The Laryngoscope

Transoral Approach for Plunging Ranula—10-Year Experience

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Objective/Hypothesis: Plunging ranulas (PRs) are infrequently encountered. The origin of PRs is mostly from the sublingual gland (SLG). Different routes to manage PRs include marsupialization, simple SLG excision, and combined SLG and submandibular gland (SMG) excision either transcervically or transorally. In this study, we demonstrated our experience managing PRs via transoral excision of the SLG with marsupialization.

Study Design: Retrospective study.

Methods: We retrospectively reviewed 20 patients at Chang Gung Memorial Hospital, Linkou, between January 1999 and April 2009. All patients received preoperative computed tomography or magnetic resonance imaging and were clinically diagnosed with PR. At surgery, the SLG was excised transorally with preservation of Wharton's duct and lingual nerve. The mucus contents of the PRs were drained through the posterior edge of the mylohyoid muscle.

Results: Two patients were found to have final diagnosis of lymphangioma. Eighteen patients were eligible for analysis. The age distribution ranged from 6 to 48 years old. Sexual distribution was equally distributed. The successful rate was 17/18 (94.4%) with a lingual nerve paresthesia rate of 2/18 (11.1%). The paresthesia persisted for 3 and 6 months, respectively. One recurrent patient was salvaged by excision of the SMG and ranula. The other two recurrent patients received excision of the SMG and cyst and had a final diagnosis of lymphangioma.

Conclusions: Transoral approach with excision of the SLG alone provides a high success rate (94.4%) with minor complications and could be the first choice in managing PRs. The results of this study confirm PRs mostly originate from the SLG.

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INTRODUCTION

The term "ranula" describes the blue, translucent swelling in the floor of mouth.[1] A simple ranula can be either a mucus retention cyst or a mucus extravasation pseudocyst confining in the floor of the mouth.[2] A plunging or cervical ranula is a mucus extravasation pseudocyst more commonly arising from the sublingual gland (SLG) and presents as a swelling in the neck. It may appear as a submandibular mass without apparent intraoral involvement, in which case the diagnosis is usually difficult. It is most commonly centered on the submandibular triangle with an average size of 4 to 10 cm, but can extend superiorly in the parapharyngeal space as far as the skull base, inferiorly to the supraclavicular area, posteriorly into the retropharyngeal space, or across the midline anteriorly.[3]

Different approaches were used to treat plunging ranulas (PRs), including marsupialization, simple SLG excision, combined SLG excision with marsupialization, PR excision alone or combined with SLG excision and sclerotherapy, or combined excision of the SLG and submandibular gland (SMG).[4,5] Parekh et al. [5] reviewed the results of all forms of treatment in the literature until 1989 and found a recurrence rate in excess of 50% for any surgery that did not involve excision of the SLG. Excision of the SLG alone or in association with other treatments resulted in a recurrence rate of 2% or less, and they concluded that the SLG should always be excised in PRs. The excision of the SLG could be approached transcervically or transorally. Transcervical approach is usually combined with SLG, SMG, and ranula excision.

Transcervical approach provides access to the SLG in the sublingual space, when adequate exposure of this space is achieved by raising the mylohyoid muscle anterosuperiorly. In PRs, excision of the SMG and incision of the cyst facilitates this exposure. The transcervical approach of treating PRs was reported to be an optimal access both on the lesion and the SLG.[6,7] However, the

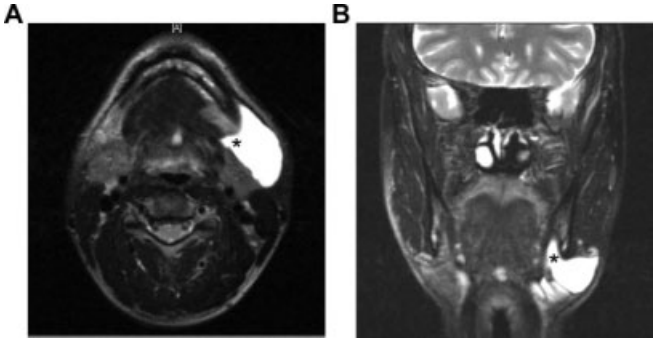

Fig. 1. (A) Axial view magnetic resonance imaging (MRI) (T2-weighted) of patient 11, showing the high intensity mass (*) in the left oral floor. T1-weighted MRI shown in low intensity. (B) Coronal view MRI (T2-weighted) of patient 6, demonstrating the high-intensity mass (*) in the oral floor and typical tail sign, which showed SLG origin of plunging ranula.

transoral approach provides a direct access to the SLG with minimal complications.

In this study, we retrospectively reviewed 20 patients in our institute with a clinical diagnosis of PR, and analyzed the treatment outcomes and complication rates through transoral approach during a period of 10 years.

MATERIALS AND METHODS

All patients managed at the Department of Otolaryngology, Chang Gung Memorial Hospital, Linkou, between January 1999 and July 2009 with a clinical diagnosis of PR were included in our study. All patients received preoperative computed tomography (CT) or magnetic resonance imaging (MRI) of head and neck. All of them were found to have a "tail-sign" or diagnosis of PR on imaging studies (Fig. 1 and Fig. 2). Demographic data, including age at presentation, lesion side, and gender were obtained for all patients from the medical records. Specific questions regarding local trauma or treatment, and symptoms at presentation were recorded.

At surgery, the patients were under general anesthesia via oral intubation. A mucosal incision was made along the SLG. The SLG was freed from both the Wharton's duct and the lingual nerve by blunt dissection (Fig. 3). Small vessels in the field were coagulated using a bipolar coagulator. After removing the

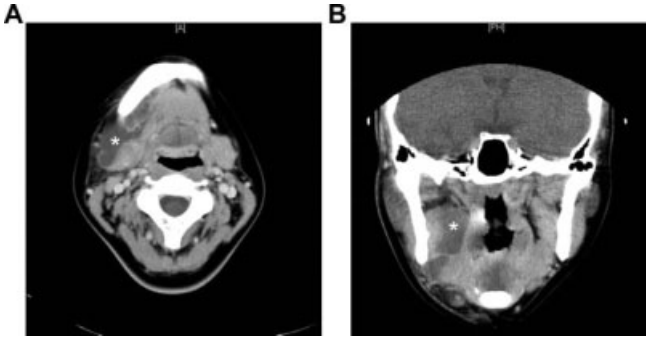

Fig. 2. (A) Axial view computed tomography CT scan of patient 14, showing low-intensity mass in anterior neck region. (B) Coronal view CT showing a submandibular region cystic mass (*), which had no definite relationship with SLG and was difficult to differentiate from lymphangioma.

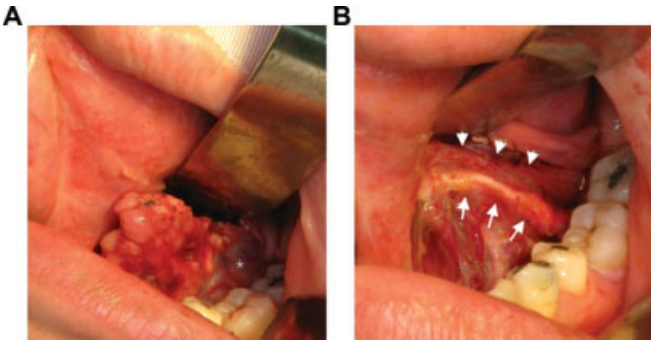

Fig. 3. (A) Transoral approach to excise the SLG. The intraoral part of plunging ranula (*). (B) After removal of the SLG, preservation of Wharton's duct (arrowheads) and lingual nerve (arrows) in patient 15. The dehiscence posterior to the mylohyoid muscle could be found after removal of the SLG. [Color figure can be viewed in the online issue, which is available at www.interscience.wiley.com.]

SLG, a simple drainage of the mucus within the ranula was performed by blunt dissecting of the posterior edge of the mylohyoid muscle. A Penrose drain was placed for potential residual cystic contents to disappear after complete resection of the source of secretions (i.e., the SLG). Mucosal approximation using absorbable sutures (Vicryl; Ethicon, Somerville, NJ) was performed coarsely to prevent hematoma formation. Any postoperative complications were recorded, and all patients were followed until the time of this writing.

RESULTS

The demographic data of the patients and the characteristics of the ranulas in these patients are described in Table I. Two patients with lymphangioma (Table I, patients 12 and 18) in final pathology were excluded from this analysis. The mean age of patients at presentation was 23.1 years (range, 6–48 years). Of the 18 patients, 9 (50.0%) were female and 9 (50.0%) were male. The locations of the PRs were evenly distributed on both sides. Of the PRs, all the patients had cervical swelling, and eight had intraoral swelling. The mean duration of ranula prior to presentation was 5.1 months. Seven patients had undergone previous surgery, which had comprised excision of sublingual mucocele or sublingual ranula (n = 4), surgery for Wharton's duct stones (n = 2), and SMG excision (n = 1). The mean ages in patients with and without surgical history were 21.29 and 24.18 years old, respectively (Student t test: P = .608).

No immediate complications, including hematoma or wound infection, were encountered in our patients. The recurrence rate in the transoral approach was 1/18 (5.6 %). Two patients in transoral group had postoperative lingual numbness, which persisted 3 (patient 11), and 6 (patient 15) months, respectively. The patient with recurrence occurred at 3 months after surgery. The recurrence was salvaged by excision of the SMG and ranula transcervically. Two patients with recurrence of ranulas were treated with excision of the SMG and cyst walls and had a final diagnosis of lymphangiomas. No more recurrences occurred after the surgeries in these three patients.

TABLE I.
Clinical Data of the Patients.

No.	Age (Yr)	Sex	Side	Surgery	Trauma History	Complications	Recurrence
1	48	F	L	I	—	—	—
2	31	M	L	I	(+) Removal of Wharton's duct stones × 2 times	—	—
3	25	M	L	I	(+) Excision of the SMG for sialoadenitis	—	—
4	31	F	L	I	(+) 1. Wharton's duct stone removal; 2. Sublingual ranula s/p transoral excision	—	—
5	6	F	L	I	(+) 2 times marsupialization and excision of the SLG for sublingual ranula	—	—
6	23	M	L	I	—	—	—
7	13	M	R	I	—	—	—
8	24	F	R	I	(+) S/p excision for sublingual ranula	—	—
9	8	M	R	I	(+) S/p excision for sublingual ranula	—	—
10	22	F	R	I	—	—	—
11	27	M	L	I	—	+*	—
12	33	F	L	I+II	—	—	Lymphangioma
13	22	M	R	I	—	—	—
14	24	F	L	I	(+) S/p excision for sublingual ranula	—	—
15	18	F	R	I	—	—	—
16	37	F	L	I	—	+*	—
17	30	F	R	I	—	—	+
18	27	M	R	I+II	—	—	Lymphangioma
19	8	M	R	I	—	—	—
20	18	M	R	I	—	—	—

*Lingual nerve paresthesia.
F = female; M = male; L = left; R = right; I = transoral approach; II = transcervical approach.

DISCUSSION

Elevation of the floor of the mouth on the affected side and a bluish discoloration hint of the origin of PRs. It commonly occurs in young adults[3,4,8] and a slight female preponderance of about 1.3:1 (F:M).[3] In our study, no sexual predilection was found, and the mean onset of age (23.1 years old) is consistent with previous reports. CT/MRI study with contrast is helpful in demonstrating the origin of these lesions. In PRs, a radiolucent and duct-like extension exists between the lesion and the SLG, and is usually located at the posterior margin of the mylohyoid muscle or in a dehiscence in the muscle.[9] Usually, a so-called tail sign is diagnostic for PRs in CT/MRI studies (Fig. 1 and Fig. 2).[10]

The etiology of the ranula has been described in association with the SLG.[11,12] SMG is also a possible source of ranula. However, it was encountered in rare cases.[13] In this study, the successful treatment of PRs by excision of the SLG alone implies the PRs mostly originated from the SLG. Histologically, a PR consists of a central cystic space containing mucin and a wall composed of loose, vascularized connective tissue. The SLG secretes continuously in the interdigestive period, and mucus extravasation probably occurs as a result of a ruptured acinus due to increased secretory back pressure when the duct is obstructed, or due to direct leakage if the duct or acini are damaged.[14] Extension of ranula into the neck occurs through two pathways: 1) along the deep lobe of the SMG between the mylohyoid and hyoglossus muscles, or 2) through a congenital dehiscence in the mylohyoid muscle,[15] in which some part of the SLG projects in up to one third of normal subjects, being the mylohyoid "boutonnière" of Gaughran.[16]

A wide variety of surgical procedures were undertaken in the therapy of PRs (Table II).[5,14,17–25] Many factors affect the choice of surgery, including the uncertain etiology of PRs decades ago, the surgeon's preference, and the correct diagnosis being made before surgery. Nowadays, excision of the SLG is the essential part of treating PRs, because Parekh et al.[5] found a recurrence rate of 2% after excision of the SLG. The intraoral approach was suggested to provide direct and better access for complete removal of the SLG,[14] and it was proved to be a safe way to treat PRs in our study. Three patients had recurrence of neck swellings after surgery: one had PR recurrence and the other two patients had submandibular lymphangiomas. Residual SLG or Wharton's duct stenosis caused by injuries during dissection are the two most probable causes. Another less frequently met origin of recurrence is the ectopic SLG. Bridger et al.[26] made the point that exploration below the mylohyoid muscle for any evidence of ectopic SLG in transoral approach for PRs is important, as this tissue could be a source of recurrence. From our point of view, residual glands could be the most possible origin of recurrence.

TABLE II.
Literature Discussing the Treatment of Plunging Ranula.

Authors/Year	No. of Patients	Operation Method	Recurrence Rate (%)	% Complication Rate
Literature before 1987 (Parekh)[5]	17	Incision and drainage	12/17 (70.6)	ND
	19	Marsupialization or other oral procedures	10/19 (52.6)	ND
	33	Cervical excision of cysts	28/33 (84.8)	ND
	26	Cervical excision of cyst combined SLG	1/26 (3.8)	ND
	14	Intraoral SLG and drainage of the cyst	0/14 (0)	ND
	30	Radiation therapy	1/30 (3.3)	ND
Parekh D, et al. (1987)[5]	3	Transcervical SLG+SMG	0	ND
Ichimura K, et al. (1996)[17]	7	Transcervical SMG+SLG (five)		ND
		SMG 2		ND
Davison MJ, et al. (1998)[14]	13	Transcervical SMG+SLG (one intraoral)	15.4	23.1, lingual paresthesia
	1	Transcervical SMG	0	
	1	Transoral SLG	0	
	2	Transcervical SLG	0	
	3	Transcervical excision of ranula	0	66.7 (lingual paresthesia, or mandibular n palsy)
Anastassov, GE, et al. (2000)[18]	2	Transcervical SLG+SMG with cyst	0 (submandibular in origin)	ND
Iida S, et al. (2001)[19]	1	Transcervical SLG+SMG	0	0
Morita Y, et al. (2003)[20]	2	SMG*	100	ND
	1	SMG+SLG	0	ND
	2	SLG†	0	ND
Kobayashi T, et al. (2003)[21]	6	Intraoral SLG	0	0
Takagi S, et al. (2003)[22]	4	Fenestration with continuous pressure	0	0
ZhaoYF, et al.(2004)[23]	119	No detailed information about surgery in the patients	ND	ND
Mahadevan M, et al. (2006)[24]	21 (pediatric)	Intraoral SLG	0	0
Chidzonga MM, et al. (2007)[25]	6	SLG	0	ND
		SLG+SMG	0	ND
Present study (2009)	18	Intraoral SLG	5.6	11.1

*Both of the patients had recurrences.
†Recurrent cases from the two patients (*).
ND = not described; SLG = excision of sublingual gland, SMG = excision of submandibular gland.

Extensive dissection of the pseudocyst is unnecessary[5] because the pseudocapsule is devoid of epithelial lining and has no potential for mucus production itself. Removing the origin of the disease, usually the SLG, is the key to successful treatment of PRs. In all our patients, drainage of the mucus content was done through the posterior edge of the mylohyoid muscle and no further dissection of cyst wall was ever attempted.

Meticulous hemostasis is important for successful excision of the SLG. The sublingual artery, a branch of lingual artery, penetrates between the SLG and mylohyoid muscle. Inadvertent injury to this vessel during dissecting the deep part of the SLG will make the artery retract into mylohyoid muscle and cause persistent oozing and poor surgical field. Residual SLG usually results from this situation. In addition, lingual nerve, lying deep to the SLG and in the lateral side of the tongue, is vulnerable during electrocauterization to control the bleeding in this region (Fig. 3). In our experience, bipolar cauterization in this area could possibly minimize the injury to the lingual nerve. Two patients (11.1%) in our study suffered from temporary paresthesia in the distribution of the lingual nerve lasting between 3 and 6 months. Zhao et al. reported that one patient had persisted lingual numbness for 2 years.[27] This complication resolves spontaneously in most patients. On the contrary, the transoral approach can avoid the risk of marginal mandibular branch of facial nerve injury. The sizes of PRs are usually large, ranging from 4 to 10 cm.[3] Extensive dissection via the transcervical approach puts marginal mandibular branch at higher risk of injury and cosmetically leaves a scar in the neck after surgery.

From our results, the transoral approach provides a safe and effective method for treatment of PRs. The submandibular swelling in two of our patients persisted after surgery, and was proved to be lymphangioma after excision of the SMG transcervically. PRs could be confused with lymphatic malformations or venous malformations on image studies.[23] At present, echo-guided aspiration of the content of neck lesions and analysis of the amylase level could be helpful in the differential diagnosis. High amylase level suggests a PR,

and the transoral approach seems to be the method of choice.

CONCLUSION

Transoral excision of the SLG with drainage of ranula provides a high success rate (94.4%) in managing PRs with acceptable (minimal) complications. Completely removing the origin of the ranula is the key to successful treatment, and the transoral approach is the treatment of choice for PRs. In this study, it also proved that most of the PRs originated from the SLG instead of SMG.

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