

New strategy for the management of oral ranulas: lingual frenectomy

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Abstract

Objective: To explore the role of lingual frenectomy in the treatment of ranulas.

Methods: In this study, we retrospectively reviewed 45 patients with ranulas accompanied by short lingual frenulum, they were randomly assigned to the frenulum correction group (n=24) and the control group (n=21). The following data were obtained, including age, sex, site of ranula, preoperative period of observation, diameter of ranula, duration of follow-up and the results.

Results: Our results demonstrated that the mean age of the patients was 13.6 ± 2.56 years. Among 24 cases treated by frenulum correction, 15 ranulas had a full regression of the lesion after one month. 9 ranulas had no change after frenulum correction. By comparison, in the control group, only 5 cases were disappearance. There was significant difference between two groups ($p < 0.05$).

Conclusion: The findings indicated that increased tongue movement after lingual frenulum surgery is beneficial to the disappearance of ranulas, and lingual frenectomy is an adjuvant approach in the treatment of ranulas adjacent to lingual frenulum.

Keywords: Ranulas, short lingual, frenulum, lingual frenectomy

1. INTRODUCTION

Oral ranulas are pseudocysts without epithelium resulting from the retention or extravasation of saliva from the sublingual gland (Baurmash HD 2003). They typically present as bluish swellings over the floor of the mouth that resemble a frog's abdomen, thus the term "ranula". Intraoral ranulas are the common type, which is restricted in its inferior extension by the mylohyoid muscle.

These ranulas can be treated by various minimally-invasive approaches. Despite of simplicity and convenience, the approaches still have a certain degree of recurrence rate (10%-60%) (McGurk M, et al. 2008; Sandrini FA, et al. 2007). To date, the most appropriate treatment protocols have not yet been established, particularly for pediatric cases.

Reports have demonstrated that mucoceles had the self-limited property of spontaneous disappearance, which is also observed in a few ranulas (Bentley JM, et al. 2003; Mínguez-Martinez I, et al. 2010; Zhao YF, et al. 2005). Recently, we found that ranulas occurred in patients accompanied by short lingual frenulum, surprisingly, we observed its spontaneous disappearance after frenulum corrections. Though the relationship between ranulas and short lingual frenulum remains unclear, we infer that short lingual frenulum may be an unfavorable factor for ranulas disappearance. The increase of tongue movement in speech, eating, chewing, and the biomechanical stimulation may promote the disappearance of ranulas (Hiemae KM; Palmer JB 2003).

To evaluate the role of lingual frenulum surgery in the treatment of ranulas, in this study, we analyzed 45 patients with ranulas accompanied by short lingual frenulum, and observe the outcome of ranulas after frenulum correction.

2. MATERIALS AND METHODS

The protocol for this prospective clinical study was approved by the Independent Ethics Committee of the Ninth People's Hospital of Shanghai Jiao Tong University School of Medicine. From January 2013 to June 2019, 45 patients with ranulas accompanied by short lingual frenulum who visited our out-patient clinic were initially enrolled. The inclusion criterion was the patients with oral ranulas diagnosed by fine needle aspiration cytology. The diameter of ranulas was no more than 10mm, and the distal end is less than 10mm from the

lingual frenum (Fig. 1). The ranulas were caused by mucus retention from a torn duct of Rivinus of LSL 1 (Harrison JD, et al. 2013). All enrolled patients provided written informed consent to enroll in the study. Exclusion criteria included the presence of any of the following: history of head and neck malignancy, history of radiation therapy in the head and neck, inflammation detected by blood tests, and preoperative history of ranula. 45 patients were assigned to the frenum correction group (n=24) and the control group (n=21). Clinical characteristics of the patients were recorded, including age at presentation, sex, site of ranula, preoperative period of observation, diameter of ranula, duration of follow-up. A descriptive analysis was performed for each of the variables, and correlation determined using the chi-square test. Statistical significance was considered for $p < 0.05$.

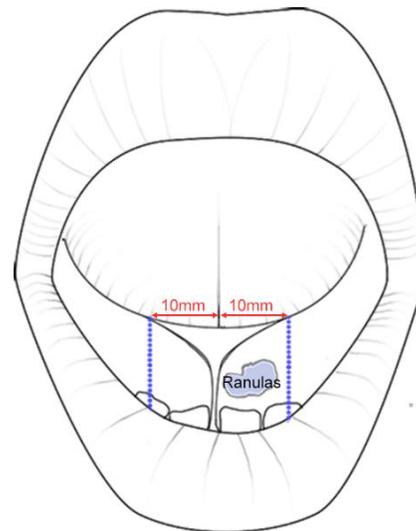


Fig. 1. Schematic model of ranulas accompanied by short lingual frenulum.

2.1. Evaluation of lingual frenulum

All short lingual frenulum was assessed following the previous report (Marchesan IQ 2005). First, the patients opened mouth wide (Position A) or opened mouth wide when the tongue tip touched the incisive papilla (Position B), the doctor examined the distance from the superior the right or left incisive to the inferior the right or left incisive using caliper (Distance A was measured at position A, distance B was measured at position B). Then the percentage (Distance B/Distance A) was calculated. According to different scores, short lingual frenulum was classified into three types: Type I (20-30%), Type II (31-40%), Type III (41-50%).

2.2. Lingual frenectomy

Under local anesthesia, surgery was performed in clinic. It involves cutting the frenulum to free up the tongue and then several stitches. We didn't treat the ranulas with any methods. The aim of the operation was to increase the movement of the tongue. The patients were placed on a liquid diet with regular oral rinses for 7 days, and given prophylactic antibiotics for 3 days.

3. RESULTS

Comparison of the frenum correction group and the control group in Table 1. In this study, 45 patients (male 18, female 27) were included, and the mean age was 13.6 ± 2.56 years. There were 23 cases (51.11%) in the 0-10 years age group, 13 cases (28.88%) in the 10-20 years age group, 6 cases (13.33%) in the 20-40 years age group, 3 cases (8.89%) in the >40 years age group. The mean size of the lesion was 0.68 ± 0.12 cm, 23 patients were <0.5 cm, and 22 patients were 0.5-1.0 cm. Duration of ranulas development in the frenum correction and the control were 8, 10 (<3 months), 11, 6 (3-6 months), 4, 3 (6-10 months), 1, 2 (>10 months) respectively. Among 24 cases treated by frenulum correction, 15 ranulas had a full regression of the lesion within one month (Fig. 2). 9 ranulas had no change after frenulum correction. By comparison, in the control group, only 5 cases were disappearance. According to the chi-square test analyses, there was significant difference between frenulum correction group and the control group ($p < 0.05$).

Table- 1: Comparison of the frenum correction group and the control group

	frenum correction (n=24)		the control (n=21)		
Variables	n (%)		n (%)		<i>P</i>
Gender					
Male	10(41.67%)		8(61.90%)		0.764
Female	15(58.33%)		12(38.10%)		
Age					
0-10	11(45.83%)		12(57.14%)		0.541
11-20	7(29.17%)		6(28.57%)		
20-40	4(16.67%)		2(9.52%)		
>40	2(8.33%)		1(4.76%)		
Side					
Right	14(58.33%)		13(61.90%)		0.818
Left	10(41.67%)		8(38.10%)		
Diameter of ranula (mm)					
< 5	13(54.17%)		10(47.62%)		0.448
5 – 10	11(45.83%)		11(52.38%)		
Type of lingual frenulum (%)					
I	9(37.5%)		7(33.33%)		0.786
II	8(33.33%)		10(47.62%)		
III	7(29.17%)		4(19.05%)		
Duration of ranulas development(months)					
< 3	8(33.33%)		10(47.62%)		0.637
3 – 6	11(45.83%)		6(28.57%)		
6 – 10	4(16.66%)		3(14.29%)		
>10	1(4.17%)		2(9.52%)		

Outcome				
Disappearance	15(62.5%)		5(23.81%)	0.0157
No change	9(37.5%)		16(76.19%)	

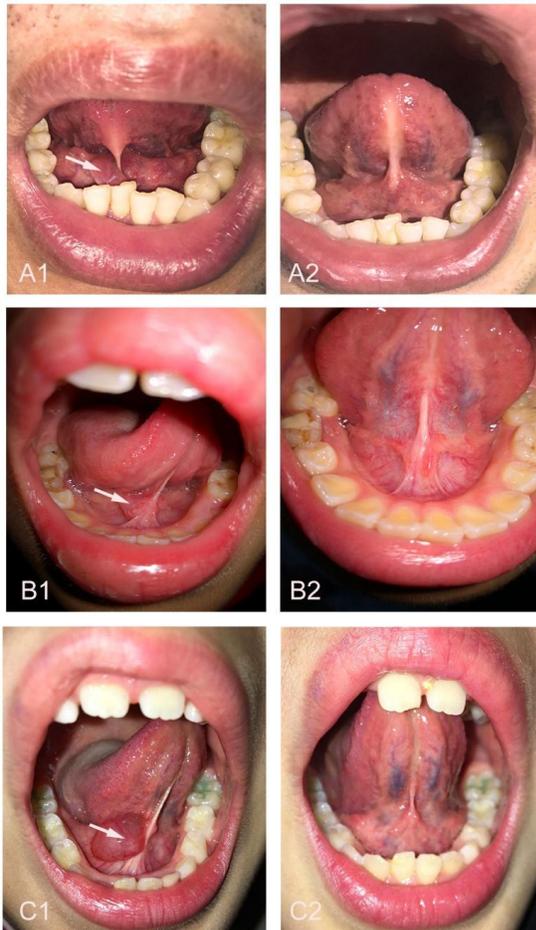


Fig. 2. Patients with ranulas were performed using lingual frenulum correction, and ranulas were collapsed after lingual frenulum correction (A1-2, B1-2, C1-C2).

4. DISCUSSION

In this study, we demonstrate that frenulum correction is an alternative treatment for ranulas, especially for those adjacent to lingual frenulum. Among 24 cases of ranulas treated by lingual frenectomy, 15 ranulas gradually disappeared within one month, and 9 ranulas had no change. In the control group with no frenulum correction,

only 5 ranulas were disappeared. Our results indicated that lingual frenectomy enhanced the movement of tongue and increased mechanical stimulation in oral cavity, which might promote repeated rupture and the disappearance of the ranulas.

The removal of sublingual gland is still considered as the gold standard, however, it also has a high rate of morbidity with risk of

injury to the submandibular duct and lingual nerve(Seo JH, et al. 2010). Attempts have been made to develop conservative therapies that avoided excision of the sublingual gland(Harrison JD 2010). So far, numerous minimally invasive methods have been proposed to treat oral ranulas, but there is still a certain recurrence rate(Goodson AM, et al. 2015). We have previously confirmed that two-incision fistula technique is an effective approach for the treatment of introral ranulas. Also, we preliminarily explored its potential mechanism and the management for recurrence(Wang S, et al. 2016;Zhao Q, et al. 2018).

Recently, studies have reported that ranulas had the ability of self-limiting, it could gradually disappear after repeated rupture of the cyst. Nevertheless, it does not mean that ranula has real self-healing ability, the possible cause of disappearance may be that the fistula formed in ranulas drains the retained cystic fluid(Lee HM, et al. 2006). According to our observation, those ranulas elapsed are usually located on the superficial sublingual gland, it is difficult for those with a slightly deep retention of the sublingual gland. From this point of view, the mechanical stimulation factors in oral cavity could promote repeated rupture of ranulas and led to their disappearance.

It is well known that the tongue is an important mechanical stimulation factor, and it plays an important role in oral mechanics and physiological function(Hiiemae KM;Palmer JB 2003;Napadow VJ, et al. 1999). When tongue is restricted because of short lingual frenulum, oral mechanical stimulation will decrease. In this study, we found that there were 9 cases (Type I), 8 cases (Type II), and 7 cases (Type III) in frenulum operation group. After frenulum correction, the limited tongue is remarkably relieved, which resulted in the increase of the oral mechanical stimulation. The enhanced mechanical stimulation, produced

by chewing, speech and feeding, increased the possibility of mucocoeles spontaneous rupture, as a result, led to the disappearance of oral ranulas(Marchesan IQ 2005).

Studies have shown that the vast majority of ranulas are extravasation cysts that develop as the result of Rivinus duct rupture, so the retained saliva was often located on the superficial sublingual gland and the repeated rupture is common(Harrison JD 2010;McGurk M 2007). Researchers have also observed that most of the cysts did not exist in the oral cavity for more than 24 hours, then rupture in a few hours and left a small white pseudomembrane composed of collapsed squamous epithelium(Jensen JL 1990). In the present study, we performed lingual frenulum correction in 24 patients, and we found that the approach could achieve good results. 15 ranulas were disappeared in one-month post frenulum correction, 9 ranulas had no change. Compared with the operation groups, only 5/21 ranulas were disappeared in the control. There is a significant difference between two groups ($p<0.05$). Unlike previous approaches reported for ranulas, frenulum correction provided with us the new insight in treating ranulas(Harrison JD 2010).

Although frenulum correction may be related with the disappearance of some ranulas, we are not convinced that the occurrence of ranulas is in associated with short lingual frenulum. Nevertheless, before the surgery, the following points need to be emphasized. First, all ranulas were located on the superficial sublingual gland and not the thicker cyst wall type; Second, ranulas less than 10 mm in diameter are suitable for this procedure; Third, the lesions mainly occurred due to a torn duct of Rivinus of LSL 1, and the location was near the lingual frenum(<10mm)(Harrison JD, et al. 2013). Because of limited samples of patients in this study, whether lingual frenulum correction is conducive to promoting the

disappearance of cysts required further investigations. Therefore, our next step is to increase patient sample size and evaluate its long-term outcome.

In conclusion, increased tongue movement after lingual frenulum surgery is beneficial to the disappearance of introral ranulas, and lingual frenectomy is an adjuvant approach in the treatment of ranulas. Our results indicated that we should not overlook the value of lingual frenectomy in the treatment of ranulas.

CONFLICTS OF INTEREST

No potential conflict of interest relevant to this article was reported.

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