Case Report

Management of Ankyloglossia with different severity: a case series in dental surgery

Stephanus Christianto[1]
Winarno Priyanto[2]
Endang Sjamsudin[2]

[1]Department of Oral Surgery, Faculty of Dentistry, Maranatha Christian University
[2]Department of Oral Surgery, Faculty of Dentistry, Padjadjaran University, Indonesia

* Correspondence:
endang.sjamsudin@fkg.unpad.ac.id

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ABSTRACT

Introduction: The lingual frenulum is an important oral structure that plays an important role in the act of suction, speech, and feeding. *Ankyloglossia* (tongue tie) is a congenital anomaly that results when lingual frenulum is too short and adhere to tip of tongue, limiting the tongue movement. This case report aimed to explain management of Ankyloglossia with different severity. **Case Report:** This case report reported three cases of *ankyloglossia* with different ages and complaints of difficulty in feeding and speech. The patients were treated for lingual frenectomy procedure under general anesthesia using one haemostats method. The haemostats were used to clamp the upper aspect of frenulum which might have been helpful to guide the incision close to ventral surface of tongue. Care had to be taken not to injure the submandibular ducts when making incision at lower part of tongue. The result after 1 week of surgery: the wound was completely healed. **Conclusion:** *Ankyloglossia* will impair the function of suction, speech and feeding that affecting a considerable number of infants and children. It is important that accurate information and guidance are given to parents with regard to the indications and potential benefit of tongue tie revision. These case series offer guidelines which can be used by general and pediatric dentist for examination, diagnosis and treatment of *ankyloglossia* in different severity.

KEYWORDS

Ankyloglossia, congenital, lingual frenulum

INTRODUCTION

The tongue is a vital anatomical structure in the oral cavity, which plays an important role in the process of swallowing, speaking, tasting and feeding.[1] The lingual frenulum is a mucous membrane attached to the ventral part of the tongue that connects the tongue to the floor of the mouth and is part of the tongue's anatomy.[2] Normal tongue movement is defined as the ability of the tongue tip to protrude out of the mouth without changing the shape of the tongue to be clefting, when the tongue is capable of normal swallowing, when there is no difficulty speaking due to limited tongue movement, and when the tongue can protrude toward the upper and lower lips without feeling resistance or pain.[3]

Ankyloglossia is a medical term that derives from two Greek words: kilos (curved) and gloss (tongue). This term is also known as tongue tie, and it describes the limited movement of the tongue that prevents it from performing normal tongue movements. This condition is distinguished by the presence of a short and thick lingual frenulum that restricts tongue movement.[3,4] Based on the

restriction of tongue movement, *ankyloglossia* is categorized into different severity.[5]

According to many case studies, the prevalence of *ankyloglossia* ranged from 0.3% 5 to 16%,[6] with men having a larger ratio than women, namely 2.5:1.0.[7] *Ankyloglossia* treatment options include observation, speech therapy, and frenectomy surgery, each treatment could be indicated in different severity.[8]

The dentist faces a unique challenge when diagnosing and treating *ankyloglossia* in children. Children with *ankyloglossia* may experience problems such as malnutrition, difficulty in swallowing, speech difficulties, and mandibular prognathism. [9-11] There are numerous surgery and therapeutic options for cases of *ankyloglossia* depending on its severity.[12-15] In this article, a series of three cases of *ankyloglossia* will be presented in different ages of patient (6 months, 3 years and 6 years old), which aim to provide a specific description of the case ankyloglossia and guidelines that must be considered before treatment in case *ankyloglossia*. This case report aimed to explain management of Ankyloglossia with different severity.

Case Report

Case 1.

A 9-month-old female infant patient was referred to the oral and maxillofacial surgery department at Hasan Sadikin Hospital, Bandung, with the main complaint of difficulty in being breastfed. The medical and family history showed no contribution to the patient's main complaint. In intraoral examination, the lingual frenulum was short and thick and obstructed the protrusion of the tongue (Figure 1). The patient was diagnosed with differential diagnosis of severe *ankyloglossia* based on classification *Kotlow's* [5](Table 1). In the treatment plan, this patient was scheduled for surgery to undergo lingual frenectomy with incising of lingual frenulum to release the restriction of tongue movement and suture was placed in the incision area (Figures 2 and 3). The patient was under control one week after surgery, and the prognosis of surgery was good with the improvement of the tongue movement. Accordingly there was no more difficulty in breastfeeding (Figure 4).

Table 1: Kotlow's classification

Class I: Mild *ankyloglossia*	12-16 mm
Class II: Moderate *ankyloglossia*	8-11 mm
Class III: Severe *ankyloglossia*	3-7 mm
Class IV: Complete *ankyloglossia*	<3 mm

Figure 1. Presence of thick and short lingual frenulum pre operatively

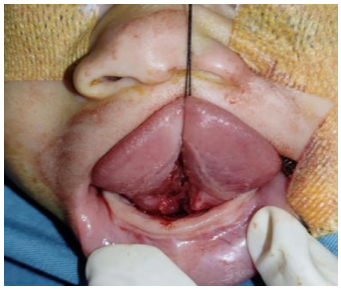

Figure 2. Lingual frenulum incised to free the tongue movement

Figure 3. Sutures placed post operatively

Figure 4. Postoperative day-7 showed no more restriction movement of tongue

Case 2.

A 3-year-old boy patient came to the Oral and Maxillofacial Surgery at Hasan Sadikin Hospital, Bandung, with the complaint of difficulty in swallowing, the patient's parents also complained that the child had speech difficulties. According to family and medical history examinations, no abnormalities were found. Meanwhile, based on clinical examination, it was found that the lingual frenulum was thick, short, and limited in the movement of the tongue, especially when touching the anterior palate (Figure 5). Based on clinical examination the patient was diagnosed with a differential diagnosis of class III of severe *ankyloglossia* according to Kottlow classification. The treatment plan of the patient was frenectomy under general anesthesia to release the lingual frenulum (Figure 6). Postoperative 1 week showed tongue movement within normal limits without any difficulty in swallowing (Figure 7). The patient was then referred to speech therapy to train his tongue movement, especially when speaking. Accordingly the treatment of *ankyloglossia* could be considered success in this case with good prognosis.

Figure 5. Preoperative view of *ankyloglossia*

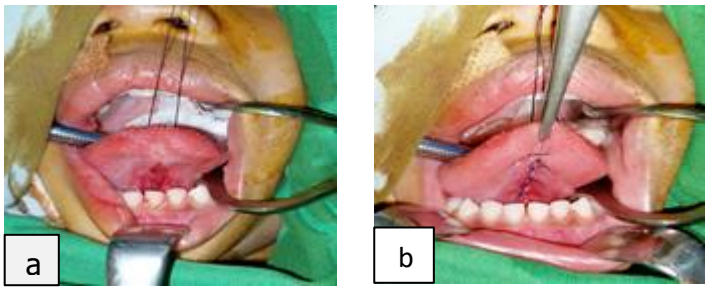

Figure 6. (a) Frenectomy was done at lingual frenulum, (b) Sutures placed

Case 3.

A 6-year-old male patient was referred from the medical rehabilitation department to the Oral and Maxillofacial Surgery Department at Hasan Sadikin Hospital, Bandung, with the chief complaint of speech difficulties, especially in the pronunciation of the letters "t" and "n". Based on the patient's medical and family history, no other abnormalities were found. A thick, short frenulum and restricted tongue mobility, especially when the tongue's upward movement met the anterior palate, were discovered during an intraoral examination (Fig. 8). The patient was diagnosed as having a differential diagnosis of class II moderate *ankyloglossia* according to Kotlow's classification. Treatment plan of frenotomy was planned to release the lingual frenulum attachment with the purpose of removing resistance from the lingual frenulum of the tongue (Figure 9). The patient was referred for postoperative speech therapy a week after the operation, when it was confirmed that the tongue movement was normal. The treatment showed a good prognosis.

Figure 8. Preoperative view of *ankyloglossia*

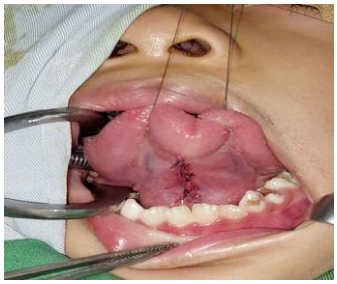

Figure 9. Suturing post frenotomy

Figure 10. Postoperative day-7 showed no restriction movement of tongue to superior direction

Operation Techniques, frenectomy and frenotomy[13] are two surgical treatments that can be used to treat *ankyloglossia*. In the course of a surgical frenectomy, local anesthetic is administered to the lingual region, and the tip of the tongue is sutured with 2/0 silk in order to facilitate elevation of the tongue by the use of a hemostat that is clipped to the middle of the lingual frenulum. The incision is made using a blade size no.15 while keeping it in contact with the upper surface of the hemostat, then proceeded to the frenulum that is located below the hemostat.

The frenulum will leave a "diamond shaped wound" after being incised, and a dissection is performed using the tip of a tissue scissor on the edge of the frenulum with the goal of releasing the fibrotic tissue around the lingual frenulum muscle. Suturing is done by using simple interrupted techniques and 4/0 vicryl (as in Figure 11).

Whereas in the frenotomy surgery method after anesthesia is given to the lingual area, suturing using 2/0 silk thread is performed on the tip of the tongue to help elevate the tongue during a frenectomy procedure. The lingual frenulum incision is made with surgical scissors in the lingual frenulum section with the goal of opening the mucosal tissue in that area. Following blunt dissection with the ash 49 instrument to free fibrous tissue in the lingual frenulum area, additional incisions from the lateral side of the lingual frenulum are made using tissue scissors to further free the lingual frenulum. Suturing the lingual mucosa with 4/0 vicryl thread can be done once there is no resistance from the lingual frenulum (as in Figure 12).

After the frenectomy procedure is completed, the patient is asked to do a follow up visit on the 7th postoperative day. Furthermore, patients are advised to be referred to speech therapy (such as in case report 2 and 3), particularly if they come with complaints of speech disturbances, in order to improve the quality of their speech.

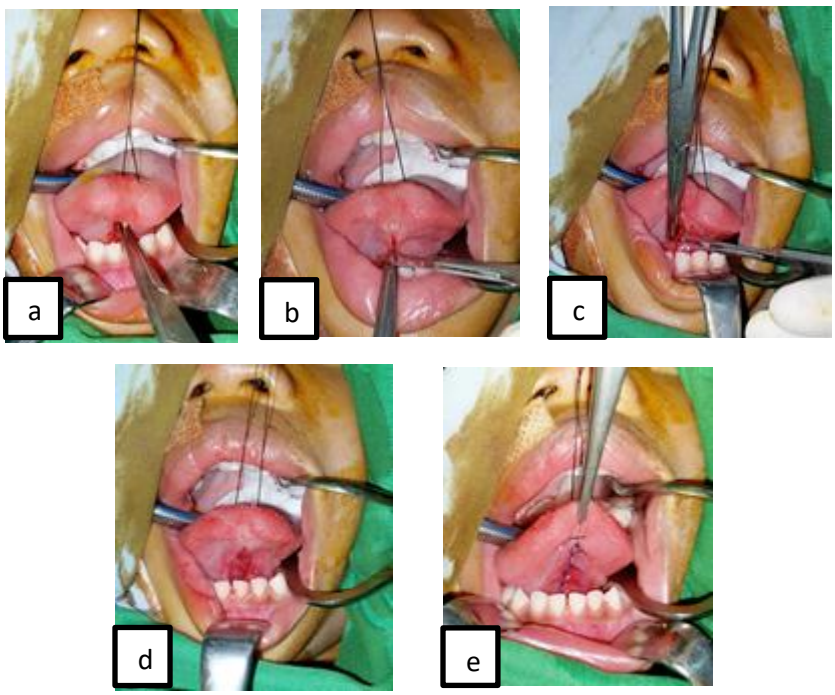

Figure 11. Frenectomy Technique (a) Application of straight hemostat at lingual frenum, (b) The scalpel is always in close contact with the upper surface of the hemostat, (c) The procedure is similar involving the incision at portion beneath the hemostat (d) surgical field after incision with diamond shaped, (e) suturing with vicryl 4/0

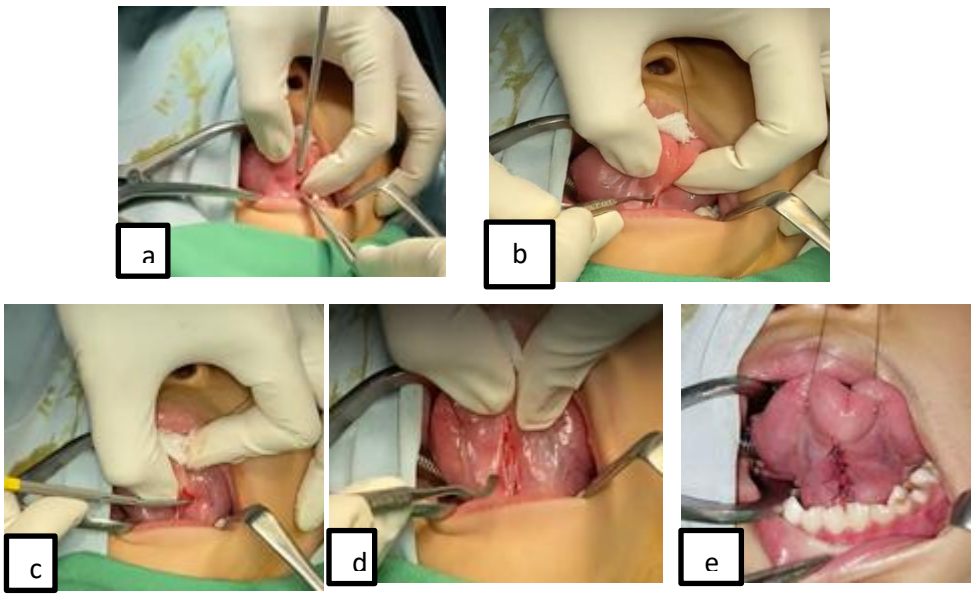

Figure 12. Frenotomy Technique (a) Incision at lingual frenum with iris scissors, (b) Dissection of frenum by ash 49 instrument, (c) Similar incision done at the lateral lingual frenum with iris scissors (d) surgical field after incision (e) suturing with vicryl 4/0.

DISCUSSION

The tongue originates from the first, second, and third pharyngeal arches, which begin to form in the uterus during the 4th month of pregnancy. *Ankyloglossia* is a rare congenital disorder caused by cellular degeneration failure, resulting in a long anchorage between the tongue and the floor of the mouth. [11] Although most patients with *ankyloglossia* do not have other congenital abnormalities, *ankyloglossia* may be associated with a number of syndromes, including Beckwith-Wiedemann syndrome, Van der Woude syndrome, Ehler Danlos syndrome, and others.[13]

Until now there is still a debate about the most optimal diagnostic criteria and therapy of *ankyloglossia*. According to Cuetas et al[14] the diagnosis of *ankyloglossia* according to the anatomical criteria (inspection and palpation of the lingual frenulum) and functional criteria (lifting, protrusion and lateralization of the tongue). Some experts think that ankyloglossia will disappear spontaneously with growth, or in some cases mild *ankyloglossia* children will learn how to compensate to increase the movement of the tongue. [3,13]

In children with moderate to severe *ankyloglossia*, there are a number of issues that can develop if they are not treated. The first issue can result in malnutrition due to disturbances in the movement of the tongue during breastfeeding in infant ages. Other issues include speech and articulation problems in toddler ages. In addition, open bites can happen due to the tongue's abnormal movement, which prevents it from moving towards the palate during swallowing. Some growth disorders such as mandibular prognathism and maxillary hypoplasia have also been reported as a result of this abnormality due to low position of the tongue and pressure from the tongue towards the front and bottom part of the mouth .[10,15,16]

In some cases of *ankyloglossia*, surgical intervention therapy may be beneficial. In a study conducted by *Messner* et al, [13] it was revealed that more than 75% of patients showed improvement in speech articulation based on assessments performed by postoperative speech therapy compared to patients who did not undergo surgery. Clinical guide to ankyloglossia therapy is highly necessary in making decisions of when to do surgery in cases of *ankyloglossia*.

Based on Kotlow's classification criteria, we found several clinical disorders that needed surgical treatment in this study.

These conditions include the formation of a concavity at the tip of the tongue when making protrusive movements, the tongue's inability to traverse the upper and lower lips, the restriction of tongue movement by the frenulum during breastfeeding in infant patients, and the restriction of tongue movement by the frenulum that makes speaking challenging, especially in pediatric patients and the frenulum that inhibit the swallowing process. Patients with the aforementioned disorders can be categorized as having Class III-IV Kotlow's conditions, when surgical therapy is advised as a treatment option.

The first and second cases in this article are categorized according to Kotlow's classification system into class IV, whereas the third case is categorized into class III. According to Susanto et al[17], surgical management is indicated; if the tongue places an excessive force on the anterior mandibular teeth; if the child has difficulty in speaking due to limited tongue movement; and if the defective frenulum prevents the baby from sticking to the mother's nipple during breastfeeding. Therefore, surgical treatment was chosen as a first line treatment in this case report.[13-15.]

There are a variety of surgical techniques that can be used to treat this particular case of *ankyloglossia*. In this case report, we used two surgical techniques depending on the severity of the *ankyloglossia*. In the first and second cases, the "diamond" frenectomy was performed using a hemostat, while in the third case, the surgical technique was performed using the frenotomy method by making a small incision in the frenulum and then blunt dissection of the frenulum. Some of the advantages and disadvantages of each surgical technique had been stated in several previous case studies, in which the technique of using a hemostat has advantages such as predictable operating results and low recurrence rates, while the disadvantage of this technique is that the procedure is quite invasive so it has a higher risk of bleeding.[13, 18, 19]

In contrast to the second surgical technique, some surgeons may choose for a simpler, less intrusive operation, albeit this technique has drawbacks due to a higher recurrence rate. Additionally, a number of other techniques are chosen to decrease the risk of bleeding during surgery and hasten the healing of postoperative wounds, such as the use of a diode laser as an alternative to traditional techniques using a scalpel. However, the disadvantage of the diode laser technique is that it is quite expensive and require the use of safety precautions during the procedure for the patient, operator and assistant. [20-,23]

There are several criteria that can be used to determine the success rate of an *ankyloglossia* therapy, such as the absence of complications from surgery (postoperative bleeding and infection), increased activity during breastfeeding (by observing an increase in body weight), and increased tongue protrusion that can exceed the upper and lower lips without pain or resistance in the frenulum.[3,23]

In this case study, the patient showed successful treatment based on the above criteria during the follow up visit on the 7th day. In the first and second cases, patients showed improvement in breastfeeding and swallowing after treatment, while in speech impairment complaints in the second and third cases with the improvement in tongue movement, it is expected that the patient will have a better pronunciation.

Moreover, the addition of speech therapy consultation is crucial for patients whose primary complaints are speech disorders after surgery. It aims to restore the tongue's movement to its correct position in cases where the patient's condition has grown accustomed to or adapted to incorrect movements when the patient speaks,[14,24] so that in these patients, the success rate can only be assessed after the patient has undergone postoperative speech therapy training.

However, there are some limitations of this case report. First, we don't follow up closely our patient, regarding the first case report, our success treatment only according to mother's statement not the BMI of the patient after the treatment,

furthermore regarding the speech limitation we don't assess the speech result of the patient after undergoing the speech therapy. Second, in accordance with the advance technology such as the laser therapy, we should compare the treatment of the laser therapy with the surgical treatment as we done above for the future cases.

CONCLUSION

Ankyloglossia is a congenital disorder that affects infants and children. Even though the disorder is a simple one, accurate information about clinical guidelines for the patient's parents regarding the timing, indications, and benefits of performing *ankyloglossia* surgery is required. Several clinical criteria were provided in determining the diagnosis and guiding therapy in cases of *ankyloglossia* in this case report. Implication of this case report is encouraging good improvement of the function of feeding and speaking. Accordingly, we highly recommend that all dentists or pediatric dentists use our clinical criteria and guiding therapy when discovering patients with these conditions.

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