



Plunging ranula with lingual nerve tether: Case report and literature review

Krish Suresh^{a,*}, Allen L. Feng^b, Mark A. Varvares^b

^a Northwestern University Feinberg School of Medicine, Chicago, IL, United States of America

^b Department of Otolaryngology, Mass. Eye and Ear Infirmary, Harvard Medical School, Boston, MA, United States of America

ARTICLE INFO

Keywords:

Plunging ranula
Transcervical approach
Lingual nerve

ABSTRACT

Plunging ranulas are most often treated surgically; various surgical approaches may be necessary depending on the unique characteristics of each case. Here, we present the case of a plunging ranula noted on imaging to have a cordlike tether, which was revealed intraoperatively to be the lingual nerve. This case illustrates the importance of preoperative imaging for surgical planning, and when a transcervical approach may be the best choice for plunging ranulas.

1. Introduction

Ranulas are extravasation mucocoeles arising from the sublingual gland (SLG), due to obstruction and rupture of its draining ducts. Ranulas are pseudocysts, with linings composed of granulation and connective tissue, as opposed to epithelium which forms the lining of true cysts such as mucous retention cysts. A plunging ranula (PR) results when the mass herniates through or around the mylohyoid muscle into the submandibular space [1].

Surgery is the mainstay of therapy for PR – however, the method of surgery is less clear. A survey sent out to members of the American Head and Neck Society as part of a 2009 study on ranulas found that of 220 surgeons, 27% favored a transoral approach for PR, 23% a cervical approach, and 49% a combined approach. There was even greater variability regarding the specific procedure [2]. One important consideration in determining the method of surgery is the unique anatomy of each lesion as seen on imaging. Herein, we present the case of a PR seen on imaging to be bifurcated by a cordlike tether, driving our team to perform a transcervical excision of the PR and right submandibular gland (SMG).

2. Case report

A 29-year-old woman presented reporting a “bubble” in the right floor of mouth and an ipsilateral neck mass for two months. She reported that the floor of mouth mass occasionally expressed mucous into the oral cavity. She had never experienced such a lesion before. She denied pain or difficulty with speech, swallowing, or breathing. She also denied prior surgery or trauma to the floor of mouth.

Exam confirmed a fluctuant, nontender mass primarily in the right

floor of mouth crossing midline by 3 cm and extending back to the second molar. In the right neck, there was a 3 cm submandibular mass as well as fullness in the submental region. These lesions were contiguous and together were measured to have a diameter of 7 cm. T2-weighted MRI (Fig. 1) demonstrated a hyperintense, homogenous right floor of mouth mass extending into the submandibular space. There was a cordlike structure wrapped around the mass, bifurcating it into a dumbbell-like shape.

The patient underwent transcervical excision of the PR and right SMG. Intraoperatively, the lingual nerve was identified tethering the ranula (Fig. 2A), conforming it into the dumbbell-like shape noted on imaging. In the course of the dissection, the very distal lingual, glossopharyngeal and hypoglossal nerves were also identified (Fig. 2B). The final pathology report demonstrated an inflammatory pseudocyst, consistent with PR. The patient had no complaints postoperatively; notably, function of the lingual, hypoglossal, and glossopharyngeal nerves was intact.

3. Discussion

While different surgical approaches may be necessary for PR depending on the unique features of each case, the method most consistently supported in the literature is transoral excision of the sublingual gland (SLG). One of the early, often cited studies on this subject reported that excision of the SLG was essential for successful treatment, resulting in the lowest recurrence rate (2%) [3]. More recent studies have corroborated this finding, and also compared transoral and transcervical approaches. Most have advocated for a transoral excision of the SLG, arguing that this achieves similarly low rates of recurrence with lower attendant morbidity [1,2,4–10]. For example, a 2009

* Corresponding author at: Northwestern University Feinberg School of Medicine, 675 N St Clair St, Ste 15-200, Chicago, IL 60611, United States of America.
E-mail address: krish.suresh@northwestern.edu (K. Suresh).

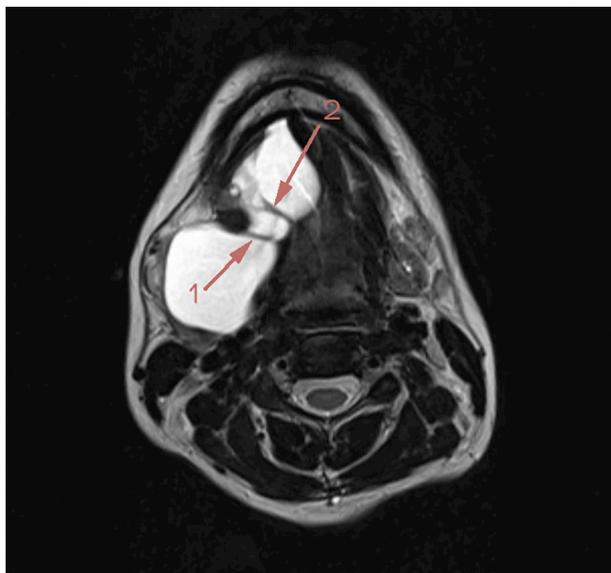


Fig. 1. T2-weighted MRI showing plunging ranula with lingual nerve tether. 1 – mylohyoid posterior edge, 2 – lingual nerve.

literature review reported that transoral excision of SLG + PR (n = 24) and transcervical excision of SLG + PR (n = 16) both resulted in 0% recurrence rates. The review also analyzed their own institution's experience with PR, reporting recurrence in 1/16 transoral cases and 1/10 transcervical cases [2].

Regarding the morbidity associated with these procedures, one potential complication particularly relevant in this case is lingual nerve injury. Several studies have reported lingual nerve injury following surgery for PR, the largest being the aforementioned literature review. The authors reported tongue hypesthesia in 1/3 (33.3%) cases of excision of SMG + PR, and 3/14 (21.4%) cases of excision of SLG + SMG + PR. In contrast, just 1/37 (2.7%) cases of transoral excision of SLG developed tongue hypesthesia [2]. Similarly, several primary series of transoral SLG excision for PR have reported relatively low rates of lingual nerve injury: 2/18 (11.1%) and 0/129 in two series

from China [5,10], 7/82 (8.5%) in a series from Australia [7], 0/21 in a series from New Zealand [6], and 0/22 in a series from the Massachusetts Eye and Ear Infirmary [4].

Despite the relatively low complication rates with transoral approaches for PR, a transcervical approach may still be appropriate in select cases. A 2013 best practice recommendation proposed that a transcervical approach would be appropriate for complex PRs that could not be adequately drained transorally, such as large, loculated pseudocysts or revision cases [11]. They cited a series of 95 PR drained transorally, of which 4 developed infected cervical fluid collections requiring readmission for drainage. Using a slightly different technique, another group of 21 PR was drained transorally with additional placement of a retrograde drain through the neck; this technique eliminated the development of postoperative fluid collections [6]. Transcervical approaches may allow for more thorough evacuation of PR contents. One other point in the best practice recommendation was that a transcervical approach would allow for excision and pathologic examination of a portion of the cyst wall, in order to confirm the diagnosis in uncertain cases [11].

The case presented herein provides important clinical context for deciding when to perform a transcervical approach. Based on the pre-operative imaging shown in Fig. 1, there was significant concern for a neurovascular structure tethering the PR, splaying it into two distinct foci. In addition, concern was raised regarding the significant size of the lesion and involvement of surrounding structures including the lingual nerve and SMG. Given these factors, transcervical excision of SMG + PR was deemed the safest and most definitive treatment for this patient. Intraoperative findings of significant fibrosis of the pseudocyst capsule involving the overlying lingual nerve and close proximity to the glossopharyngeal and hypoglossal nerves validated this pre-operative decision.

4. Conclusion

Most PR can be treated by transoral excision of SLG, however alternative approaches may be necessary depending on the unique characteristics of each case. This case highlights the importance of pre-operative planning based on relevant imaging in the surgical management of plunging ranula.

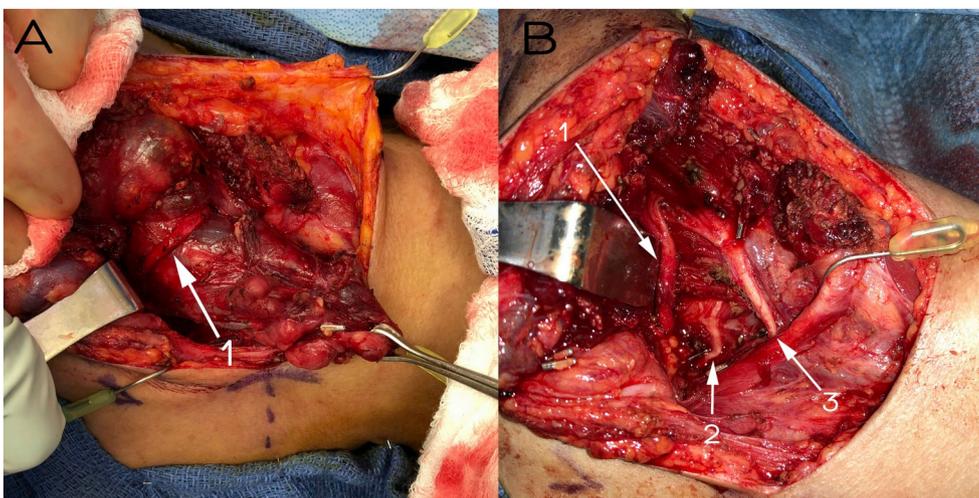


Fig. 2. Intraoperative images before (A) and after (B) ranula excision with the following nerves dissected: 1 – lingual nerve, 2 – glossopharyngeal nerve, 3 – hypoglossal nerve.

Funding

None.

Declaration of Competing Interest

None.

References

- [1] Harrison JD. Modern management and pathophysiology of ranula: literature review. *Head Neck* 2010;32:1310–20.
- [2] Patel MR, Deal AM, Shockley WW. Oral and plunging ranulas: what is the most effective treatment? *Laryngoscope* 2009;119:1501–9.
- [3] Parekh D, Stewart M, Joseph C, Lawson HH. Plunging ranula: a report of three cases and review of the literature. *Br J Surg* 1987;74:307–9.
- [4] Chen JX, Zenga J, Emerick K, Deschler D. Sublingual gland excision for the surgical management of plunging ranula. *Am J Otolaryngol* 2018;39:497–500.
- [5] Huang SF, Liao CT, Chin SC, Chen IH. Transoral approach for plunging ranula—10-year experience. *Laryngoscope* 2010;120:53–7.
- [6] Mahadevan M, Vasan N. Management of pediatric plunging ranula. *Int J Pediatr Otorhinolaryngol* 2006;70:1049–54.
- [7] Samant S, Morton RP, Ahmad Z. Surgery for plunging ranula: the lesson not yet learned? *Eur Arch Otorhinolaryngol* 2011;268:1513–8.
- [8] Zhao YF, Jia J, Jia Y. Complications associated with surgical management of ranulas. *J Oral Maxillofac Surg* 2005;63:51–4.
- [9] Zhao YF, Jia Y, Chen XM, Zhang WF. Clinical review of 580 ranulas. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2004;98:281–7.
- [10] Zhi K, Wen Y, Zhou H. Management of the pediatric plunging ranula: results of 15 years' clinical experience. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2009;107:499–502.
- [11] Lesperance MM. When do ranulas require a cervical approach? *Laryngoscope* 2013;123:1826–7.