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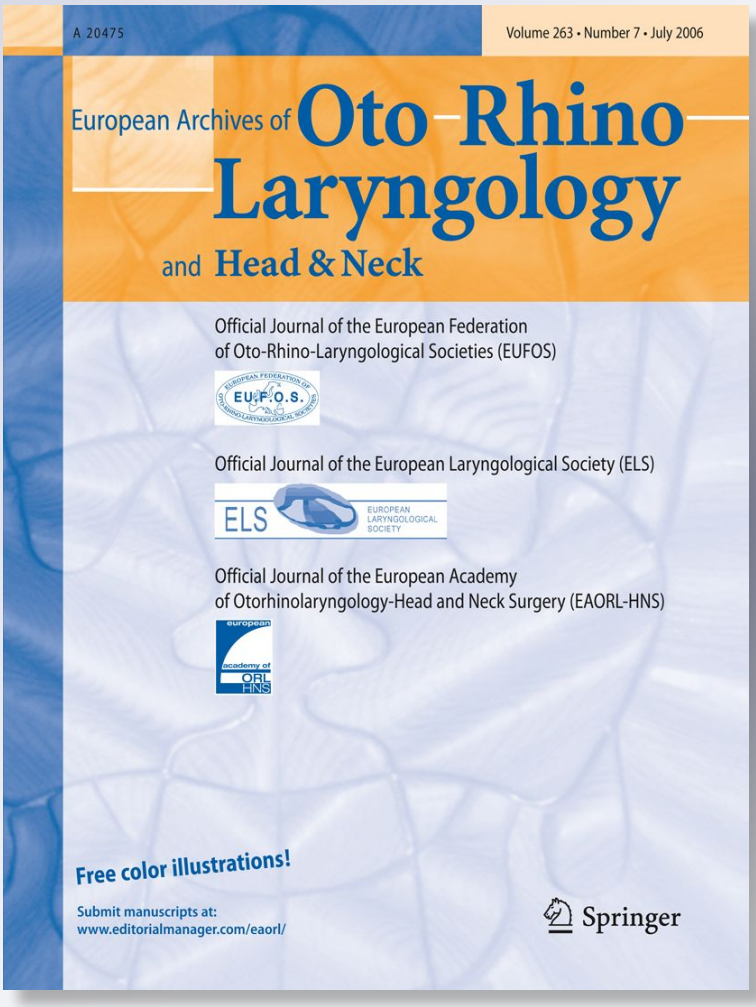

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HEAD AND NECK

Surgery for plunging ranula: the lesson not yet learned?

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Abstract Our objective is to review our experience with treatment of plunging ranula and examine the efficacy of transoral excision of sublingual gland as the principal treatment. This study comprises a case series with chart review. A secondary otolaryngology service was used as the setting. Retrospective analysis of patient records was performed for a series of 95 consecutive cases of plunging ranula, which presented to our department between January 2001 and February 2010. Clinical presentation, investigations, diagnosis, treatment, complications and outcome were recorded. Literature search was performed using MEDLINE and OLD MEDLINE. 81 cases of plunging ranula were treated surgically by transoral excision of sublingual gland and evacuation of ranula contents. Mean operating time was 75.3 min. Twelve patients had undergone previous surgery elsewhere. One patient in our series had a recurrence, needing excision of sublingual gland remnant. Two patients had trauma to submandibular duct requiring excision of submandibular gland. Other complications were minor and transient. Review of literature revealed many diverse methods of treating ranula, with varying results. Our series makes a substantial contribution to the number of plunging ranulas reported in the world, and supports the use of transoral sublingual gland excision as first-line treatment of plunging ranula.

Keywords Plunging ranula · Sublingual gland excision

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Introduction

Plunging ranula, also known as diving or cervical ranula, is a pseudocyst formed by extravasation of mucous from the sublingual gland (SLG) into the submandibular space. It may or may not have an additional oral component.

Although most plunging ranulas appear in second or third decades of life, there have been reports of presentation in infancy [1] which, along with marked preponderance of ranula in peoples of Asian origin [2–4], and anatomical anomalies in the mylohyoid muscle [5, 6] points toward a congenital etiology [4].

The diagnosis of plunging ranula may be made using clinical presentation (Fig. 1) [7]; imaging in the form of Ultrasound, CT scan or MRI [8, 9] and fine needle aspiration for cytology [2] or biochemical analysis [10].

Up to twelve different methods of treating plunging ranulas have been described [6]. Patel et al. [11] recently reported on an online survey of members of the American Head and Neck Society. Among the 220 respondents, the preferred management (39%) of plunging ranulas was excision of SLG plus excision of ranula. A combined approach involving excision of ranula, SLG and submandibular gland (SMG) was favored by 23%, whereas excision of ranula alone was favored by 14% and only 13% opted for excision of SLG with evacuation of ranula. The surgical approach used would be combined (transoral and cervical) for 49%, transoral only in 27%, and cervical only in 23%. About half (52%) of the respondents had experience with 10 or fewer plunging ranulas. Such variation in therapeutic approach would suggest either that all techniques are equally effective, or that many practitioners are unaware of an optimal technique, presumably because the etiologic basis for the disease is poorly understood.

Fig. 1 Clinical presentation of a patient with bilateral ranulas (**a**) and right-sided ranula (**b**)

In this paper, we present a large series of patients with plunging ranula, where the principal operation was transoral excision of SLG with simple evacuation of ranula contents. We believe that our approach to diagnosis and treatment represents the preferred management for this condition.

Patients and method

A retrospective analysis was performed on the data from all patients presenting to South Auckland Health, Department of Otolaryngology, with a diagnosis of plunging ranula. The details included standard demographic information (racial group, age and gender), side and extent of lesion, investigations, surgical time and surgical outcomes. The demographics and epidemiologic considerations of this group have been described elsewhere [4].

Between January 2001 and February 2010, there were 95 cases with a diagnosis of plunging ranula. 57 of these (60%) were male and 38 (40%) were female. There were 45 Maori patients (47%), 36 Pacific Islanders (38%), 7 patients of European descent (7%) and 7 of other racial origins (7%) including Arab, Chinese, and African.

Fifty lesions (52.5%) were on the right side. Four patients (8.4% cases) had bilateral ranulas; two of these presented synchronously, while the other two occurred metachronously. An intraoral component was present in 24 ranulas (26%).

Fine needle aspiration of the cystic collection was performed in 45 cases (47.4%). The finding of mucoid aspirate was considered to support the diagnosis in most of those cases, and in some, it was the only investigation. In 17 cases, the aspirate was positive for amylase, thereby confirming the salivary origin of the fluid. In five cases, the specimen was too viscous for amylase analysis, and for the remaining samples, no biochemical assessment was made. Some samples were sent for cytology, which generally showed inflammatory changes.

Eighty-six patients underwent imaging as part of diagnostic evaluation. Only nine patients had no imaging at all, while three patients had all three imaging modalities performed. Table 1 shows the distribution of the imaging modalities in our series. Virtually all of the radiology reports described a defect in the mylohyoid muscle (Fig. 2). There was evidence of herniation of SLG through the mylohyoid defect in 27 patients, with real-time herniation of SLG on moving the tongue from side to side—observed on Ultrasound—in 25 of them.

This study was approved by the Institutional Clinical Board in accordance with the Auckland Regional Ethics Committee requirements for clinical audit. Anonymized data was collected on a retrospective basis from patients' hospital records.

Surgical technique

All patients had surgery performed under a general anesthetic. About one-third had surgery in the presence of an orotracheal tube, while the remainder had a nasotracheal tube to facilitate intraoral access. The oral cavity was first decontaminated with a throat pack soaked in a solution

Table 1 Ultrasound × CT scan × MRI scan crosstabulation

MRI scan			CT scan		Total
			No	Yes	
No	Ultrasound	No	9	30	39
		Yes	19	28	47
	Total		28	58	86
Yes	Ultrasound	No	3	0	3
		Yes	3	3	6
	Total		6	3	9

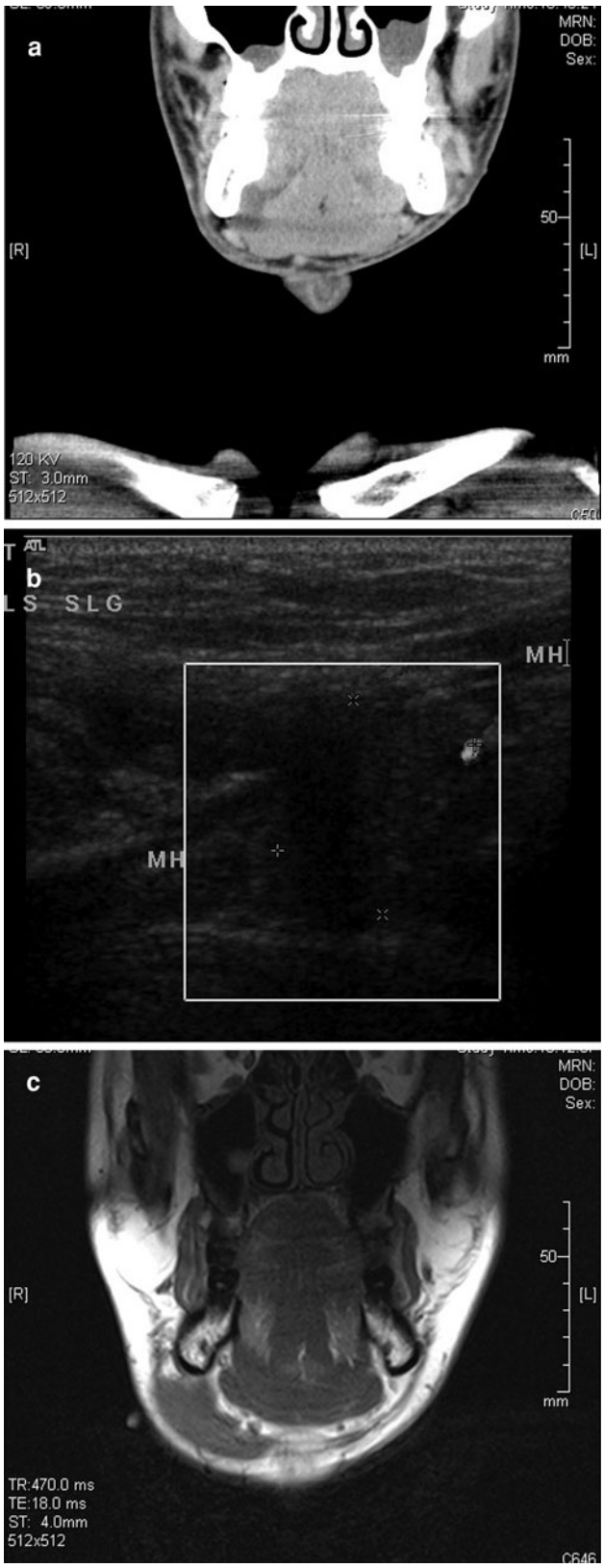

Fig. 2 Pseudocyst with mylohyoid defect seen on (**a**) CT scan, (**b**) ultrasound, and (**c**) MRI

containing 10 ml of normal saline and 40 mg of gentamycin (Fig. 3). The ipsilateral floor of the mouth was then infiltrated with 0.5% bupivacaine and 1:200,000 adrenaline and the incision marked (Fig. 4).

The incision was made in the line of the submandibular duct, on its medial, or lingual, aspect (Fig. 5). Mucosa overlying the SLG was then dissected off the gland using scissor dissection. Traction was then applied to the gland using Babcock's forceps (Fig. 6) and blunt dissection performed to identify the submandibular duct and the lingual nerve (Fig. 7). These structures were gently reflected off the SLG and any sublingual duct joining the submandibular duct was divided. The SLG was then dissected from its bed.

A fibrous tract was usually encountered passing through a dehiscence in the lateral attachment of the mylohyoid muscle, adjacent to the mandible, at approximately one-third of the way along the length of the attachment from the midline.

Some patients had obvious herniation of the SLG through this dehiscence (Fig. 8). Occasionally, the pseudocyst tracked around the posterior-free margin of the mylohyoid, rather than passing directly through a dehiscence in the muscle. The tract was followed into the neck and any residual collection expressed through the dehiscence into the mouth.

A drain was inserted through the cystic space, emerging in the neck, for the first few cases. Thereafter, no drain was used. The mucosa was closed with two or three interrupted dissolvable sutures.

Results

Eighty-two patients had surgery. Of the other 13 cases, four resolved spontaneously, while six patients failed to attend

Fig. 3 Operative setup with gentamicin-soaked throat pack in oral cavity

Fig. 4 Floor of mouth after infiltration, showing incision site

Fig. 5 Mucosal incision

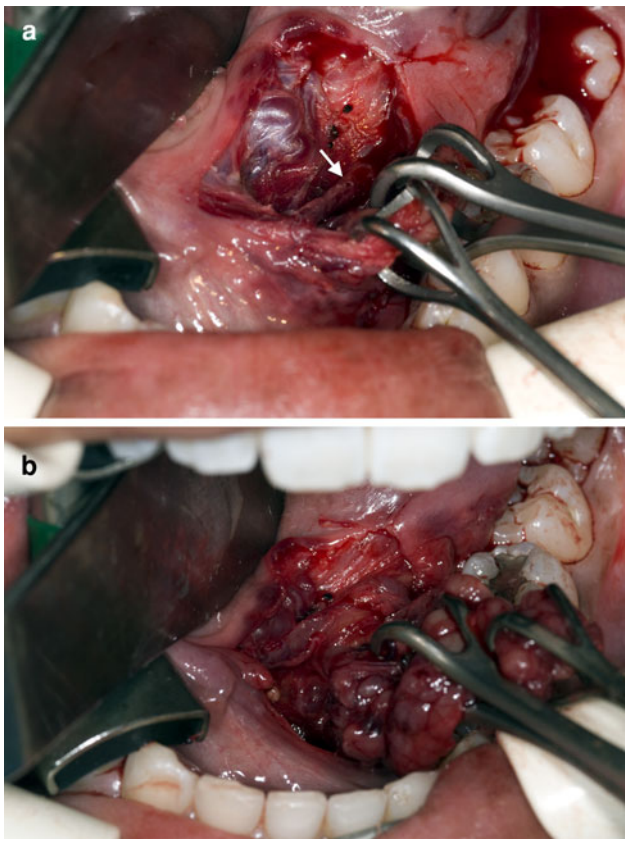

Fig. 6 Dissection of SLG with arrow indicating SM duct (**a**) and with retraction of SLG using Babcock's forceps (**b**)

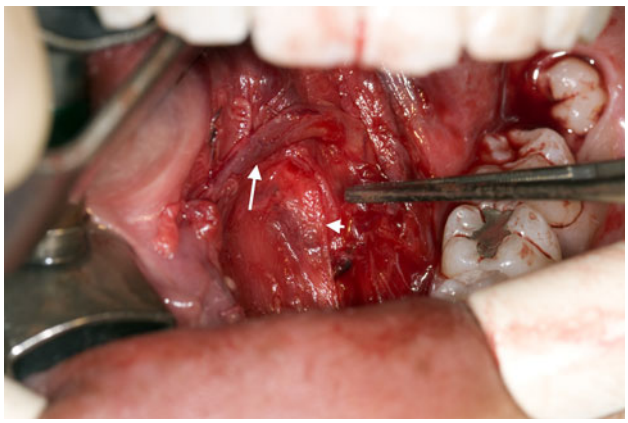

Fig. 7 Submandibular duct (*arrow*) and lingual nerve (*arrowhead*) after removal of SLG

their radiologic examination or follow-up appointments. Three patients were awaiting surgery.

The first patient in the series underwent external excision of ranula and ipsilateral SMG and SLG and was excluded from further analysis.

Of the remaining 81 patients, 12 (15%) had had previous surgery for plunging ranula, and some more than once. Table 2 summarizes these cases.

The mean operating time was 75.3 min (standard deviation 30 min). The operating time ranged between 38 and 151 min. The surgeon who had done the greater number (30 operations) had a mean operating time of 53.1 min (standard deviation 10.4 min) as compared with 111.5 min for the occasional surgeon (six operations).

All operative specimens of SLG were subjected to histological evaluation and virtually all showed extravasation of mucous within the gland. One report did not comment on extravasation and another did not find any evidence of extravasation.

One of our patients had recurrence. He underwent revision transoral surgery, at which time residual SLG tissue was found passing through the mylohyoid defect, and was excised.

Most patients (69%) had no complications. Six patients (7%) had significant morbidity; four developed a postoperative infection requiring readmission to hospital and drainage of a submandibular collection. Two patients had trauma to the submandibular duct requiring subsequent resection of the SMG. Other minor complications

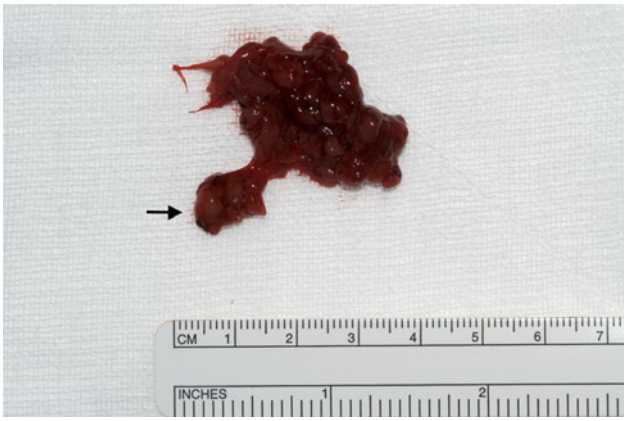

Fig. 8 SLG specimen showing herniated portion (*arrow*)

comprised bruising or minor collection in the submandibular region that subsided over time. Seven patients experienced lingual nerve neuropraxia. Three of these also had edema or collection, whereas other two had associated submandibular duct trauma.

Discussion

Our approach, involving transoral SLG excision and evacuation of cystic contents as sole treatment for plunging ranula, resulted in a 99% success rate for this large series that included both primary and recurrent ranulas. Our experience is supported by a recent study of 20 cases by Huang et al. [12] that also concluded that transoral excision of SLG with evacuation of the ranula provides a high success rate and acceptable complications.

Morbidity in our patients was mostly minor. However, the submandibular duct was unintentionally divided twice: once by a trainee under supervision, and once in a recurrent case, where there was considerable fibrosis making dissection difficult. A few other major complications related to the cystic collection in the neck, which may have been avoided with the insertion of a drain into the neck at the time of surgery.

Harrison recently performed a meta-analysis of published treatment for plunging ranula [6]. He showed an overall recurrence rate of 31% from 449 cases and 13 different treatment options (varying from incision and drainage, through instillation of OK-432 or botulinum toxin, to various combinations of SMG, SLG and cyst excision). The best results (i.e., cure rates more than 90%) were those that included SLG excision, either alone or in combination with other structures. Harrison [6] concluded that a successful approach to the treatment of plunging ranula depends on complete excision of SLG or excision of that part of SLG from which extravasation occurs.

Harrison [6] also considered that successful treatment might follow induction of fibrosis or inhibition of secretion. In general, however, non-surgical treatments designed to avoid complications of surgical treatment, especially those that were associated with more aggressive surgery (such as excision of the pseudocyst and excision of the SMG), are less successful at controlling the disease [6, 10, 11, 13, 14].

The histological finding of extravasation of mucus in the excised SLG explains the pseudo-cystic nature of the fluid collection and why simple drainage of the pseudo-cyst is sufficient management of the collection for the large majority of our cases. It also supports the concept of a disrupted SLG as the source of the cyst [3, 6, 10]. Residual, disrupted SLG was encountered in all recurrent cases that we treated. Clearly it is important to perform a complete excision of the SLG in order to remove the disrupted segment of the gland.

We would add a note here on the diagnosis of plunging ranula: Mahadevan and Vasan [2] considered cytological diagnosis to be definitive with no need for imaging. Others have reported on the radiological features of ranulas such as incompletely septate lesions in the neck, to differentiate them from cystic hygromas, thyroglossal cysts and lymphangiomas [8]. In our experience, it is largely sufficient to demonstrate the mucoid nature of the aspirate or the presence of amylase with imaging as an ancillary investigation. Ultrasound in the hands of an experienced practitioner will demonstrate the presence of a mylohyoid dehiscence, even more reliably than CT scanning [15]. Moreover, many studies will show actual herniation of the SLG through the dehiscence.

Table 2 Median time to recurrence after the previous procedure in months

Previous procedure	Number of cases	Median time to recurrence (months)
External SLG excision	2	6
External SMG excision	3	6
Incision and drainage	3	8
Transoral SLG excision	3	24
External excision of SMG and cyst	1	24
Excision of cyst	4	38

Finally, from Harrison's [6] literature review, it is apparent that authors continue to exhibit a lack of understanding of the pathophysiology of plunging ranula; this is manifest in descriptions of treatment methods that are either unnecessary or inappropriate. Hopefully, as more papers appear with evidence supporting successful treatment that addresses the SLG disruption, the true nature of plunging ranula will become more widely understood and accepted.

Conclusion

There are many methods of treating ranulas, with varying results, reflecting some uncertainty about the most effective treatment modality. Our experience with a series of 81 patients supports the following conclusions. Clinical identification of a mucoid aspirate, which may be tested for the presence of amylase, is usually sufficient for diagnosis. This may be further endorsed by identification of a mylohyoid defect with Ultrasound or other imaging. Simple transoral SLG excision, with evacuation (but not excision) of the cervical cystic contents provides excellent results, and should be the preferred surgical approach.

Conflict of interest The authors declare that they have no conflict of interest.

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