

Management of Familial Ankyloglossia in Pediatric Patient Under Electrocautery Procedure

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ABSTRACT

Ankyloglossia, commonly referred to as tongue-tie, is a congenital condition affecting the tongue. It is characterized by a short, thick, and tight lingual frenulum that attaches to the tip of the tongue, leading to restricted tongue movement. This condition can result in various issues, such as difficulties in mastication, challenges in pronouncing consonants, and poor oral hygiene. A nine-year-old male patient came to the Department of Pediatric Dentistry, Universitas Airlangga Dental Hospital, Surabaya, complaining of restricted tongue movement, which was causing difficulty in articulating certain letters such as "r, s, n, d." Intraoral examination revealed a shortened lingual frenulum, measured at 8.5 mm, and was diagnosed as ankyloglossia class II according to the Kotlow's Classification. A lingual frenectomy using electrocautery was performed with parental consent and no sutures were needed. The patient was prescribed antibiotics, analgesics, and mouthwash, and instructed not to drink or eat hot food and to maintain oral hygiene. Follow-up visits after one week showed successful wound healing and improved tongue mobility. Ankyloglossia can significantly impact patients by restricting tongue movement and causing speech difficulties, highlighting the importance of timely treatment through surgical intervention. The advantages of using electrocautery for lingual frenectomy include a bloodless surgical field, the absence of sutures, and a shorter treatment duration, making it an excellent choice for pediatric patients.

Keywords: familial ankyloglossia; lingual frenulum; electrocautery; human & health

INTRODUCTION

Ankyloglossia, commonly known as tongue-tie, is a congenital condition characterized by a short, thick, and tight lingual frenulum attaching to the tip of the tongue, resulting in restricted tongue movement [1,2]. The reported prevalence of ankyloglossia in the literature varies widely, ranging from 0.1% to 10.7%. Its occurrence is higher in neonates (1.72%–10.7%) compared to children, adolescents, or adults (0.1%–2.08%), with an incidence ranging between 4.2% and 10.7% and a male-to-female ratio of 3:1 [3]. Ankyloglossia can lead to difficulties in breastfeeding during infancy, mastication, pronunciation of consonants, oral hygiene, gingival recession of lower front teeth, malocclusion, excessive salivation, and instability of mandibular prostheses [3,4].

Ankyloglossia can occur as an isolated condition in an otherwise healthy child or be associated with rare syndromes such as X-linked cleft palate, Van der Woude syndrome, Oral-facial digital syndrome, Beckwith

– Weidman syndrome, and Smith–Lemli–Opitz syndrome [1,2].

According to Kotlow, the severity of ankyloglossia is determined by the length of the "free tongue," defined as the distance from the insertion of the lingual frenulum into the base of the tongue to the tip of the tongue [5]. Management options for ankyloglossia include observation, surgical interventions (frenotomy, frenectomy, frenuloplasty) using conventional techniques (scalpel), and electrocautery [6]. This case presents the management of familial ankyloglossia in pediatric patient under electrocautery procedure.

CASE REPORT

A nine-year-old male patient came to the Department of Pediatric Dentistry, Universitas Airlangga Dental Hospital, Surabaya, Indonesia, with the main complaint of difficulty moving his tongue freely.

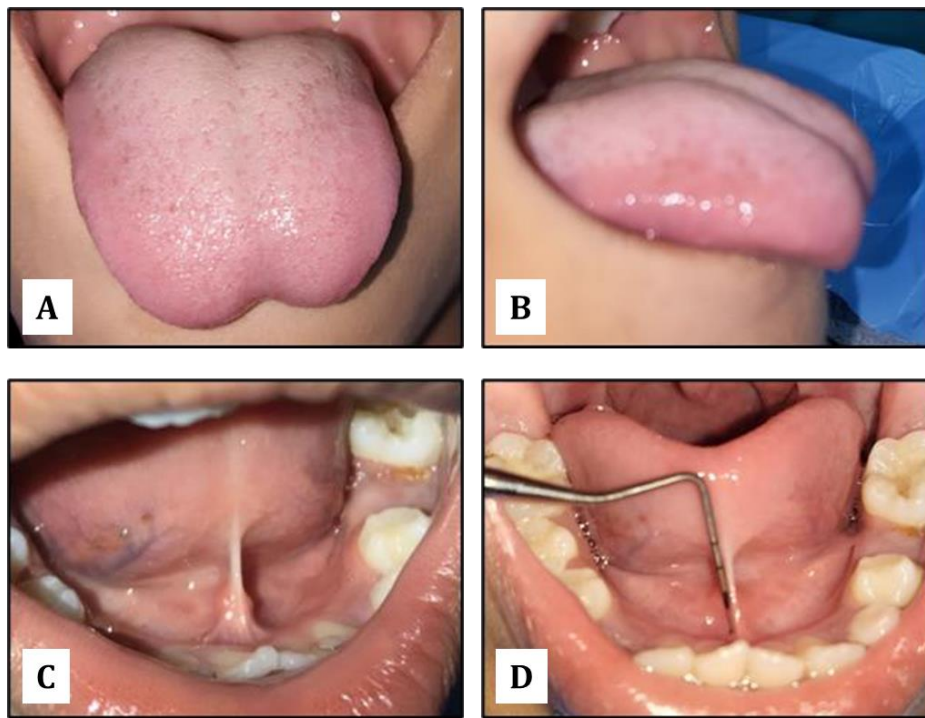


FIGURE 1: Pre-operative intraoral photograph. (A, B) Heart-shaped tongue during protrusion. (C) Limited movement of tongue. (D) Lingual frenulum with class II ankyloglossia.

Resulting in speech difficulties when pronouncing certain letters, such as "r, s, n, d". Family history revealed that the patient's father and sibling also have ankyloglossia. The extraoral examination showed no pathological abnormalities. Intraoral clinical examination revealed limited tongue movements due to a short lingual frenulum, and a heart-shaped appearance when the tongue is protruded (Figure 1A to C). The distance between the insertion point of the lingual frenulum and the tip of the tongue was approximately 8.5 mm (Figure 1D). Therefore, the patient was diagnosed with class II ankyloglossia according to Kotlow's classification (Table 1).

TABLE 1: Kotlow's Classification.

Category	Length of the free tongue
Normal	>16 mm
Class I: mild	12-16 mm
Class II: moderate	8-11 mm
Class III: severe	3-7 mm
Class IV: complete	<3 mm

The management of this case is a lingual frenectomy using electrocautery. Following consent from the patient's parents, the procedure begins with extraoral asepsis using 70% alcohol, followed by intraoral asepsis of the operating area using 10% povidone iodine (Figure 2A). Subsequently, a topical anesthetic gel is applied to the underside of the tongue, followed by local anesthesia (1:100,000 adrenaline) administered on both sides of the frenulum (Figure 2B). To facilitate visualization of the frenulum without disturbing tongue mobility, suture silk is threaded through the tip of the tongue for traction (Figure 2C).

The procedure utilizes an ART-Electron® (E1) unit set to "cut + coagulant 1" mode with a speed of 5 and a T5 tip for the incision (Figure 2D), followed by the removal of muscle fibers using a T2 tip (Figure 2E). The frenulum leaves a diamond-shaped wound with a diameter of approximately 8 mm and brown color (Figure 3A). Monitored for 10 minutes. There was no bleeding, thus suturing was not necessary. Immediate post-operative improvement was observed in tongue protrusion and mobility (Figure 3B). The patient is prescribed antibiotics (amoxicillin 250 mg), analgesics (paracetamol 250 mg), and 0.2% chlorhexidine gluconate mouthwash. Post-operative instructions include refraining from eating and drinking for one hour, avoiding hot, cold, sour, and spicy foods, following a recommended soft diet for several days, gently rinsing the mouth, avoiding touching the wound, and maintaining oral hygiene at home. Follow-up appointments are scheduled for further assessment.

The observation one day post-operative revealed the presence of slough at the surgical site, indicating healing process. The scar presents a diamond-shaped appearance with white and yellow colors (Figure 4A). The patient reported no pain at the surgical site. At the one-week follow-up appointment, the wound was observed to be healing without any complications. The diameter has further reduced, and the color transitions to white, indicative of the ongoing healing process (Figure 4B). The patient demonstrated improved tongue mobility. Post-operative exercises commenced after one week, including touching the palate with the tip of the tongue, moving the tongue laterally, rotating it, and stretching it with a protrusive motion.

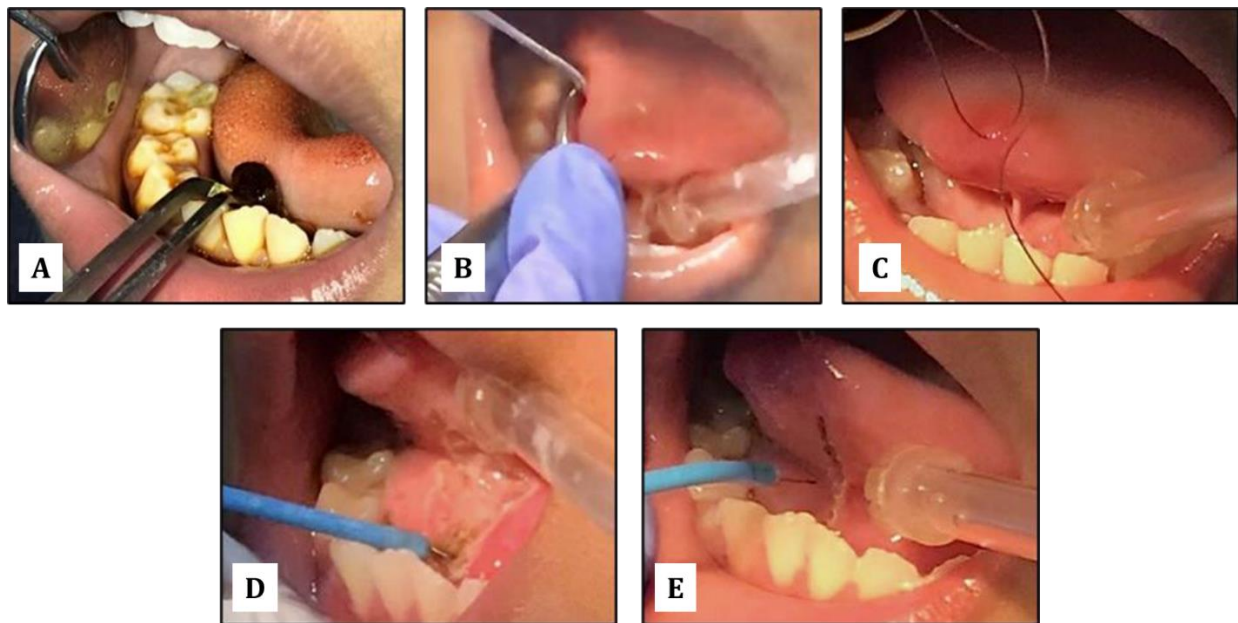


FIGURE 2: Management of lingual frenectomy. (A) Asepsis in the working area. (B) Local anesthesia. (C) Traction of the tongue. (D) Incision of the lingual frenulum. (E) Removing the muscle fibers.

The patient was instructed to perform these exercises 3–4 times daily for 2 minutes each session. Additionally, the patient's parents were advised to consult a speech therapist for further improvement in speech concerns. After one year and a six-month observation period, the electrocautery frenectomy showed signs of a complete healing process without any complications (Figure 4C).

Chewing, breastfeeding in infants, and oral hygiene. The tension from a tight frenulum on the tongue may lead to gaps between lower front teeth (diastema) and receding gums. Limited tongue mobility particularly affects the articulation of consonants like "s," "t," "z," "d," "l," "ch," "j," "zh," and "th." [3,4]. Familial ankyloglossia is an exceptionally uncommon condition. The precise pathological mechanism of ankyloglossia is still unknown.

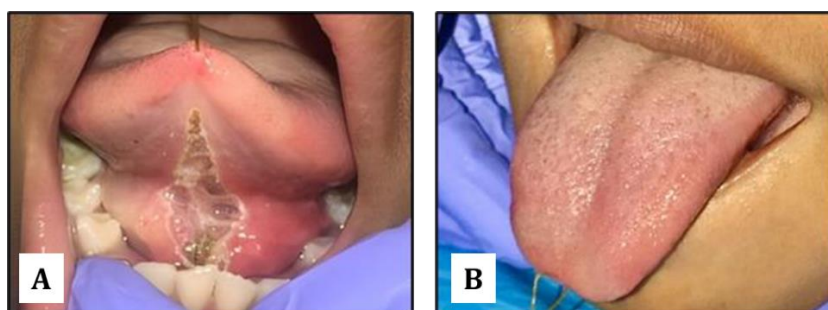


FIGURE 3: (A) Lingual frenectomy done by using electrocautery. (B) Immediate post-operative when tongue protrusion.



FIGURE 4: (A) One-day follow-up intraoral photograph. (B) One-week follow-up intraoral photograph. (C) One year and six months follow-up intraoral photograph.

DISCUSSION

Ankyloglossia, commonly referred to as tongue-tie, is a congenital condition characterized by clinical indications of an abnormally short lingual frenulum, resulting in restricted movement of the tongue. The lingual frenulum is a thin membrane that extends from the underside of the tongue to the floor of the mouth [1,2]. Ankyloglossia can impact speech, uncertain, and its definitive hereditary characteristics have yet to be clarified. There have been reports ankyloglossia seems to be inherited as an autosomal dominant trait with incomplete penetrance or due to an autosomal recessive trait. To identify the defective gene(s) causing ankyloglossia in patients, linkage analysis should be feasible. Reports indicate that mutations in the T-box transcription factor (TBX22) gene during palatogenesis are responsible for ankyloglossia [7,8]. Ankyloglossia can also manifest as part of the X-linked cleft palate due to TBX22 gene mutations. [9,10].

In normal conditions, the frenulum should attach to the tongue approximately 1 cm behind the tip, while its attachment to the inferior alveolar ridge should be close to or within the genioglossus muscle, located on the floor of the mouth. Ankyloglossia is categorized into four groups according to Kotlow's Classification, which is based on the length of the "free tongue", defined as the distance from the tip of the tongue to the insertion point of the lingual frenulum. A normal free tongue length is 16 mm. Class I represents mild ankyloglossia with a free tongue length of 12-16 mm; class II signifies moderate ankyloglossia with a free tongue length of 8-11 mm; class III denotes severe ankyloglossia with a free tongue length of 3-7 mm; and class IV indicates complete ankyloglossia with a free tongue length of ≤ 3 mm [5].

Multiple treatment options exist for ankyloglossia, ranging from observation to surgical interventions such as frenotomy, frenectomy, and frenuloplasty, using either conventional techniques like a scalpel or electrocautery [6]. Frenectomy involves the complete removal of the frenulum. In this case, frenectomy was conducted using electrocautery, which offers several advantages over conventional techniques. These include shorter operative working time, tissue cauterization and sterilization, hemostasis, minimal bleeding, clearer surgical fields, less local anesthesia, and fewer post-operative complications such as pain, swelling, and infection [11]. Conventional frenectomy procedures with a scalpel often result in post-operative pain and discomfort due to blood loss, larger surgical wounds, and the necessity for sutures [12,13].

In the present case, there were no reports of swelling, pain, or edema post-operative. The patient showed positive signs of healing after one week. Lingual frenectomy with electrocautery shows good wound healing. During the one-day follow-up after the frenectomy, the wound presents a diamond-shaped appearance with white and yellow colors. By the one-week follow-up, the wound diameter has

further reduced, and the color transitions to white, indicative of the ongoing healing process. After one year and six months of follow-up, complete healing of the wound is observed. The patient's parent was satisfied with the improved tongue movements. It's in accordance

With the advantages of frenectomy using electrocautery. It's essential to engage in post-operative tongue muscle training to stretch the tongue muscles through lateral, upward, twisting, and protrusive motions. These exercises should be done 2-3 times daily for 2 minutes each session. The patient encountered difficulty articulating certain sounds, which necessitated speech therapy for improved pronunciation. These exercises can be practiced independently at home or under the guidance of a speech therapist in the hospital. Ideally, surgery should take place before any abnormal speech or swallowing patterns develop [14]. For older individuals undergoing surgery, it's advisable to consult a speech therapist to restore normal tongue function [15,16]. Follow-up was conducted one year and six months post-operative. There were no complaints regarding the surgical site of the frenectomy. The patient practiced tongue exercises at home with parents and can articulate the letters that were previously of concern.

CONCLUSION

Management of familial ankyloglossia through frenectomy in pediatric patients under electrocautery procedure gives positive outcomes without complications. Utilizing electrocautery for frenectomy presents a favorable choice. Ankyloglossia should be treated as early as possible since early detection and timely surgical intervention typically help patients avoid prolonged complications.

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