

Plunging ranula: a report of three cases and review of the literature

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Three cases of plunging ranula are reported and the literature reviewed. Extravasation of saliva from the sublingual gland due to trauma or obstruction of its ducts appears to be the most likely cause of plunging ranula. Available data suggest that the submandibular gland is usually not involved, although at the time of surgery it may be extremely difficult to exclude a submandibular origin of the cyst in the neck. Communication between the oral and cervical components of the plunging ranula probably occur via a hiatus in the mylohyoid muscle. Such communication passing directly into the submandibular compartment may simulate submandibular gland involvement. Since 1910, 139 procedures in 89 patients with plunging ranula have been reported in the English literature. The recurrence rate was 70 per cent after incision and drainage of the cyst, 53 per cent after marsupialization, 85 per cent after excision of the cyst in the neck and 2 per cent after excision of the sublingual gland via the cervical or intra-oral route. This review suggests that excision of the sublingual gland with intra-oral drainage of the cervical swelling appears to be the treatment of choice for the plunging ranula.

Keywords: Plunging ranula

Ranula describes the bluish, translucent cystic swelling that occurs in the floor of the mouth. Occasionally an oral ranula gives rise to a cervical extension which is then termed a plunging ranula. Three cases of plunging ranulas are presented and the literature reviewed. Uncertainty with regard to the exact aetiology and pathogenesis of the plunging ranula has resulted in controversies in surgical management.

Case reports

Case 1

A 20-year-old man presented with a 2 year history of a painless swelling in the right submandibular region, which developed 6 months after marsupialization of an oral ranula. Examination revealed scarring in the floor of the mouth and an 18 × 10 cm cystic swelling in the right side of the neck. Aspiration yielded straw-coloured fluid with an amylase content of 1183 units/l. The cyst which appeared to be originating from the submandibular gland was excised with both the sublingual and submandibular glands through a cervical incision. Follow-up at 3 months showed no evidence of recurrence.

Case 2

An 18-year-old man presented with a 4-month history of an oral swelling which had ruptured spontaneously one month before the present admission. Examination revealed a cystic swelling in the floor of the mouth associated with a swelling on the left side of the neck. At surgery the sublingual and submandibular glands were removed via a left cervical incision. Follow-up at 2 years showed no evidence of recurrence.

Case 3

A 15-year-old male presented with a 5-year history of swelling in the floor of the mouth which had discharged spontaneously on two occasions. Eight months before the present admission, marsupialization of the oral cyst had been performed. Examination revealed a cystic swelling in the left submandibular triangle associated with a swelling in the floor of the mouth. The sublingual and submandibular glands were excised through a cervical incision and the cyst cavity aspirated. The patient was free of recurrence at 2 years.

Histology in all three patients showed an inflammatory wall to the cyst with no true epithelium.

Discussion

Controversy over plunging ranula has revolved around the pathogenesis and management of this condition. Although several authors^{1,2} have supported Thompson's³ postulate that the plunging ranula originates from a cervical sinus in a manner similar to that of a branchial cleft cyst, the raised amylase content of the cyst fluid⁴, the initial intra-oral presentation of the ranula and the characteristic absence of a true epithelial lining has refuted this theory.

Currently it is accepted that the plunging ranula is of salivary gland origin⁵. Early authors^{6,7} thought that it was a retention cyst of the sublingual or submandibular glands that had gravitated into the neck. However, the plunging ranula is now thought to be due to mucous extravasation into the tissues from a traumatized sublingual gland or ducts^{4,8}.

Ranula formation has been described in association with congenital anomalies⁹, trauma¹⁰ and disease of the sublingual gland¹¹. Bhasker *et al.*¹² showed that partial severance of the feline sublingual duct produced mucous extravasation with sialocoele formation. Harrison and Garrett⁸ produced sialocoeles following ligation of the feline sublingual duct presumably as a result of mucous extravasation from ruptured acini secondary to excessive secretory back pressure. These series of experiments failed to produce mucocoeles following ligation of the parotid or submandibular ducts⁸.

The differences in response of the major salivary glands to conservative treatment following glandular injury or ductal obstruction have been explained on the basis of their differing secretory physiology. Naturally occurring plunging ranula probably occur from the sublingual gland which secretes continuously during the interdigestive period¹³. In contrast the parotid and submandibular glands secrete after major stimuli such as eating¹³. Sialocoeles following parotid trauma resolve when the patient is starved¹⁴ and ligation of the duct produces atrophy of the gland¹⁵.

Obstruction of the submandibular duct leads to retention cyst formation. Trauma to the gland may lead to sialocoele formation¹⁰. The two reported cases^{5,10} of sialocoeles from the

Table 1 Summary of the operative experience on 89 cases (including our 3 cases) reported in the English literature since 1910

Surgical procedure	Total number of operations	Reported number of recurrences	Reported number with successful outcome with less than 1 year follow-up	Reported number with successful outcome with at least 1 year follow-up
Incision and drainage	17	12	5	0
Marsupialization or other oral procedure	19	10	5	4
Cervical excision of cysts	33	28	5	0
Cervical excision of cyst combined with excision of the sublingual gland	26	1	13	12
Intra-oral excision of the sublingual gland and drainage of the cyst	14	0	2	12
Radiation therapy to neck	30	1	25	4
Total	139	52	55	32

References 1-4, 6, 7, 18-21, 24-41

submandibular gland were secondary to iatrogenic injury, and both resolved on conservative management. A follow-up perchnetate scan in one case¹⁰ showed atrophy of the gland. Thus, while this review suggests that the submandibular gland is usually not the cause of a naturally occurring plunging ranula, its role in the pathogenesis of the plunging ranula has not been completely excluded.

The oral and cervical components of the plunging ranula probably communicate through naturally occurring defects in the mylohyoid muscle. Prolongation of fat or sublingual gland through such defects into the submandibular compartment occur in up to 36 per cent of the normal population¹⁶. The intimate relationship between sublingual and submandibular glands¹⁷ accounts for the intra-operative difficulty^{18,19} in deciding which gland is giving rise to the plunging ranula (Case 1). This complex interrelationship also explains the danger of injury to the sublingual gland during surgical excision of the submandibular gland^{20,21,30} and injury to the submandibular gland during sublingual gland excision^{5,10}.

The sequence of events giving rise to a plunging ranula is probably as follows:

Trauma, infection or disease may result in injury to the sublingual ducts or gland. Alternatively obstruction from atretic²² or abnormal ducts⁴ may lead to acinar rupture from excessive secretory back pressure⁸.

Continuous extravasation of saliva from the injured gland into the floor of the mouth with its associated inflammatory reaction²³ results in the formation of an oral ranula.

Rising tension from continuous extravasation commonly leads to rupture of the oral ranula. However, it may also open up a potential hiatus in the mylohyoid forcing the saliva to extravasate into cervical tissue planes. Marsupialization or other surgical procedures for oral ranula may cause cicatrization in the floor of the mouth. The extravasated saliva cannot now be accommodated by the expansion of the oral compartment and is forced into the neck through the hiatus in the mylohyoid muscle. The saliva in the neck is limited by an inflammatory membrane and presents clinically as a plunging ranula.

Management

Management of plunging ranula over the years has paralleled the theory of pathogenesis in vogue at the time. *Table 1* lists all the various reported procedures and results. Eight-nine cases with an average of 1.5 operative procedures per case have been reported in the English literature. As recurrences are frequent, a follow-up period of at least 1 year is necessary. Radiation therapy, incision and drainage and marsupialization have all fallen into disrepute. Excision of the sublingual gland appears to be essential for cure^{4,5} as clearly shown in *Table 1*. The single reported recurrence after sublingual gland excision was due to incomplete excision of the gland²⁹.

When faced with cystic swelling in the neck, it is tempting to explore it through a cervical approach. However the amylase content of the cyst⁴ and a history of oral ranula should make the diagnosis of a plunging ranula obvious. The cervical approach, as used in our patients, gives access to both the sublingual gland and the cyst. Excision of the submandibular gland facilitates exposure of the sublingual gland with this approach¹⁸. However, extensive dissection of the cervical cyst is probably unnecessary^{4,21} and often tedious as there is no true capsule and the cyst tends to burrow into cervical and parapharyngeal tissue planes. Recent reports indicate that intra-oral excision of the sublingual gland with oral drainage of the cervical cyst is adequate treatment^{4,21}. Care must be exercised to avoid damage to the lingual and hypoglossal nerves and the submandibular gland and duct.

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Management of parotid sialoceles: a simple surgical technique

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Most post-traumatic parotid sialoceles can be managed conservatively with repeated aspirations and compression^{1–3}. However, if a sialocele is resistant to this form of treatment a more aggressive approach is necessary. Re-exploration of the wound and ligation of the proximal duct, tympanic neurectomy, superficial or total parotidectomy, and irradiation, are some of the suggested radical forms of management^{2–4}. These are obviously major procedures with significant risks. In the present paper a simple and effective alternative is described.

Method

The procedure can be performed under local or light general anaesthesia. The cyst is opened by means of a 0.5 cm incision on the overlying skin. A small forceps is forced through the base of the cyst and through the masseter into the mouth. With the help of this forceps a 5 cm long Jacques catheter (EG 6) is positioned with one end in the sialocele and the other end in the mouth. The tube is secured by suturing it to the

mucosal surface of the cheek. The skin incision is sutured and a compression dressing is applied. The patient is put on oral penicillin and antiseptic mouthwashes. Oral mixed fluids are given for the first 2 days and thereafter a light diet. The tube is removed 3 or 4 days after it stops draining saliva (about 10 days after the insertion).

Discussion

We used this technique in two patients with post-traumatic sialoceles. In both cases a conservative approach consisting of repeated aspirations and compression over a period of 8–10 weeks had failed. Because of thinning of the overlying skin and imminent external rupture, it was decided to perform the operation described above. The tube drained saliva for 5 or 6 days and then stopped. The sialocele disappeared completely and the artificial internal orifice closed within days of the tube being removed. Three months later there was no recurrence and the cosmetic results were excellent. A parotid scan showed significant atrophy of the affected parotid.

In conclusion we have described a simple technique of managing traumatic sialoceles resistant to conservative treatment. It is easy, effective and can be performed under local anaesthesia.

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