Observational Study

Medicine

OPEN

Evaluation of tongue mobility following release operation of ankyloglossia with genioglossus myotomy

Ji Seon Choi, MD[a], Ji Hwan Park, MD[b], Min Cheol Kim, MD[b], Jin Soo Lim, MD, PhD[b], Hyung-Sup Shim, MD, PhD[b,*]

Abstract

Ankyloglossia is a congenital anomaly characterized by a short lingual frenulum and severe form needs release surgery. Our goal in this study was to confirm the long-term safety and efficacy of Z-plasty with genioglossus myotomy compared with traditional Z-plasty alone and to develop a proper measurement tool to validate the postoperative results of release operations for ankyloglossia. Patients were divided in 2 groups, release with Z-plasty only group (group 1) and Z-plasty combined with genioglossus myotomy group (group 2) In group 2, the release of central longitudinal fiber of genioglossus muscle was added in the conventional Z-plasty operative procedure. To analyze the results of the surgery, we developed an objective assessment tool which is the direct length from the innermost point of the floor of mouth to the tip of the tongue, preoperatively and postoperatively, which is called "functional tongue length." A total of 101 patients with ankyloglossia ranging in age from 36 months to 8 years underwent release operation. Although there was no significant difference in terms of postoperative measurements between groups in Kotlow class II, group 2 patients in Class III and IV showed greater postoperative functional tongue length. Also, there was no significant complication requiring secondary surgery. Our study demonstrated that adding genioglossus myotomy to a simple Z-plasty is a safe and effective method for improving the tongue mobility required to make lingual sounds, especially in moderate to severe form of ankyloglossia, along with suggestion of a new measurement tool, which can objectively assess tongue mobility with possibility for universal utilization in ankyloglossia release operation.

Keywords: ankyloglossia, genioglossus myotomy, release with Z-plasty, speech problem, tongue-tie

1. Introduction

Ankyloglossia, or tongue-tie, is a congenital condition characterized by a short lingual frenulum. Because diagnosis criteria have not been unified and various classification systems have been applied, incidence varies from 0.02% to 10.7% depending on the study. Patients exhibit various clinical manifestations depending on the degree of tongue mobility. The patient can be asymptomatic or have difficulties in breastfeeding, speech articulation, and mechanical tasks such as licking the lips, kissing, and oral cavity cleaning.[1–5] It is also reported that ankyloglossia can affect maxillofacial development.[6]

Frenotomy (simple release of the frenulum) and frenuloplasty (release with repair) are the most common surgical procedures, using various techniques such as myotomy and Z-plasty.[7–10]

We previously reported a modified method for ankyloglossia correction, Z-plasty combined with genioglossus myotomy. In general, patients with ankyloglossia have not only a short and tight frenulum but also contracture of the central fiber of the genioglossus muscle. Thus, better clinical results were obtained by releasing the contracted portion of the muscle as well as mucosal lengthening using Z-plasty than by offering only simple release.[9] However, because we had no objective variable with which to assess the effectiveness of the method, no significant associations were obtained.

Our goal in this study was to confirm the long-term safety and efficacy of Z-plasty with genioglossus myotomy compared with traditional Z-plasty alone and to develop a proper measurement tool to validate the postoperative results of release operations for ankyloglossia.

2. Methods

This study was approved by the Institutional Review Board of the Catholic University of Korea. All data were analyzed anonymously and according to the principles in the 1975 Declaration of Helsinki, revised in 2008.

This study was a retrospective cohort study. All patients who were diagnosed with ankyloglossia Class II–IV (Kotlow

This work was supported by the St. Vincent's Hospital, Research Institute of Medical Science (5-2021-B0001-00278).

The authors have no conflicts of interest to disclose.

The datasets generated during and/or analyzed during the current study are available from the corresponding author on reasonable request.

[a] Department of Plastic and Reconstructive Surgery, Eunpyeong St. Mary's Hospital, College of Medicine, The Catholic University of Korea, Seoul, Korea, [b] Department of Plastic and Reconstructive Surgery, St. Vincent's Hospital, College of Medicine, The Catholic University of Korea, Suwon, Korea.

** Correspondence: Hyung-Sup Shim, MD, PhD, Department of Plastic and Reconstructive Surgery, St. Vincent's Hospital, College of Medicine, The Catholic University of Korea, 93 joongbu-daero, Suwon, Korea (e-mail: sharpshim@catholic.ac.kr).*

How to cite this article: Choi JS, Park JH, Kim MC, Lim JS, Shim H-S. Evaluation of tongue mobility following release operation of ankyloglossia with genioglossus myotomy. Medicine 2022;101:32(e29648).

Received: 23 March 2021 / Received in final form: 22 April 2022 / Accepted: 9 May 2022

http://dx.doi.org/10.1097/MD.0000000000029648

classification system) and received release surgery in our institute between January 2012 and December 2018 were identified. Patients younger than 3 were excluded because cooperation is difficult to attain during the evaluation at the time of outpatient follow-up. Also, patients with a past history of tongue trauma, impaired tongue motion due to brain injury, or a previous release operation that required revisional surgery were excluded from the study.

Four surgeons were involved in this study, and they each used their own preferred surgical technique: Z-plasty with or without genioglossus myotomy.

In release surgery with Z-plasty without genioglossus myotomy (group 1), under general anesthesia, the tip of the tongue was pulled in the cephalic direction using a traction suture, and the frenulum was released close to the tongue base using a scalpel. Then a mucosal Z-plasty flap was designed, elevated, and transposed to prevent mucosal lengthening and contracture. The wound was closed with a 5-0 Vicryl suture.

In release surgery with Z-plasty and genioglossus myotomy (group 2), release of the central longitudinal fiber of the genioglossus muscle was added to the operative procedure just described. The genioglossus myotomy is performed before the mucosal Z-plasty while the contracted longitudinal fiber was visible to prevent injury to the adjacent normal muscle fibers. After myotomy, the acquired mobility was checked with gentle tongue tip traction (Fig. 1).

To analyze the results of the surgeries, we developed an objective assessment tool that we call "functional tongue length" using the difference in direct distance from the innermost point of the floor of mouth to the tip of the tongue preoperatively and postoperatively. This length was measured in the operating room before and after the procedure while the patients were under general anesthesia. During the measurement, the tip of the tongue was gently projected in an upward direction to simulate the tongue motion used in producing lingual sounds (Fig. 2).

The difference between the pre- and postmeasurements was recorded as a postoperatively increased ratio compared with preoperative length to account for individual differences in total tongue length. All patients were observed in the outpatient department at least 6 months postoperatively, and postoperative complications (hematoma, infection, postoperative wound disruption, and scar contracture) were evaluated. The functional tongue length was also checked at each follow-up visit and compared with the immediate postoperative measurement to evaluate postoperative scar contracture over at least 6 months.

Comparisons between the 2 groups were performed using the Chi-square test and Fisher exact test. A $P < .05$ was considered statistically significant.

3. Results

We identified 101 patients with ankyloglossia (30 females and 71 males) ranging in age from 36 months to 8 years (mean age 4.01 years). A total of 43 patients received conventional release with Z-plasty (group 1), and 58 patients (group 2) received additional genioglossus myotomy. The mean age of group 1 was 4.04 ± 0.85 years (range, 36 months to 8 years), and that of group 2 was 4.13 ± 0.59 years (range 37 months to 8 years). In group 1, 16 patients (37%), 17 patients (40%), and 10 patients (23%) had class II, III, and IV ankyloglossia, respectively, and in group 2, those numbers were 17 patients (29%), 19 patients (33%), and 22 patients (38%), respectively. The mean age and proportion of sex in each group did not differ significantly ($P = .44$) (Table 1).

The mean preoperative functional tongue lengths in groups I and II were 24.58 ± 3.44 and 23.67 ± 5.10 mm, respectively, which was not a significant difference. The preoperative to postoperative change in functional tongue length was calculated for each class in both groups. The mean differences were 13.44 ± 2.10 mm for class II, 16.24 ± 3.51 mm for class III, and 21.4 ± 4.71 mm for class IV in group 1 and 13.64 ± 1.90 mm for class II, 22.47 ± 3.09 mm for class III, and 25.72 ± 4.19 mm for class IV in group 2. The difference of measurement was significantly greater in group 2 with class III and IV ($P = .037$, .041, respectively) (Table 2).

Hematoma was found in 2 patients (4.7%) in group 1 and 3 patients (5.2%) in group 2, but no case required revisional surgery, and all cases spontaneously regressed. Postoperative partial wound disruption was found in 2 patients (4.7%) in group I and 2 patients (3.4%) in group II, but all cases healed without additional intervention. No major wound disruptions, infections, or scar contracture occurred.

There was no recurrence in the long-term follow-up period due to contracture when the length was measured at the outpatient clinic. In addition, no cases of reduced length compared with the immediate postoperative result were found.

4. Discussion

Ankyloglossia is sometimes asymptomatic, but can be associated with various functional limitations, including breastfeeding difficulties in infants, speech articulation problems, and mechanical problems such as an inability to lick the lips, kiss, or clean the oral cavity.[1–3] Because most studies have focused on breastfeeding difficulties in infants,[11] several studies have reported the effects of ankyloglossia on breastfeeding difficulties.[12,13] However, there is much controversy about the effect of ankyloglossia on speech articulation.[2,3,12–15]

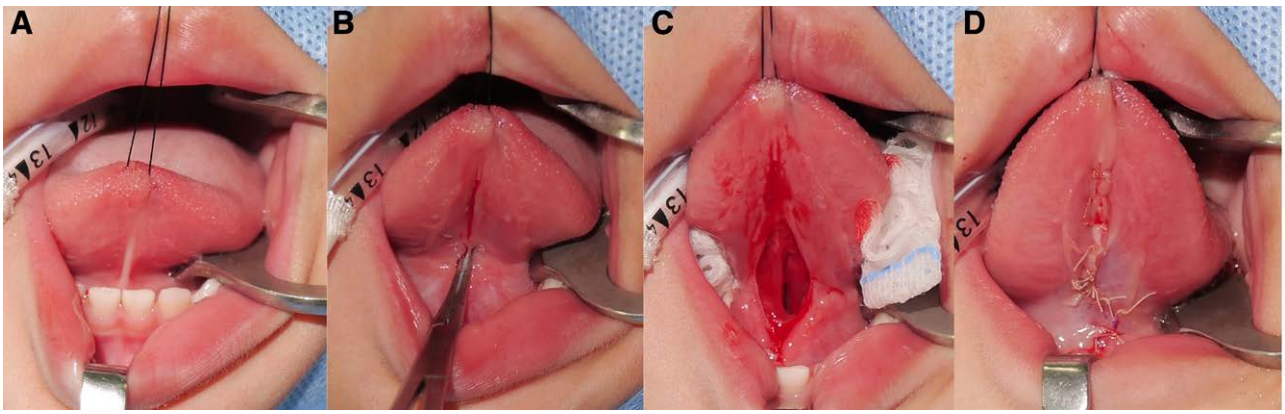

Figure 1. Operative procedure of release surgery with Z-plasty and genioglossus myotomy. (A) Traction suture in the cephalic direction; (B) release of frenulum; (C) release of central longitudinal fiber of the genioglossus muscle; and (D) mucosal Z-plasty and closure.

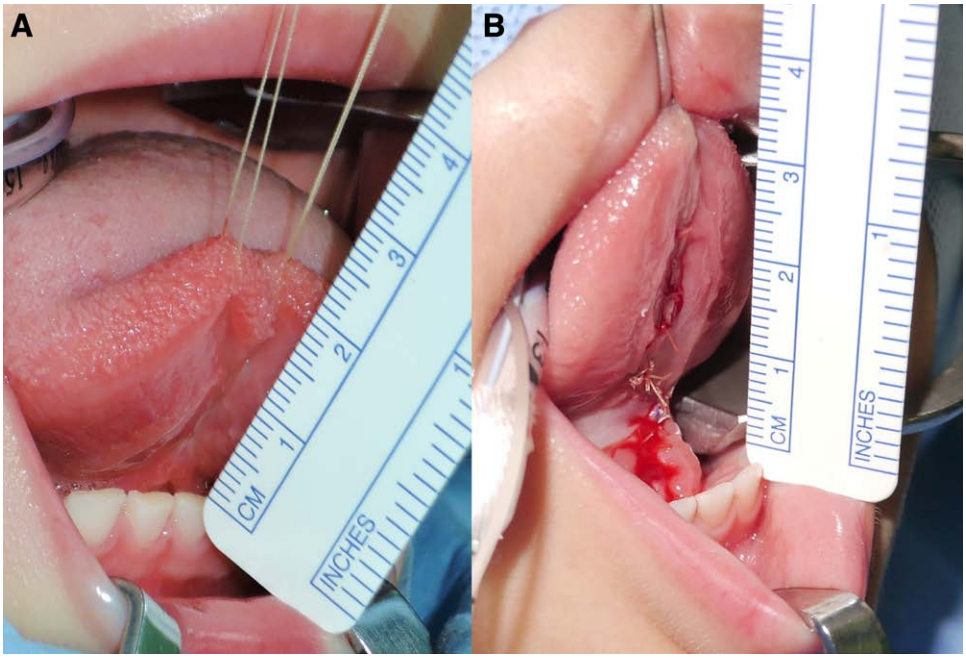

Figure 2. Measurement of the preoperative and postoperative "functional tongue length." (A) Preoperative functional tongue length and (B) Postoperative functional tongue length.

Table 1

Patients demographics.

	Group 1 (n = 43)			Group 2 (n = 58)		
	Class II (n = 16)	Class III (n = 17)	Class IV (n = 10)	Class II (n = 17)	Class III (n = 19)	Class IV (n = 22)
Age (y)	3.88 ± 0.96	4.24 ± 1.30	4.00 ± 1.63	4.18 ± 1.42	4.11 ± 1.10	4.14 ± 1.46
Gender (%)						
Male	56	76	90	76	74	59
Female	44	24	10	24	26	41

Table 2

Preoperative measurement and postoperative differences in each group and classes.

		Group 1 (without myotomy)	Group 2 (with myotomy)	*P* value
Preoperative measurement (mm)		24.58 ± 3.44	23.67 ± 5.10	.39
Difference from postoperative measurement (mm)	Class II	13.44 ± 2.10	13.64 ± 1.90	.57
	Class III	16.24 ± 3.51	22.47 ± 3.09	.037
	Class IV	21.40 ± 4.71	25.72 ± 4.19	.041

Several grading systems have been proposed. One of them categorizes the frenulum into 4 types based on the point of tongue attachment to it (Coryllos system), and another measures the free tongue length from the tip of the tongue to the frenulum attachment (Kotlow system). We used the Kotlow system in our study, and it classifies ankyloglossia into normal, class I (mild), class II (moderate), class III (severe), and class IV (complete). Other grading systems use tongue protrusion distance. Although various classification systems have been applied, none of them have been related to symptomatic severity,[6,7] and no consensus about the indication and optimal timing for surgical release has been established.

Generally, surgical release has been performed on infants with breastfeeding difficulties, young children with articulation problems, and older children and adults with mechanical and social problems.[2] However, because classification and grading systems are not commonly defined, it is difficult to determine definite indications for surgery and to evaluate the postoperative results.

Because most reports on ankyloglossia have focused on breastfeeding difficulties in infants, associations between breastfeeding difficulties and ankyloglossia have been confirmed. However, the relationship between ankyloglossia and speech articulation remains controversial.[2,14,15] Speech problems, especially articulation, have been considered a hallmark manifestation of symptomatic ankyloglossia because it has been thought that restrictions in tongue movement cause difficulty in pronouncing lingual sounds such as t, d, z, s, th, n, and l. Pronunciation of lingual-alveolar sounds (particularly l) and interdental sounds (particularly th) seems to cause most problems because the tongue needs maximal elevation and protrusion to produce those consonants.[2,3,14–16]

Fletcher and Meldrum found a highly significant increase in the number of articulation errors in their limited lingual

freedom group, which provided strong evidence for the relationship between tongue mobility and speech articulation.[2,17] Our experience in our institution also suggests that ankyloglossia might not cause speech development delays, but it does interfere in speech articulation in certain individuals.

Another debate surrounds the optimal timing of surgery.[3] Some investigators prefer to perform prophylactic release before speech articulation difficulties occur because the surgery is minor. If patients have a significant potential for speech difficulties, performing prophylactic surgery regardless of age will reduce or eliminate the need for speech therapy and social embarrassment. Other investigators recommend withholding surgery until a patient manifests a speech or mechanical problem because the frenulum could recede on its own during the first years of a child's growth and development.[2,5]

In the absence of an appropriate tool to evaluate the outcome of surgery, controversy continues regarding the potential association between ankyloglossia and speech articulation, the effectiveness of surgery, and the appropriate timing of surgery to improve articulation. Without a grading system clearly related to symptomatic severity, it is not easy to demonstrate an association between improved pronunciation and surgery even when postoperative results are evaluated using one of the existing grading systems. Furthermore, no high-quality study has compared the effects of various procedures.

In previous publications, the following scales were used to evaluate tongue mobility: the degree of tongue elevation using a maximum interincisal distance; the degree of tongue protrusion, measured as the distance from the tongue tip to the lower dentition; the degree of improvement in pronunciation in a speech pathologist's assessment; and a nonvalidated telephone questionnaire about parental perceptions (subjective assessment by parents).[3,14] Heller et al evaluated surgical outcomes using the distance from the tongue tip to the tongue base measured when the tongue was maximally elevated.[10] Because that indicator is a measure of distance along curved surfaces from the tip to the base, it is inevitably affected by the volume or thickness of the tongue.

In this study, we sought an indicator relevant to the surgical outcome. Thus, to measure the actual mobile tongue length required to produce a lingual sound, we used the distance from innermost point of the floor of mouth to the tip of the tongue, the linear distance from the frenulum attachment to the tongue tip while projecting the tongue tip upward, which has not been used in previous studies. This measurement is an actual indicator of how much the tongue can move, which makes it more suitable than previous measures for identifying the tongue mobility needed for lingual sounds. We call this distance "functional tongue length" because it is relevant to making lingual sounds, and we offer it as a functional measurement tool for use after ankyloglossia surgery.

In our evaluation of the results, we found no statistically significant differences between the surgical groups in class II. However, in classes III and IV, the increased postoperative functional tongue length was significantly greater in group 2, indicating that the genioglossus myotomy improved the actual tongue mobility required to make lingual sounds. The degree of increase was the greatest in class IV, presumably because the contracture of the central longitudinal fiber in the genioglossus muscle is the most severe in class IV. This also confirms that the contracture of the genioglossus muscle affects the severity of ankyloglossia. In addition, we suggest that surgeons can improve their results by using intraoperative myotomy in patients with severe ankyloglossia.

In addition, long-term follow-up was possible because our patient selection process did not include patients too young to evaluate during follow-up. We confirmed that there was no recurrence due to contracture or other scar-related complications, possibly because of the high mobility of the tongue.

The surgical technique used in this study added genioglossus myotomy to the established release operation (conventional Z-plasty). This technique is an easy and simple procedure for conducting myotomy with electrocautery in a portion of the contracted central fiber of the genioglossus muscle. Not only does it carry little risk of complications such as bleeding and hematoma since it is performed via electrocautery but it is also safe because it is performed at a site that does not compromise the function of the tongue. In addition, it does not lengthen the surgical time much because it is simple and does not require any additional dissection or incisions.

Our study has some limitations. This indicator might not be applicable to patients younger than 36 months because cooperation in the outpatient department might be impossible to attain. Comparing follow-up measurements with the immediate postoperative measurement is a crucial point for our measurement to be used as a standard tool. Also, there is a potential risk for measurement errors because the traction force can vary with each measurement. Although we tried to measure the length of the tongue in a fixed orientation to the incisive foramen using gentle traction, errors remain a possibility.

5. Conclusion

Our study demonstrated that adding genioglossus myotomy to a simple Z-plasty is a safe and effective method for improving the tongue mobility required to make lingual sounds. This was confirmed by the new measurement tool we provide, functional tongue length, which is the direct distance from innermost point of the floor of mouth to the tip of the tongue.

Also, we found that patients with severe ankyloglossia showed greater improvement in tongue mobility when their release operation was combined with genioglossus myotomy. Functional tongue length, which can objectively assess tongue mobility, could be useful in evaluating and comparing the results of other release surgery techniques.

Author contributions

Ji Seon Choi, Ji Hwan Park: study design, data processing, writing manuscript.
Min Cheol Kim, Jin Soo Lim, Hyung-Sup Shim: study design, surgery, data analysis.

References

[1] Messner AH, Lalakea ML, Aby J, et al. Ankyloglossia: incidence and associated feeding difficulties. Arch Otorhinolaryngol- Head Neck Surg. 2000;126:36–9.
[2] Lalakea ML, Messner AH. Ankyloglossia: does it matter? Pediatr Clin North Am. 2003;50:381–97.
[3] Webb AN, Hao W, Hong P. The effect of tongue-tie division on breastfeeding and speech articulation: a systematic review. Int J Pediatr Otorhinolaryngol. 2013;77:635–46.
[4] Kupietzky A, Botzer E. Ankyloglossia in the infant and young child: clinical suggestions for diagnosis and management. Pediatr Dent. 2005;27:40–6.
[5] Tsaousoglou P, Topouzelis N, Vouros I, et al. Diagnosis and treatment of ankyloglossia: a narrative review and a report of three cases. Quintessence Int (Berlin, Germany: 1985). 2016;47:523–34.
[6] Yoon AJ, Zaghi S, Ha S, et al. Ankyloglossia as a risk factor for maxillary hypoplasia and soft palate elongation: a functional - morphological study. Orthod Craniofac Res. 2017;20:237–44.
[7] Hong P, Lago D, Seargeant J, et al. Defining ankyloglossia: a case series of anterior and posterior tongue ties. Int J Pediatr Otorhinolaryngol. 2010;74:1003–6.
[8] Junqueira MA, Cunha NN, Costa e Silva LL, et al. Surgical techniques for the treatment of ankyloglossia in children: a case series. J Appl Oral Sci: Revista FOB 2014;22:241–8.
[9] Choi YS, Lim JS, Han KT, et al. Ankyloglossia correction: Z-plasty combined with genioglossus myotomy. J Craniofac Surg. 2011;22:2238–40.
[10] Heller J, Gabbay J, O'Hara C, et al. Improved ankyloglossia correction with four-flap Z-frenuloplasty. Ann Plast Surg. 2005;54:623–8.

[11] Chinnadurai S, Francis DO, Epstein RA, et al. Treatment of ankyloglossia for reasons other than breastfeeding: a systematic review. Pediatrics. 2015;135:e1467–74.
[12] Pransky SM, Lago D, Hong P. Breastfeeding difficulties and oral cavity anomalies: the influence of posterior ankyloglossia and upper-lip ties. Int J Pediatr Otorhinolaryngol. 2015;79:1714–7.
[13] Ghaheri BA, Cole M, Fausel SC, et al. Breastfeeding improvement following tongue-tie and lip-tie release: a prospective cohort study. The Laryngoscope. 2017;127:1217–23.
[14] Suter VG, Bornstein MM. Ankyloglossia: facts and myths in diagnosis and treatment. J Periodontol. 2009;80:1204–19.
[15] Messner AH, Lalakea ML. Ankyloglossia: controversies in management. Int J Pediatr Otorhinolaryngol. 2000;54:123–31.
[16] Khan S, Sharma S, Sharma VK. Ankyloglossia: surgical management and functional rehabilitation of tongue. Indian J Dent Res: Official Pub Indian Soc Dent Res. 2017;28:585–7.
[17] Fletcher SG, Meldrum JR. Lingual function and relative length of the lingual frenulum. J Speech Hear Res. 1968;11:382–90.