-inserted in the inferior alveolar crest, besides problems with phonation, indicating the need for new surgical intervention. In the searched literature, we found no other reports of recurrence of ankyloglossia after frenectomy.

Different factors are related to the healing process of wounds in the oral soft tissues. [12] It is suggested that the relapse observed in the present case is related to an individual characteristic of the patient, with the formation of a scar tissue that is more fibrous than normally observed. It could be the reason that, even after the second procedure, the patient presented a very fibrous insertion of the lingual frenulum in the postoperative follow-up period, revealing an individual physiological problem. Despite this, no need for new interventions has been observed to date.

In addition, to avoid recurrence after frenectomy, it is important that the patient perform myofunctional tongue rehabilitation exercises with a speech therapist, as observed in many other successful cases. [13] In the present case, the patient also underwent multi-professional treatment with a speech therapist to complement the surgical treatment and improve the prognosis. The importance of maintaining this treatment was explained to his mother.

The dental management of children, especially when they are patients with special needs, is challenging. In this context, behavioral control is one of the main issues in enabling adequate management of the patient and for performing the necessary procedures effectively. [14,15] In the present case, multidisciplinary care with a dental surgeon and pediatric dentists allowed us to perform the surgical treatment without requiring sedation or general anesthesia. It should be noted that this multidisciplinary and multi-professional collaboration was of fundamental importance

for ensuring the most adequate planning, efficient management, and the best prognosis for the patient.

We can conclude that, after the 2-year follow-up period, the multidisciplinary and multi-professional treatment, including a new lingual frenectomy and speech therapy treatment, facilitated the successful management of a case of ankyloglossia recurrence in a child with cerebral palsy.

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