



Management of a Rare Case of Total Ankyloglossia Caused by POTASH Chewing. A Case Report

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Abstract Oral chemical burns result due to ingestion of a chemical accidentally into the mouth, causing burns in the oral mucosa, esophagus, stomach, and upper digestive tract if swallowed. Oral chemical burns mainly happen in children. The current case report describes a rare case of a child who ingested POTASH (potassium hydroxide (KOH) (a caustic chemical used as a detergent in clothes washing in low socioeconomic standard Egyptian places) at home and developed chemical burns that resulted in total ankyloglossia. This case was further complicated through mishandling by a general dentist. This case report describes a detailed protocol for the management of such cases.

Keywords Ankyloglossia · Tongue tie · Lingual plasty · Tongue plasty

Introduction

According to Kotlow [1] and Jamilian [2] et al., Ankyloglossia is defined as a limitation of the protrusion and elevation of the tip of the tongue either as a result of shortness of the frenum or the genioglossus muscles or both. It has a genetic etiology and occurs in children between 1 and 3 years old. The case is considered as a hyper-trophic lingual frenulum when tongue mobility is reduced [3, 4]. Limitation in tongue mobility results in poor feeding, which affects weight gain, and changes in the bone growth of the child's oro-facial structures, oral functioning, or both. The

child's speech is also affected since the phonetic sounds that depend on tongue articulation are affected [5–7]. Due to insufficient palatal support in these cases, swallowing is also affected as the patients develop atypical deglutition movements [6, 7]. This case report presents clinical examination, the presurgical activity, the surgical approach used, and the postsurgical rehabilitation.

Case Presentation

A 7-year-old male patient was referred suffering from total inability to move his tongue. The history of chief complaint according to the father's own words is the chewing of POTASH (potassium hydroxide (KOH)), a caustic chemical used as a detergent in clothes washing in low socioeconomic standard Egyptian places. It has a candy-like appearance and when diluted it looks like milk. The insult happened one year ago and led to a burn on the child's tongue that partially affected the tongue movement. The family went to a general dentist who used a scalpel to release the tongue from the floor of the mouth without suturing or performing any surgery that aggravated the condition (still according to the parent's own words). The child began to eat improperly, and they blended his meals for him. He also developed mouth breathing with habitual mouth opening and suffered from difficulty in swallowing.

Clinical examination revealed a total ankyloglossia with the absence of any lingual frenum and the presence of a tough fibrous union between the ventral surface of the tongue and the floor of the mouth (Fig. 1a). The child could not move his tongue and has flaccid and hypo-plastic tongue muscles. In this particular case, the uniqueness lies in that the fusion observed in the floor of the mouth and the ventral side of the tongue is total. In other classifications, the

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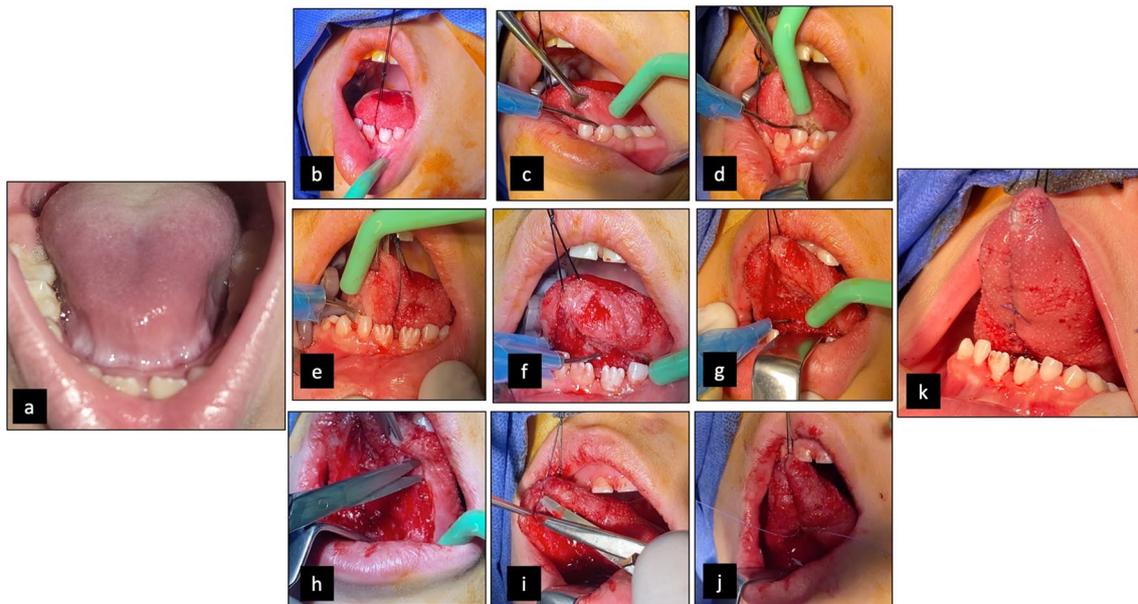


Fig. 1 **a** preoperative photograph of the patient trying to move his tongue, **b**. application of black silk retracting suture at the tip of the tongue, **c**. using the laser the floor of the mouth is separated from the ventral aspect of the tongue started posteriorly from the left side, **d** the completion of separation using the laser to the tip of the tongue, **e** the same procedure is performed in the right side, **f** a partial split

thickness of the genioglossus muscle ventrally is dissected by the laser, **g** a partial split thickness of the hyoglossus muscle laterally till the styloglossus muscle posteriorly is performed by the laser, **h** using a Metzenbaum scissors in a snip action, **i** using a Metzenbaum scissors in a snap action

term total ankyloglossia has been limited to cases where the lingual frenum is less than 4 mm long as referenced by Kotlow's classification [1].

Treatment

A hematogram in addition to liver enzymes and kidney function tests was ordered for the patient to assure his fitness for the surgical procedure under general anesthesia. The patient's weight was recorded at the time of surgery to be 18 kg. The surgery was performed under general anesthesia via nasotracheal intubation. Povidone-iodine solution was used to scrub the surgical field, intra-orally then extra-orally, followed by draping of the patient with sterile disposable towels revealing only the surgical field. Local infiltration anesthesia was injected at the sides of the tongue and the anterior region intramuscularly using Articaine hydrochloride 40 mg/ml + 0.01 mg/ml epinephrine (Inibsa Dental S.L.U. Ctra. Sabadell a Granollers, Lliçà de Vall, Barcelona, Spain). The surgical field was then sterilized and draped to ensure asepsis.

The tongue was stabilized by 1 zero black silk retracting suture at the midline, which is used to retract the tongue during the surgery. Alli's Forceps was also used to help pull the tongue during surgery (Fig. 1b). The submandibular glands ducts were identified. Using Sirolaser Diode Laser

(Dentsply Sirona. Charlotte, NC, USA), the fibrous tissue attached to the tongue's ventral surface was freed from the floor of the mouth at a level superior to the submandibular glands' ducts. The layer was continued bilaterally and then deepened to the level of the genioglossus muscle where it was dissected by the laser. A partial split in the genioglossus muscle ventrally and the hyoglossus muscle laterally till the styloglossus muscle posteriorly to construct a new ventral aspect of the tongue and to create a separate floor of the mouth to allow for proper movement of the tongue was made. When releasing the muscle tissue, the tongue started to be retracted more and freed to produce a diamond shape surgical field (Fig. 1c–g).

Using Metzenbaum scissors by the snip and snap action, the genioglossus muscle was dissected and released from the lateral aspect of the tongue to free it from the dorsal surface of the tongue and allow for approximation of edges and closure of the ventral surface of the tongue (Fig. 1h, i).

A tongue tip was created at first; then, the ventral surface of the tongue was sutured using the rule of halves; the key suture is performed as a standard interrupted suture while the other sutures are inverted (Fig. 1j, k). The ventral aspect was then sutured to the floor of the mouth. The patient was given the postoperative tongue rehabilitation instruction. The regimen included moving the tongue forward, bilaterally to open the mouth as much and trying to touch the hard palate with the tongue; in the form

of 10 sets per day, 10 times at each set. The patient was also referred to a speech and language therapist to help achieve normal phonetics. Suture removal was done after 1 week, and the postoperative image showed partial healing. (Fig. 2a–d). The patient was checked monthly for the next six months to assess healing and the improvement of the child's general condition (Fig. 2f, e). Furthermore, to clinically assess the mobility and the size of the tongue in addition to phonetics.

Treatment Outcome

Healing was uneventful. The patient just complained of postoperative pain that was treated with NSAIDs whenever needed. The interrupted key suture was removed one week postoperatively. The site of suture retraction developed an ulcer that was treated using topical protective paste. The parents were instructed not to feed him spicy and salty food. The tongue can be moved 20 mm from the floor of the mouth. The patient started to develop acceptable phonetics. At the follow-up visits, the patient weighed 20 kg and 25 kg at one and six months, respectively.



Fig. 2 Postoperative clinical photograph; (a–d one week postoperatively) a the tongue in position, b the patient moves the tongue anteriorly outside the mouth, c the patient moves his tongue toward the right, d the patient moves his tongue toward the left. (e, f six month postoperatively). the patient moves the tongue upwards, f the patient moves the tongue anteriorly outside the mouth

Discussion

In the presented case, the ankyloglossia occurred as a result of rare, combined conditions, chewing a caustic chemical (potassium hydroxide (KOH)) which caused a chemical burn to the floor of the mouth, and a faulty surgical intervention. This resulted in total ankyloglossia with the absence of lingual frenum in a 7-year-old patient. Since the insult, the child suffered from an incapability of mastication and swallowing in addition to difficulty in talking. This results in severe loss of the patient's weight, and difficulty in speech and communication. Furthermore, the child dropped a year from his school being incapable of communicating with his colleagues and teachers. According to Cuestas et al., ankyloglossia could result in breastfeeding difficulties, speech and dentition disorders, and social problems related to the functional limitation of the tongue [8].

There is continuing controversy over the diagnostic criteria and treatment of ankyloglossia [9]. Kotlow's classification was used in the present study as it quantifies the problem based on the frenal length which gives the surgeon a proper way to clinically assess the condition and the surgical prognosis [1]. It classifies ankyloglossia into four classes based on the frenal length as follows: Class I: Mild ankyloglossia (12–16 mm); Class II: Moderate ankyloglossia (8–12 mm), Class III: Severe ankyloglossia (4–8 mm), Class IV: Complete ankyloglossia (<4 mm). Class III and IV tongue-tie categories should be given special consideration because they severely restrict the tongue's movement.

Frenectomy is the line of treatment for ankyloglossia; the incision can be made with a cold scalpel, scissors, electrosurgical unit, or laser [8–12]. Heller et al. recommended the Z-plasty for the frenum rather than the horizontal to vertical frenuloplasty concerning tongue lengthening, protrusion, and articulation improvement for patients with symptomatic ankyloglossia [10]. This was debated by Choi et al., who considered the conventional surgery with Z-plasty not effective [11]. They recommended performing myotomies of the genioglossus muscles to free the muscles. Both techniques depend on the presence of a frenum and a bulk of genioglossus muscle. However, in the presented case there was a complete absence of the frenum in addition to the presence of fibrotic muscle. Therefore, the innovative technique used aimed to free the ventral aspect of the tongue from the floor of the mouth and then perform a partial split in the genioglossus muscle and the hyoglossus muscle till the styloglossus muscle posteriorly to construct a new ventral aspect of the tongue and create a separate floor of the mouth with amount to allow for proper tongue movement. Laser was used to avoid the massive bleeding that could happen while using the surgical blades.

Immediately after the surgery, the patient was instructed to perform a tongue rehabilitation regimen which helped strengthen his tongue muscles allowing for the normalization of the functions especially swallowing and correction of speech. This agreed with Ferrés-Amat et al., who recommended a rehabilitation regimen that starts before the surgery by one week and continues after the surgery. Nonetheless, in the present case, the rehabilitation program started after the surgery depending on the previous muscles and actions memory. The child was also referred to a speech and language therapist to improve his speech and phonetics [12].

Finally, it was observed that within one month postoperatively the patient gained 3 kg of weight and a total of 7 kg at the six-month follow-up. His parents reported better eating habits and physiological improvement. This case report highlighted the appropriate response to severe oral chemical burns and the importance of having referral strategies and capacity decisions.

Conclusion

The results of the current study revealed that utilizing the aforementioned surgical technique of lingual plasty in combination with myofunctional rehabilitation improves the lingual mobility, feeding ability, and psychological status of the presented case.

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Declarations

Conflict of interest The author declares no conflict of interest.

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