Conservative Treatment of Oral Ranula by Excision With Minimal Excision of the Sublingual Gland: Histological Support for a Traumatic Etiology

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Purpose: This study investigates, clinically and histologically, a new conservative technique for the treatment of oral ranula based on the premise that a discrete unit of the sublingual gland feeds the ranula, which can therefore be treated by local removal with the attached part of the sublingual gland.

Patients and Methods: The study group consisted of 8 patients with ranula treated by decompression of the ranula followed by local surgical removal together with the attached part of the sublingual gland. Detailed histologic examination of the entire specimen was undertaken in every case.

Results: The treatment was successful in all the patients and there have been no recurrences after reviews of from 13 to 29 months (median, 26 months). Histologic examination of the entire specimen showed communication between the removed part of the sublingual gland and the ranula by way of a torn duct in every case.

Conclusions: The premise that the ranula is fed by an attached, discrete unit of the sublingual gland has been vindicated and is the basis for the successful conservative treatment of ranula by decompression and local surgical removal together with the attached part of the sublingual gland. The finding of communication between the attached sublingual gland and ranula in every case indicates a traumatic etiology for these ranulas.

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Most ranulas are large extravasation mucoceles that arise from the sublingual gland and are sufficiently extensive to form a swelling that resembles the belly or vocal air sac of frog. They are cystic and are frequently blue owing to the Tyndall effect, whereby blue light is reflected more than red light at the interface of soft tissue and cyst. Most extravasation mucoceles occur in the lower lip[1] and are treated successfully by removal of the mucocele with the feeding minor salivary gland. Although the floor of the mouth is the second most common site for extravasation mucoceles,[1] the treatment of the ranula is varied and not always successful.[2-6] Treatment by incision, simple marsupialization, and excision of the ranula alone have a high recurrence rate, whereas excision of the sublingual gland with or without the ranula is almost always successful.[7-9] Although the removal of the sublingual gland as the source of the extravasated mucus may be appealing, it is technically demanding and associated with notable morbidity that can include damage to the lingual nerve, Wharton's duct, submandibular gland, and blood vessels.[8,9] This has encouraged a search for a satisfactory conservative approach to treatment. Marsupialization with packing of the ranula is successful in about 90% of cases[2-4,6] and intracystic injection of the sclerosing-preparation OK-432 has given variable results.[10,11]

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Table 1. EXTRAVASATION MUCOCELES

Site of Mucocele	Patients (n)
Left lower lip	15
Right lower lip	11
Centre of lower lip	2
Left buccal mucosa	3
Right buccal mucosa	1
Left floor of mouth	1
Ventral surface of tongue	1
Orifice of right Wharton's duct	1
Total	35

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This study describes a new method of treating ranulas based on principles applied to extravasation mucoceles elsewhere in the mouth. The premise adopted is that the ranula is fed by a discrete collection of sublingual tissue attached to it. Consequently it can be successfully treated by removal with only a portion of the gland.

Patients and Methods

In the years from 2001 to 2006, 35 consecutive patients were referred with extravasation mucoceles of minor salivary glands, most of which were in the lower lip (Table 1). Records of these patients were retrieved and reviewed retrospectively. This cohort comprised 21 males and 14 females and ranged in age from 18 to 54 years (median, 31 years).

The treatment of these mucoceles was by surgical excision under local anesthesia. A standard approach was used whereby the mucocele was excised without rupture together with any attached minor salivary glands and the wound was closed directly.

A similar approach was adopted for ranulas. However, to make the ranula amenable to local excision, it was decompressed by aspiration of some of its contents before surgery, usually by 3 to 5 days, with the aim of producing a visible, palpable cyst that had been deflated by about a third and was small enough to be amenable to enucleation. A large cyst is prone to rupture and it is important that the integrity is maintained to identify the feeding sublingual tissue. It is not advisable to aspirate at operation because this produces a persistent leak of saliva from the puncture point throughout the procedure unless the puncture point is ligated. The cyst should not be evacuated completely, otherwise the capsule cannot be identified.

Surgery was under general anesthesia, the ranula was approached through a longitudinal incision in the floor of mouth medial to the ranula. The cyst was carefully freed from local tissues. This required careful and deliberate local dissection at the margin of the capsule, which was possible when the tension within the cyst had been released by the aspiration. When the cyst was freed from local structures, it became apparent that it was firmly attached to part of the sublingual gland. At the point of attachment to the sublingual gland, a portion of the gland was excised in continuity with the ranula using cutting diathermy to liberate it from the main portion of the sublingual gland.

In 2005 and 2006, 8 consecutive patients referred with ranulas were treated in this manner and data were collected prospectively (Table 2). The ranulas were typical oral ranulas lying in the paralingual space of the floor of the mouth, not plunging, cervical ranulas.

All patients were reviewed at 1 week, and those who had ranulas were subsequently kept under regular review. Patients with mucoceles of the minor salivary glands were surveyed by post or telephone for evidence of recurrence.

The excised ranulas were fixed in formalin (Fig 1), sliced at about 4-mm intervals and labeled consecutively, embedded in wax, and sections were cut at intervals throughout the embedded material and stained with hematoxylin and eosin (H&E) or with Alcian blue at pH2.5 followed by periodic-

Table 2. RANULAS

Gender	Age (yr)	Presenting Complaint	Site of Ranula	Duration Before Presentation (wks)
M	33	Recurrent swelling	Right floor of mouth	6
M	35	Swelling	Right floor of mouth	Patient unsure
F	29	Swelling	Left floor of mouth	4
F	24	Swelling	Right floor of mouth	8
F	15	Swelling	Right floor of mouth	32
F	20	Swelling	Left floor of mouth	8
F	30	Swelling	Left floor of mouth	32
F	26	Swelling	Right floor of mouth	6

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FIGURE 1. Fixed specimen of ranula with attached part of sublingual gland, which communicated with the ranula. *Line* indicates junction of gland (*G*) and ranula. Scale = mm.

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acid-Schiff (ABPAS) for the demonstration of mucosubstance.[12]

Results

Local surgical excision of mucoceles of the minor salivary glands was successful in all patients and no recurrence was reported at review, which ranged from 11 to 65 months (median, 39 months). The only complication of surgery was a local, minor sensory loss in the mucosa of the lower lip at the site of surgery that occurred in about one third of the cases. However, it was so limited as to be of no consequence to the patient.

The 8 patients with ranula were treated under general anesthesia with 1 overnight stay. No recurrences were recorded at review, which ranged from 13 to 29 months (median, 26 months). No complications were encountered except for transient lingual paraesthesia in 2 patients. In 1 case, the lingual nerve was bound with scar tissue as a result of previous surgery to the floor of the mouth and it had to be freed at the time of surgery.

Histologic examination of the ranulas showed cystic extravasation mucoceles in all the cases (Fig 2). They varied in size from 25 to 50 mm. Lobules of sublingual gland were associated with the mucoceles. In 1 case, lobules of submandibular gland were found adjacent to the sublingual lobules. The lumina of the cysts contained mucus in which there were conspicuous macrophages. The capsules of the cysts consisted of granulation tissue in which there were macrophages. Mucus

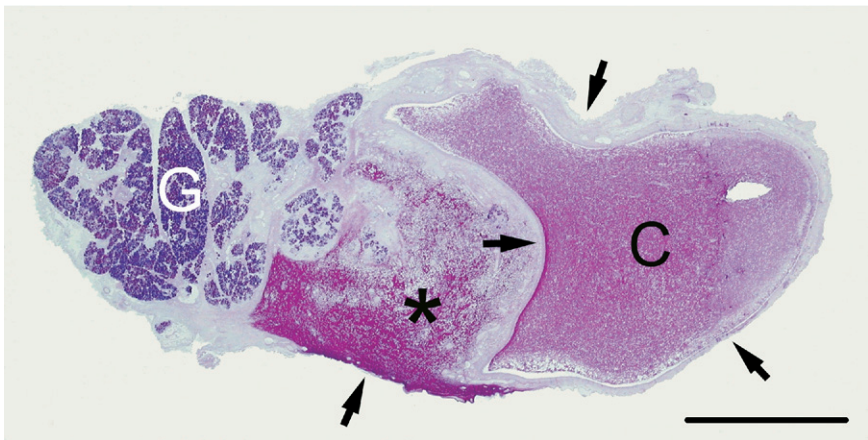

FIGURE 2. Section from transverse slice through ranula and attached part of sublingual gland. Extravasated mucus is stained *purple* and the mucus in the acini of the gland (*G*) is stained *royal blue*. The greater part of the ranula is a cyst (*C*), the lumen of which is filled with extravasated mucus. An extensive region of extravasated mucus intermixed with granulation tissue (*) is present between the cyst (*C*) and the gland (*G*). A palely stained capsule of granulation tissue (*arrows*) limits the cyst and mucus intermixed with granulation tissue. Photograph of section stained with ABPAS. Magnification ×13. Scale bar = 5 mm.

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FIGURE 3. Section from transverse slice adjacent to that of Figure 2. The sublingual gland (*G*) contains mucus in acini that is stained *royal blue*. The gland communicates with the ranula via a duct, the torn end of which (*hollow arrow*) communicates with the lumen of an extension of the cyst, which appears empty owing to a loss of mucus during processing. *Purple* stained mucus intermixed with granulation tissue (*) surrounds this extension. A palely stained capsule of granulation tissue (*arrows*) encapsulates the main cyst (*C*) and the mucus intermixed with granulation tissue at the margin of the specimen. Photomicrograph of section stained with ABPAS. Magnification ×47. Scale bar = 1,000 μm.

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intermixed with granulation tissue was sometimes found adjacent to the cysts and in 1 case was extensive (Figs 2, 3).

Communication between the lumen of the cyst and the sublingual gland was seen in all cases (Figs 3-6). Oral mucosa was seen in 4 cases, in 3 of which a communication was seen close to it (Fig 5). Two communications were seen in 3 cases. In one of these, 1 communication was superficial and close to oral mucosa and the other was to the deep surface of the cyst (Figs 5, 6). In the other 2 cases, the communications were in slices separated by an intervening slice, which indicates that they were over 4 mm apart. The communication was by an interlobular or main duct that passed through the capsule of the cyst to reach the lumen (Figs 3-6) or into mucus intermixed with granulation tissue for 1 of the 2 communications in 1 case. The epithelium that lined the duct at the communication showed a characteristic squamous metaplasia and was sometimes seen to line the luminal surface of the adjacent part of the capsule of the cyst (Figs 4-6).

There were variable inflammation, atrophy, fibrosis, and ductal dilatation in the sublingual gland. There was a small amount of intralobular extravasated mucus in 2 cases, which in 1 case was from acini and ducts that consisted of flattened epithelium surrounding a dilated lumen (Figs 7, 8), and in the other case was from ducts with lymphoreticular metaplasia that were surrounded by lymphoid follicles.

Discussion

The technique of limited removal of sublingual gland is based on the premise that a ranula arises at a discrete point along the sublingual gland. The combination of histological findings and the lack of recurrence confirms the validity of the premise.

The detailed anatomy of the sublingual gland is more complex than is generally appreciated. Leppi[13] discovered that the sublingual gland consists of a constant, lesser sublingual gland and a greater sublingual gland. The latter is posterior to the lesser sublingual gland in the paralingual space and was only found in 10 of 28 people and usually only unilaterally. Furthermore, the sublingual gland is often continuous with the part of the submandibular gland that is above the posterior part of the mylohyoid muscle. Bartholin's duct runs from the greater sublingual gland to either join or open independently of Wharton's duct. The lesser sublingual gland consists of between 8 to 30 small glands, from every one of which a duct of Rivinus passes to open independently on the sublingual fold. Therefore, according to the premise that the ranula arises from 1 small unit of gland,

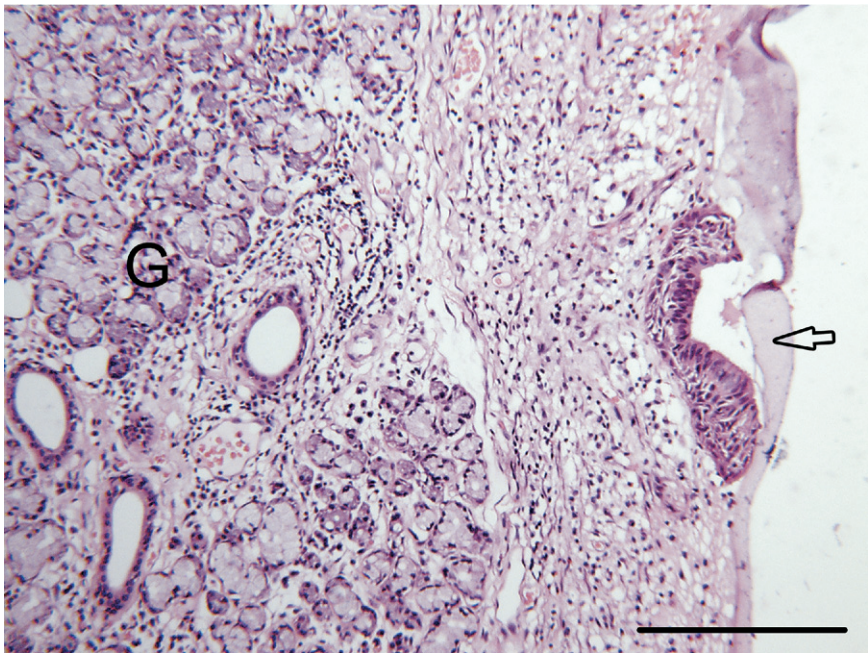

FIGURE 4. Detail of section adjacent to that of Figure 3. The sublingual gland (*G*) communicates with the ranula via a duct, the torn end of which is seen (*hollow arrow*), shows squamous metaplasia and lines the luminal surface of the capsule of the cyst. Mucus is intermixed with granulation tissue between the lumen and the gland. Photomicrograph of section stained with H&E. Magnification ×400. Scale bar = 200 μm.

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once the ranula is dissected free from local connective tissue, only that portion of the sublingual gland attached to the ranula needs to be excised and the majority of the sublingual gland is preserved.

However, the successful eradication of the ranula depends on the successful identification of the secretory unit supplying it at operation. For this to be accomplished, the circumference of the ranula has to be freed

FIGURE 5. Section from transverse slice through ranula. Oral mucosa covers the cyst, close to which a duct (*hollow arrow*) shows squamous metaplasia indicative of a nearby tear and communication with the lumen. Photomicrograph of section stained with H&E. Magnification ×118. Scale bar = 500 μm.

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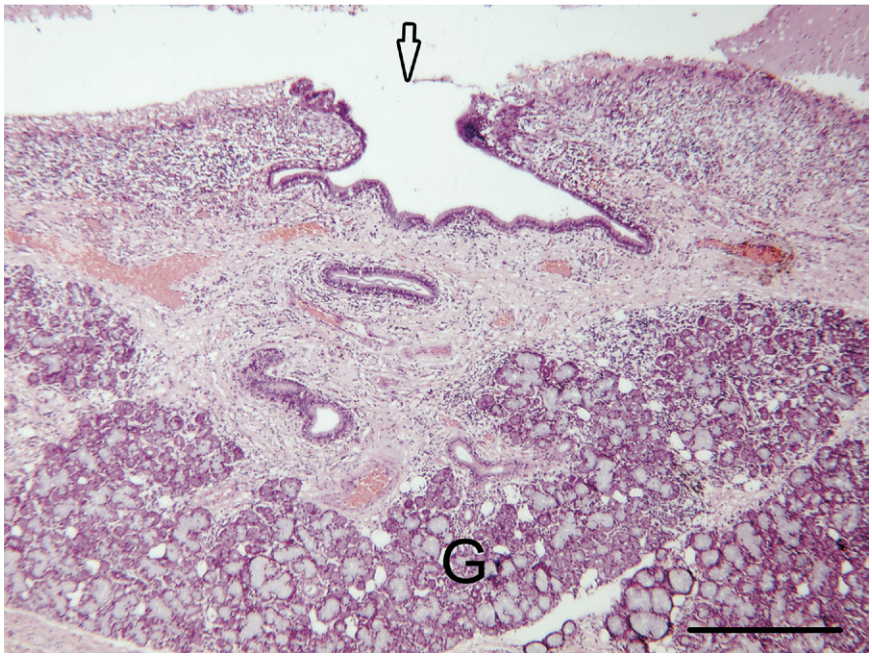

FIGURE 6. Section from transverse slice adjacent to that of Figure 5 through ranula and attached part of sublingual gland. The sublingual gland (*G*) communicates with the lumen of the cyst via a duct, the torn end of which (*hollow arrow*) shows squamous metaplasia. Photomicrograph of section stained with H&E. Magnification ×118. Scale bar = 500 μm.

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from the local tissues. In large ranulas, this is difficult to accomplish without rupturing it. Once it is ruptured, its capsule becomes indistinct and is difficult to enucleate. It is for this reason that the ranula is decompressed before surgery. This is achieved by needle aspiration. These aspirations are undertaken easily without local anesthesia in the dental surgery. Saliva is bacteriostatic and the risk of infection is minimal.

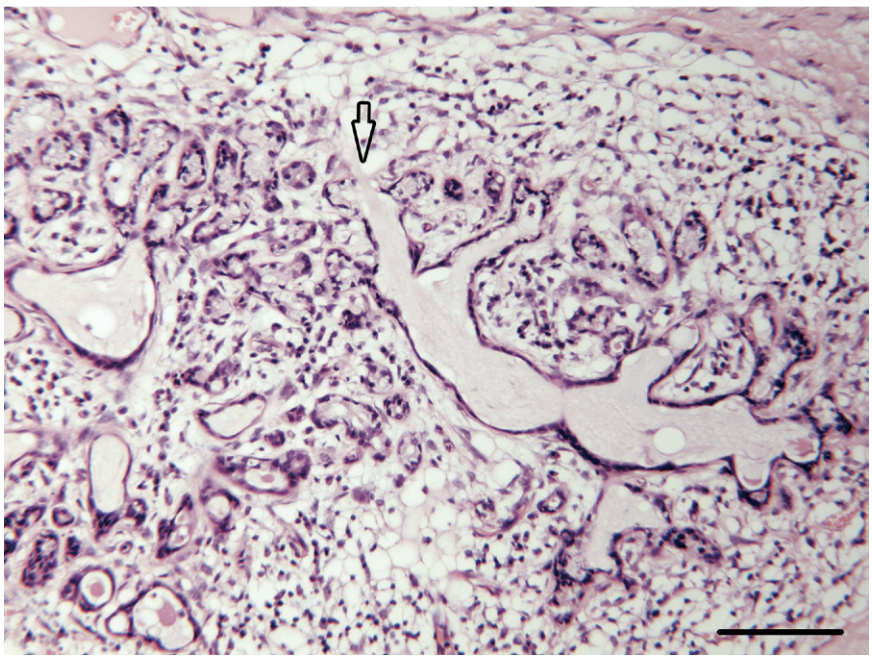

FIGURE 7. Section adjacent to that of Figure 2. Atrophic ducts and acini in the sublingual gland. Mucus has extravasated from a ruptured acinus (*hollow arrow*). Extravasated mucus and inflammatory cells are present interstitially. Photomicrograph of section stained with H&E. Magnification ×476. Scale bar = 100 μm.

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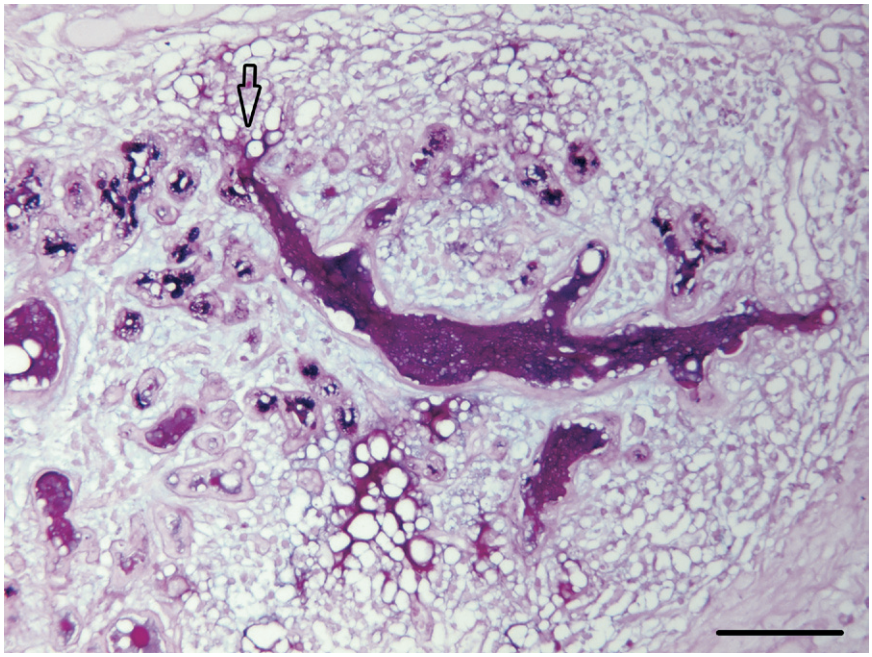

FIGURE 8. Detail of section of Figure 2. The mucus is stained from purple to royal blue, is present in reduced amount in atrophic acini, fills the lumina of the ducts and acini, extravasates through the rupture of the acinus (*hollow arrow*) and is present interstitially. Photomicrograph of section stained with ABPAS. Magnification ×476. Scale bar = 100 μm.

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The histological findings have confirmed that the ranulas in the present investigation are extravasation mucoceles that arise from the sublingual gland. The etiology of extravasation mucoceles elsewhere in the mouth and particularly in the lower lip is usually mechanical trauma.[1,9] However, Zhao et al[7] found a history of trauma to the floor of the mouth in only 2.7% of patients with ranulas. Furthermore, the extravasation had often been found to occur deep in the gland where it is unlikely to be mechanically traumatized.[2,3,14] The etiology of the ranula was considered obscure.[3,15]

One possible etiology is obstruction, for both the sublingual and minor salivary glands are spontaneous secretors with a great resistance to obstruction.[9,16] Supportive evidence for this is that ductal ligation of the feline sublingual gland often results in an extravasation mucocele from an accumulation of mucus that extravasates from ruptured acini and not the ducts.[16] Furthermore, an increased incidence of ranulas associated with untreated HIV infection in Zimbabwe[17,18] is possibly a result of obstruction for the following reasons: there is an increase of inflammation and fibrosis in minor salivary glands of patients with untreated HIV[19] (this would also involve the biologically similar sublingual glands), inflammation and fibrosis cause obstruction of salivary glands,[20-23] and obstruction of the sublingual glands leads to extravasation and possibly the development of ranulas in patients with untreated HIV infection.

However, the findings in the present study support a traumatic etiology for most ranulas. Thus, naturally occurring ranulas have been found in the present and previous investigations to be fed by a damaged duct,[14,15] and although extravasation from acini and intralobular ducts was found in 2 of the present cases, the amount of extravasated mucus was insignificant compared with the amount in the ranulas. The finding that communication between sublingual gland and ranula was close to the oral mucosa in 3 of 4 cases in which oral mucosa was seen, and the finding of both a superficial and a deep communication between sublingual gland and ranula in 1 case suggest that a sublingual duct was originally severed close to the oral mucosa and the 2 ends became separated by the expansion of the ranula. This is the likely explanation for the previous finding that the extravasation often occurs deep in the gland where it is unlikely to be mechanically traumatized,[2,3,14] and for the finding of more than 1 communication between sublingual glands and ranulas in dogs.[14] Thus it seems that mechanical trauma to the floor of the mouth is more common than assumed and passes unnoticed.

The investigation by Glen[14] confirmed that ranulas arise from the sublingual gland and not the submandibular, although he found a coexistent leakage from the anterior border of the submandibular gland in 1 mucocele out of 58. The submandibular gland is not a spontaneous secretor and is far less resistant to obstruction than the sublingual gland, and even when

the submandibular gland is damaged and there is extravasation of saliva, the ensuing granulation tissue can obstruct further extravasation and seal the leak, which is an outcome less likely with the sublingual gland.[9,15,16]

This histological investigation indicates that trauma is the usual etiology of the ranula, as it is of extravasation mucoceles elsewhere in the mouth.

The conservative, enucleation technique described in this investigation preserves most of the sublingual gland and has been successful in 8 consecutive patients. This method merits further evaluation and comparison with other conservative techniques.

Acknowledgment

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