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**Case Report**

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# Sequestered plunging ranula: a case report and literature review

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## ABSTRACT

Ranula are swellings arising from the sublingual or submandibular salivary glands either due to mucus extravasation or the formation of mucus retention cysts. They are located on the floor of the mouth, generally have an insidious course and remain asymptomatic unless they grow very large or develop secondary complications. Occasionally, by dividing through cervical facial planes, they present as swellings in the neck, aptly called plunging ranulas. Plunging ranulas, therefore, have an intra-oral component and an extra-oral component; however rarely, the intra-oral component resolves leaving behind an isolated swelling in the neck, making the diagnosis a clinical challenge.

**Keywords:** Ranula, Plunging ranula, Salivary gland, Sublingual gland, Submandibular gland, Neck swellings, Retention cyst, Oral surgery, Cervical fascia

## INTRODUCTION

Ranula are diffuse swellings located in the floor of the mouth and result from mucus extravasation or the formation of a retention cyst, the former being the more usual pathology. These swellings originate from the sublingual or submandibular salivary glands.[1] Named because of their resemblance of a "frogs belly" (Latin, rana = frog), they may present at any age, are most common in the second decade and exhibit a slight female preponderance.[2]

Occasionally, by dividing through the facial planes, the ranula finds its way into the neck, and gains the name "plunging ranula", the synonyms include 'deep', 'diving' and 'cervical' ranulas. These swellings in the neck either coexist with their source, the oral swelling, or there is a history of the oral swelling being treated earlier, and thus, the clinical diagnosis is straightforward. Rarely, the extra-oral component exists on its own, without the parent swelling in the floor of the mouth. The lack of an oral connection makes the neck swelling a challenge to diagnose, owing to the multitude of differential diagnoses that apply. These isolated or sequestered plunging ranulas are a rare occurrence and clinical challenge.[3]

## CASE REPORT

A 12-year-old male patient presented with a swelling beneath the right jaw which was present since one month. It was insidious in onset and slowly progressive in size and unassociated with pain, difficulty in chewing or swallowing or fever.

Examination revealed an elliptical, soft, cystic swelling in the right submandibular region measuring 3×2×2 cm. It was non-tender, freely mobile, displayed no inflammatory signs and did not move with tongue protrusion. There was no clinically significant loco-regional lymphadenopathy. Routine blood investigations were within normal limits.

Ultrasound of the neck revealed a right para-median, well-defined, hypo-echoic, cystic lesion in the intramuscular plane measuring 3.3×1.1×2.5 cm with low level internal debris with suspicious deeper oral

extension, prompting the consideration of thyroglossal cyst and ranula as possible differentials.

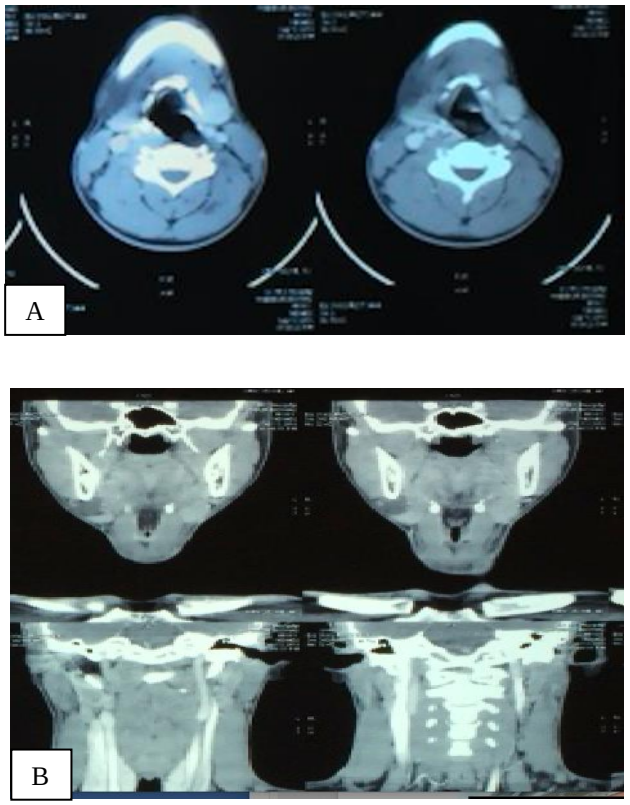


**Figure 1: CT findings revealing plunging ranula: right para-median hypo-echoic mass [A: axial, B: coronal views] with connectivity to the ipsilateral sub-lingual space [C: sagittal view].**

Computed tomography (CT) of the neck with contrast revealed a cystic density (20 Hounsfield units) measuring 4.1×1.4×1.7 cm, with indiscernible walls, showing no significant enhancement post contrast study, located in the right submandibular region, antero-lateral to the right submandibular gland and showing communication with the right sub-lingual space in the antero-lateral aspect, suggestive of a simple plunging ranula.

After an unremarkable pre-operative assessment and marking of the swelling (Figure 2), surgery was performed with the patient under general anesthesia and positioned supine. Aspiration into the swelling revealed mucoid content (Figure 3). A 4 cm skin incision was made along the long-axis of the swelling, corresponding to the skin crease and deepened, opening the platysma. The submandibular gland, its duct and the marginal mandibular nerve were identified and preserved. With no traceable connections, the cyst was excised in to-to (Figure 4), and the wound closed in layers after the placement of a suction drain (Figure 5).

The post-operative period was uneventful, and there was no recurrence of swelling at 6 months of follow-up.

**Figure 2: Pre-operative marking of the swelling.**

**Figure 3: Mucoid aspirate from the swelling.**

**Figure 4: Specimen of extracted swelling.**

**Figure 5: Post-operative image with suction drain.**

## DISCUSSION

Arising from sublingual or submandibular salivary glands as organized extravagated mucus or in the form of retention cysts, ranulas have a prevalence of 0.2 per 1000 population and account for 6% of all silo-cysts. Less than 1-10% of these are true retention cysts, and less than a quarter of these true retention cysts would potentially develop into plunging ranulas. Plunging ranulas have two components, an intra-oral (primary) swelling and an extra-oral (secondary) swelling, usually located in the neck. In very rare instances the intra-oral component is not found, leaving the clinical picture of an isolated neck swelling, opening up the clinical diagnosis to include a multitude of differential diagnoses.[3]

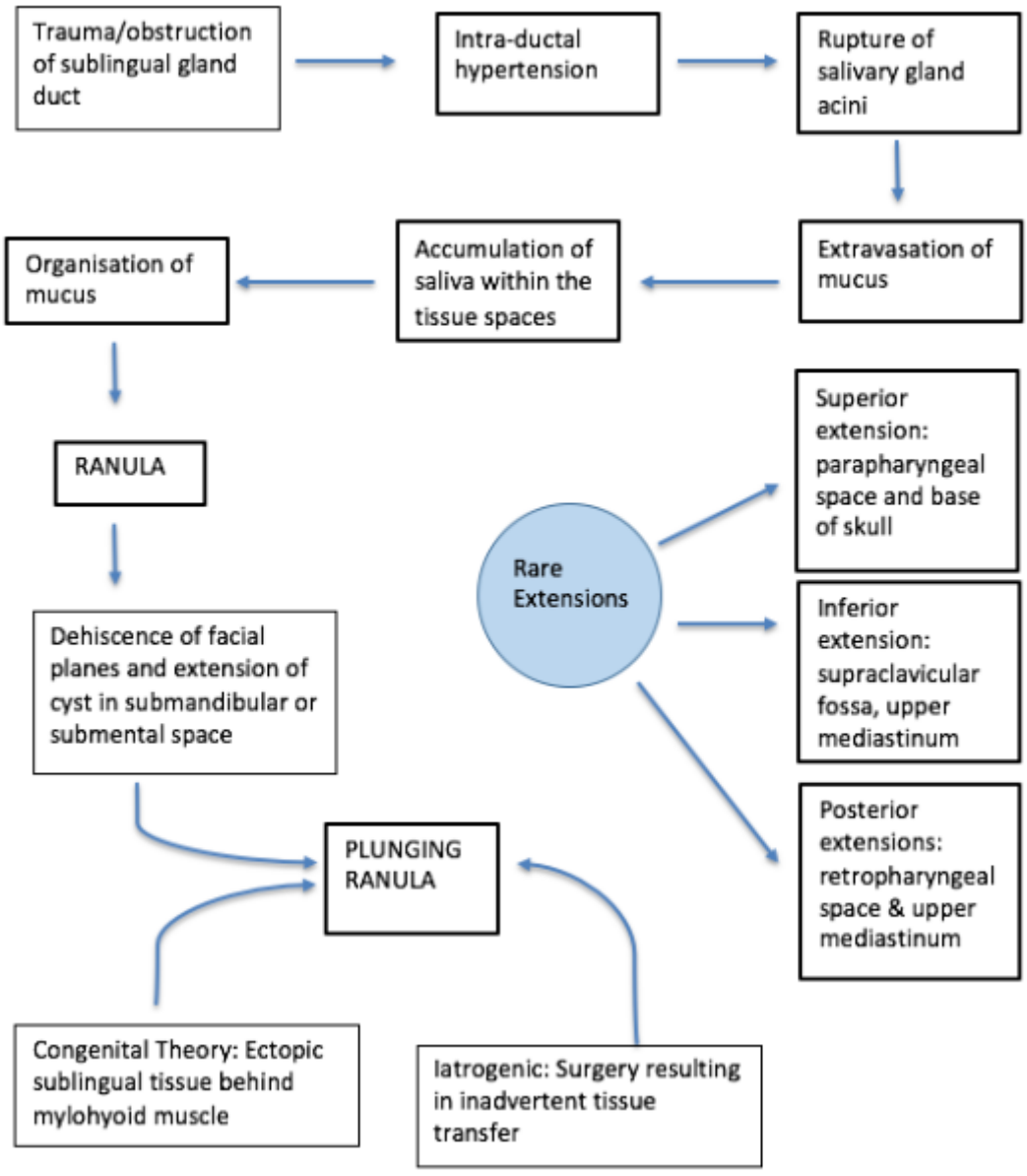


**Figure 6: Pathogenesis of plunging ranula.**

**Table 1: Differential diagnosis of isolated plunging ranula.[3,7,9.]**

| Type of lesion | Example |
|---|---|
| **Neoplastic lesions** | Secondary tumor deposits in cervical nodes<br>Primary lymphomas<br>Lymphangioma |
| **Congenital lesions** | Branchial cleft cyst<br>Thyroglossal cleft cyst<br>Cystic hygroma |
| **Benign lesions** | Lipoma<br>Epidermoid cyst |
| **Post-traumatic lesions** | Hematoma<br>Larygocoele<br>Dermoid cyst |
| **Infectious cervical lymphadenopathy** | Epstein-Barr virus mononucleosis<br>Bacterial infection (draining)<br>Cat-scratch disease |
| **Granulomatous lesions** | Sarcoidosis<br>Tuberculosis |

The pathogenesis of the formation of a plunging ranula is given in Figure 6. [4-7]

The pathogenesis of plunging ranula (Figure 6) may be as extension of a simple ranula, congenital in origin via ectopic cell rests or iatrogenic in nature due to transposition of tissue during surgery involving the salivary structures. Extensive extension has been reported in rare occurrences, producing pressure symptoms like dysphagia and airway obstruction.[4-7] The current consensus is that submandibular glands are involved by extension of a plunging ranula into it and engulfing it, rather than being an intrinsic pathology of the gland.[3]

Diagnosis of a clinically isolated neck swelling as a ranula requires demonstration of communicating cysts which is best achieved by sialography. Injection of contrast into the sublingual space followed by a roentgenogram and ultrasonography are less sensitive techniques. On CT, ranulas are ovoid cysts with homogenous core of 10-20 HU with no clear cyst wall, and extensions from the primary swelling can be identified in case of plunging ranulas.[3] Similar findings were encountered in our case. On magnetic resonance imaging (MRI), it has low T1-weighted and high T-2 weighted signal intensity due to its high water content.[3]

Aspiration cytology reveals mucin and muciphages and amylase content confirms salivary origin.[4] The histopathological picture is that of spilled mucin surrounded by granulation tissue, foamy histiocytes and a mixture of inflammatory cells.[8]

A wide-range of differential diagnoses exist (Table 1) when the plunging ranula is sequestered, including malignancy and hence it is imperative that the diagnosis is thoroughly evaluated.[3,7,9]

Complete surgical excision is the definitive treatment. Other methods such marsupialization and sclerotherapy are associated with higher rates of recurrence.[10] Care must be taken to prevent trauma to the marginal mandibular nerve and histopathological examination of the specimen is essential as it be harboring occult malignancy.[3]

## CONCLUSION

Plunging ranulas are uncommon swelling arising from the sublingual gland that have wandered away from their origin, usually to the submandibular space or rarely, further down the neck. The communication between the intra-oral component and the neck swelling allows for a straight-forward clinical diagnosis. In rare cases, the intra-oral component is absent, leaving behind an isolated swelling in the neck. The differential diagnosis of neck swellings is vast and includes malignancies as well as infections of the lymph nodes and tuberculosis. Thus, sequestered ranula should also be considered amongst the differential diagnosis of neck swellings even when there is no identifiable intra-oral component.

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