

## Management of ankyloglossia in 2 years old male patient with epilepsy and global developmental delay

Kirana Maha Dewi <sup>1</sup>, Fami Widya Pangestika <sup>1</sup>, Lely Indriasari <sup>2</sup> and Soegeng Wahlujo <sup>1,\*</sup>

<sup>1</sup> Department of Pediatric Dentistry, Faculty of Dental Medicine, Universitas Airlangga, Surabaya, Indonesia.

<sup>2</sup> Dr. Saiful Anwar Regional General Hospital, Malang, Jawa Timur, Indonesia.

World Journal of Advanced Research and Reviews, 2024, 24(03), 228–232

Publication history: Received on 22 October 2024; revised on 29 November 2024; accepted on 02 December 2024

Article DOI: <https://doi.org/10.30574/wjarr.2024.24.3.3665>

### Abstract

**Introduction:** Ankyloglossia is defined as a developmental anomaly of the tongue characterized by an abnormally short, thick lingual frenulum resulting in limitation of tongue movement. Ankyloglossia or tongue-tie affects speech, feeding, oral hygiene as well as social environment also.

**Case History:** A 2 years old male patient was brought by his parent by referral of pediatrician to the pediatric dentist policlinic at Dr. Saiful Anwar Regional General Hospital Malang, East Java with the chief complaint difficulties to speech, choked when eating, and excessive drooling. The patient also has epilepsy and global developmental delay and under the control of his pediatrician. According to the clinical examination, the patient had a tongue tie with a thick frenum, limited tongue movements.

**Discussion:** Tongue is an important organ that plays a big role in influencing speech, tooth position, periodontal tissue, swallowing function, as well as nutritional function. The key principle of management of children with developmental concerns is early intervention. In this case the children got referred by his pediatrician to pediatric dentist specialist to give the intervention to the tongue so the child could improve the ability to drink, eating, and also the speech. Several studies have shown tongue-tie release as a safe and successful procedure with little to no complications. The purpose of post operative exercise after surgery is to develop new muscle movements particularly those involving tongue tip elevation and protrusion, inside and outside of the mouth, improved speech and mastication.

**Conclusion:** frenotomy to treat ankyloglossia in 2 years old male patient with epilepsy and global developmental delay shows no post operative complication

**Keywords:** Ankyloglossia; Frenotomy; Global Developmental Delay; Epilepsy; General anesthesia

### 1. Introduction

Tongue is an important organ that plays a big role in influencing speech, tooth position, periodontal tissue, swallowing function, as well as nutritional function. Ankyloglossia is a developmental anomaly of the tongue that results in limited tongue mobility due to an abnormally short, thick, and restricted lingual frenulum. Between 0.2% and 5% of children experience tongue tie, with male predisposition and a male to female ratio of (2.5: 1.0). About 25% to 60% of babies have ankyloglossia, which can lead to nursing issues, such as failure to thrive or even refusal of the breast, which can result in breastfeeding issues, breast rejection, and worries that it will cause failure to thrive. (1), (2), (11)

\* Corresponding author: Soegeng Wahlujo

Speech is critical to child's psychosocial growth since children with speech issues are frequently stigmatised by society. As is well known, if the tongue is constricted, it may be difficult to produce some sounds.(2) (12). Especially in children who experience global developmental delays where children do not reach developmental milestones at the expected age, then the factors that worsen the condition must be eliminated, such as lack of child stimulation, and malnutrition, which in the long term can affect productivity in adulthood.(3)

It becomes critical to perform a lingual frenotomy in order to maximise tongue movement and restore the child's ability to speak and chew the food. The decision to use general anesthesia must be considered based on behavioral modulation, patient's dental needs, as well as their emotional and medical status. A limited and particular group of patients, including extremely young children under the age of three who require extensive dental treatment, those who are not susceptible to treatments under conscious sedation, and those with special needs, necessitate the use of general anaesthesia. Most patients needing general anaesthesia for safe dental treatments are infants, kids, and even teenagers with cognitive or psychological issues.(4)

A seizure is a symptom of a disease that affects people of all races, ages, and genders. The disease is characterised by episodic disruptions of movement, feeling, or consciousness brought on by sudden, synchronous, inappropriate, and excessive electrical discharges that interfere with the brain's normal functioning and the use of conscious sedation and general anesthesia is not contraindicated in patients' epilepsy.(4) During general anaesthesia, the brain may experience transient anoxia, which could cause epileptic seizures, therefore, it must be performed by a skilled anesthesiologist.(4)

This article was aimed to report a case of ankyloglossia of 2 Years Old Male patient with Epilepsy and Global Developmental Delay with the chief complaint of difficulty in moving his tongue which causes speech difficulties, choked when eating, and excessive drooling.

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## 2. Case History

A 2 years old male patient was brought by his parent by referral of pediatrician to the pediatric dentist polyclinic at Dr. Saiful Anwar Regional General Hospital Malang, East Java with the chief complaint difficulty in speaking, choking when eating, difficulty in drinking and excessive drooling. The patient also has epilepsy, and the child is receiving occupational and speech therapy under the supervision of his paediatrician. The child has been diagnosed with both epilepsy and global developmental delay. According to the clinical examination, the patient had a tongue tie with a thick frenum and limited tongue movements.

Extraoral examination showed symmetrical face with no lymphadenopathy. On intraoral examination, was found tongue tie grade 2 based on Kotlow classification (figure 1). The parents of the patient wish for treatment on their child's tongue to prevent further difficulties during feeding and speech. Based on the Frankl behavioral rating scale, the patient showed a rating scale of 1, which is definitely negative(5). The patient's epileptic condition makes this worse. His muscular tone is thought to make the frenotomy process more difficult. Because of these conditions, it was decided to undergo comprehensive dental treatment under general anesthesia.



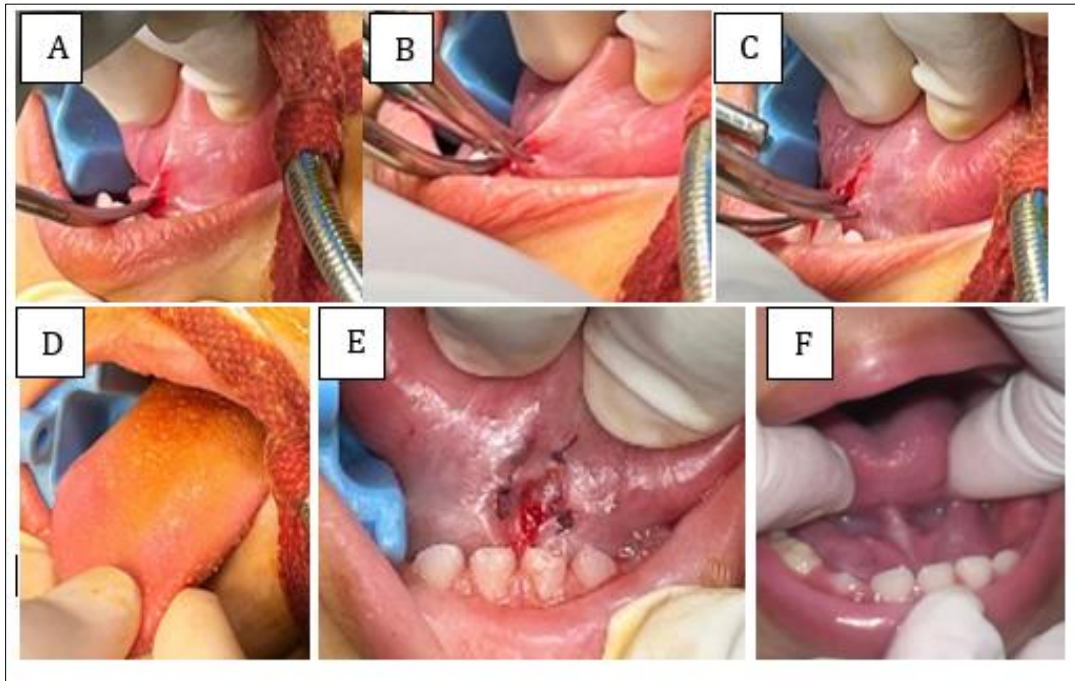
**Figure 1** Intraoral Examination

The General Anesthesia procedure was carried out after examination and approval by a pediatrician and anesthesiologist. 1 week before, a laboratory examination was also carried out including a complete blood cell examination, thorax examination and all examinations showed normal condition. Before the procedure, patient was

instructed to fast for 8 hours. In this case the patient was not given prophylactic antibiotics. General anesthesia is performed with an endotracheal tube through the mouth.

After the patient was anesthetized, frenotomy was performed with initial aseptic steps, followed by fixation of the upper and lower lingual frenulum (figure A), then cutting with surgical scissors (figure B, C), release the area of the lingual frenulum (figure D). Subsequently, 3 sutures were applied using vicryl 4.0 absorbable silk (figure E). The tongue appears to be able to protrude. (figure D)

After all procedures has been done, the patient's parents were given instructions, including avoiding hot or warm foods and drinks, refraining from playing with scars, instructions for maintaining oral hygiene, scheduling routine follow-up visits, and performing periodic tongue massages.



**Figure A** Fixation of the lingual frenulum. **Figure B, C** : cutting Lingual frenulum. **Figure D** : Visible protrusion of the tongue, **Figures E** : 3 sutures, **Figures E** : healing after 7 days follow up

### 3. Discussion

Ankyloglossia is a congenital anomaly characterized by the attachment of the tongue to the floor of the mouth.<sup>1</sup> It occurs due to failure in cellular degeneration. In most of the patients it is asymptomatic, may spontaneously resolve, or affected individuals may learn to compensate for reduced lingual mobility, or may lead to various functional abnormalities that include abnormal speech, malocclusion, and inability to swallow the food which could entail difficulty in normal life activity of an individual. (6) (13)

Based on the distance of the insertion of the lingual frenulum to the tip of the tongue, tongue-tie is classified from class I to class IV according to Kotlow's assessment, Class I: Mild ankyloglossia (12 to 16 mm) Class II: Moderate ankyloglossia (8 to 11 mm) Class III: Severe ankyloglossia (3 to 7 mm) Class IV: Complete ankyloglossia (Less than 3 mm). (7)

The four domains or streams of development are, (I) motor development— gross motor and fine motor, (II) speech and language development—expressive and receptive, (III) social and emotional, and (IV) cognitive. The key principle of management of children with developmental concerns is early intervention. A more comprehensive evaluation is guided by findings on developmental surveillance and screening. Specific clinical psychological or neuropsychological evaluation and psychological testing should be performed by clinical psychologists with appropriate expertise and experience in the evaluation of children with atypical development. Based on findings on clinical history and physical examination further medical evaluation may be indicated. Such evaluation may include neuroimaging, electroencephalography, tests for genetic disorders, and specific laboratory tests for inborn errors of metabolism and should be considered in consultation with appropriate medical specialists. In this case the children got referred by his

pediatrician to pediatric dentist specialist to give the intervention to the tongue so the child could improve the ability to drink, eating, and also the speech. (8)

Several studies have shown tongue-tie release as a safe and successful procedure with little to no complications. (9) According to Patel et al., compared to lasers, healing was better in cases treated using conventional Scalpel after the seventh day and one month postoperatively. Messner et al., in their study, reported that more than 75% of patients had demonstrable improvements in speech articulation postoperatively, as judged by a speech pathologist, compared to the preoperative states.

The purpose of post operative exercise after surgery is to develop new muscle movements particularly those involving tongue tip elevation and protrusion, inside and outside of the mouth, improved speech and mastication. It also increases self aesthetic awareness of the full range of movements the tongue and lips can perform. (10) (14)

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#### 4. Conclusion

Optimal management of a tongue tie including timely surgical intervention and appropriate technique, followed by speech therapy when necessary, has the capacity to provide good outcomes. Correction of ankyloglossia at an early age reduces the risk of complications, especially in this case the child has global developmental delays and epilepsy, frenotomy shows no post operative complication.

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#### Compliance with ethical standards

##### *Acknowledgments*

The authors thank the reviewers for their insightful suggestions.

##### *Disclosure of conflict of interest*

The authors declare that there is no conflict of interest regarding the publication of this document.

##### *Statement of informed consent*

Informed consent was obtained from patient included in the study.

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#### References

- [1] Patel S, Gupta S, Isha S, Tripathi P. Relieving the Tongue Tie-A Case Report. International Journal of Science and Research [Internet]. 2023; Available from: <https://www.researchgate.net/publication/372477296>
- [2] Fakhriani R, Robby Amsriza F. Thermal welding frenotomy on neglected tongue tie: a case report. 2023;23(02). Available from: <https://doi.org/10.24815/jks.v23i02.31560305www.jurnal.usk.ac.id/jks>
- [3] Gil JDC, Ewerling F, Ferreira LZ, Barros AJD. Early childhood suspected developmental delay in 63 low-and middle-income countries: Large within-and between-country inequalities documented using national health surveys. J Glob Health. 2020 Jun 1;10(1).
- [4] SVSG N, Degala S. Dental concerns and management of children with Epilepsy: An overview. Dental, Oral and Maxillofacial Research. 2020;6(4).
- [5] Riba H, Al-Zahrani S, Al-Buqmi N, Al-Jundi A. A Review of Behavior Evaluation Scales in Pediatric Dentistry and Suggested Modification to the Frankl Scale Cronicon A Review of Behavior Evaluation Scales in Pediatric Dentistry and Suggested Modification to the Frankl Scale [Internet]. Available from: <https://www.researchgate.net/publication/322202962>
- [6] Gyawali, N., Sundas, S., Sah, B. K., & Dhakal, N. (2021). Conservative Management of Ankyloglossia by Diode Laser: A Case Report. Journal of Nepalese Association of Pediatric Dentistry, 2(1), 28-31.
- [7] Arjmandzadeh E, Yousefi J. Cronicon EC PAEDIATRICALS EC PAEDIATRICALS Case Report Tongue Tie: A Case with Complete Ankyloglossia.
- [8] Brown KA, Parikh S, Patel DR. Understanding basic concepts of developmental diagnosis in children. Vol. 9, Translational Pediatrics. AME Publishing Company; 2020. p. S9–22.

- [9] Solis-Pazmino P, Kim GS, Lincango-Naranjo E, Prokop L, Ponce OJ, Truong MT. Major complications after tongue-tie release: A case report and systematic review. Vol. 138, *International Journal of Pediatric Otorhinolaryngology*. Elsevier Ireland Ltd; 2020.
- [10] Saskianti T, Aprillia Z, Moeharyono Puteri M, Aulia Az Zahra A, Tedjosongko U. Lingual Frenectomy as Treatment of Ankyloglossia in Children: A case report. Vol. 1, *Indonesian Journal of Dental Medicine*. 2018.
- [11] Messner AH, Walsh J, Rosenfeld RM, Schwartz SR, Ishman SL, Baldassari C, et al. Clinical Consensus Statement: Ankyloglossia in Children. *Otolaryngol Neck Surg*. 2020; 162(5):597–611
- [12] Wright JE J. Tongue-tie. *Pediatric Child Health* 1995; 31: 276-278
- [13] Dr Anand Kishore, Dr Vivek Srivastava, Dr Ashish Mahendra. 2014. Ankyloglossia or
- [14] tongue tie- A case report. *Journal of Dental and Medical Sciences*. Volume 13, Issue 7 Ver. I (July. 2014), PP 52-54