

A Rare Case of a Large Plunging Ranula with Cervical Extension: Imaging, Diagnosis, and Management

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Abstract. A plunging ranula is a mucous extravasation cyst appearing as a swelling in the submental and submandibular regions. We describe a rare case of massive deep diving cervical ranula involving multiple tissue spaces and extending to the upper mediastinum. The unusual course of events following surgical excision of the submandibular gland is presented. A Computerized Tomography (CT) scan revealed the true extent of the lesion and its relationship to the surrounding structures. Other imaging techniques and diagnostic tests are discussed and relevant literature is reviewed.

Introduction

A ranula is an extravasation pseudocyst arising from the sublingual salivary gland. The classic ranula presents as a blue-domed, translucent swelling in the floor of the mouth. The term ranula is derived from the Latin word *rana*, meaning frog, and describes a blue translucent swelling in the floor of the mouth reminiscent of the underbelly of a frog.

A plunging ranula manifests itself as a swelling in the neck (Davison *et al.*, 1998). Plunging ranula may rarely appear as a submandibular mass without visible intraoral involvement, making the diagnosis difficult (Zafarulla, 1986).

We describe a case of massive deep diving cervical ranula involving multiple tissue spaces and extending to the upper mediastinum. Other imaging techniques and diagnostic tests are discussed and relevant literature is reviewed.

Case report

A 36-year-old Caucasian came to the oral and maxillofacial surgery clinic for management of a submandibular and neck swelling (Fig. 1, A, B and C). The patient had previously presented with sialolithiasis of the right submandibular gland about 2 years previously. An otolaryngologist at another hospital has surgically excised the right

submandibular gland. On this occasion, the swelling had been present for several weeks and did not improve with antibiotics. His medical history was unremarkable.

Clinically, a soft and fluctuant bluish swelling was noted involving the floor of the mouth, the submental, and the submandibular and lower neck regions and extending to the upper mediastinum. The swelling had remained stable in size and did not change at meal times. Scarring on the floor of the mouth was noted. Radiographic examination by panoramic and lower occlusal films was unremarkable. An ultrasound scan had previously been carried out at another hospital. This was reported to show a large, simple cystic collection in the floor of the mouth that appeared to be associated with the right submandibular gland, and the submandibular duct was seen to be dilated. Under ultrasound guidance, 100 mL of straw-colored thick fluid was aspirated for cytological investigation. The specimen revealed numerous macrophages but no epithelial cells and was regarded as salivary secretion with inadequate diagnostic features. The swelling gradually increased in size, and the floor of the mouth was raised when reviewed 2 weeks later.

From discussion with the previous surgeon, it was suspected that the lesion arose from remnant of the right submandibular gland so sialography of the gland



Figs. 1. A, B and C. Clinical photographs of patient with submandibular and neck swelling.

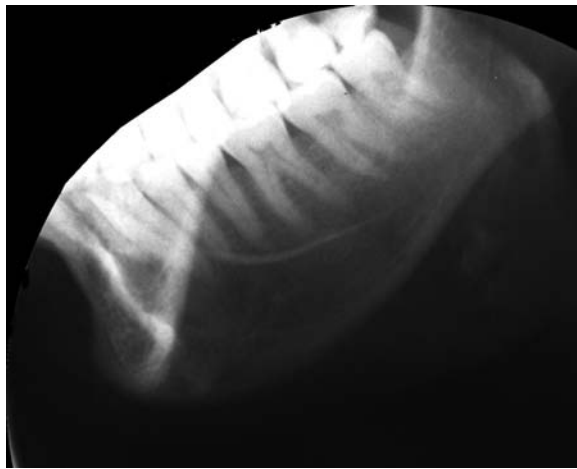


Fig. 2. Sialography of the right submandibular gland showed a dead-end main duct.

was performed and showed a dead-end right submandibular gland main duct and delayed evacuation (Fig. 2). Scintigraphy (radioisotope imaging) by ^{99m}Tc -pertichnetate showed no uptake in the right submandibular gland area and no change in uptake when stimulated with lemon juice (Fig. 3).

Computerized tomography (CT) scan of the neck was performed. Direct axial and reconstructed sagittal and coronal sections were obtained in the soft tissue window with and without intravenous contrast agent. These images demonstrated an extensive right-side round unilocular well defined superficial radiolucent

lesion superomedial to the mylohyoid muscle (Fig. 4), the lesion occupied the whole sublingual space on the right side. It extended posteriorly and inferiorly behind the mylohyoid to occupy most of the submandibular space. Anteriorly, it extended to the submental region. From the submandibular space, it extended lateral to the body of the mandible; it was bounded posteriorly by the masseter muscle. Medially and posteriorly, the lesion extended to the anterior parapharyngeal space and extended along the medial pterygoid muscle. Inferoposteriorly, it extended between the mandibular angle and retromandibular vein extending to the upper mediastinum. The lesion was considered to be an extensive plunging ranula from the right sublingual gland. The greatest dimensions that were measured in CT sections were 8.22 cm from most lateral portion from the right sternocleidomastoid muscle to midline (Fig. 4A), 14 cm superoinferiorly from the floor of the mouth to the right clavicle (Fig. 4B) and 4.96 cm from most superficial to the deepest extension at the right common carotid artery (Fig. 4C).

Under endotracheal general anesthesia, an incision was made in the right floor of the mouth parallel to the submandibular duct. The right sublingual gland was dissected free. About 80 mL of thick straw-colored fluid was drained and the swelling was decompressed. The right sublingual gland was excised. A drain was inserted into the cavity and sutured to the wound margins. The patient made an uneventful recovery and was discharged

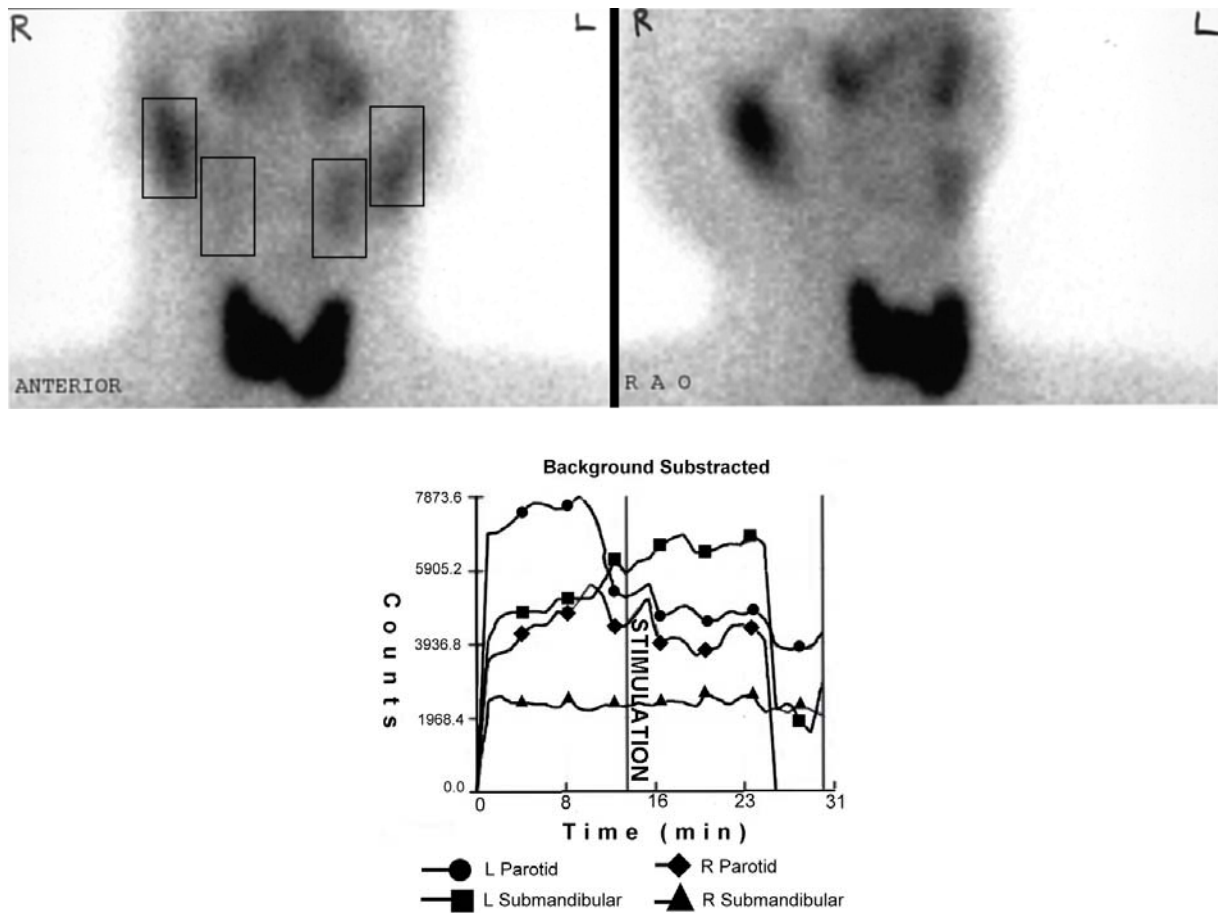


Fig. 3. ^{99m}Tc -pertchnetate scintigraphy (radioisotope imaging) showed no uptake in the right submandibular gland area and no change in uptake when stimulated with lemon juice.

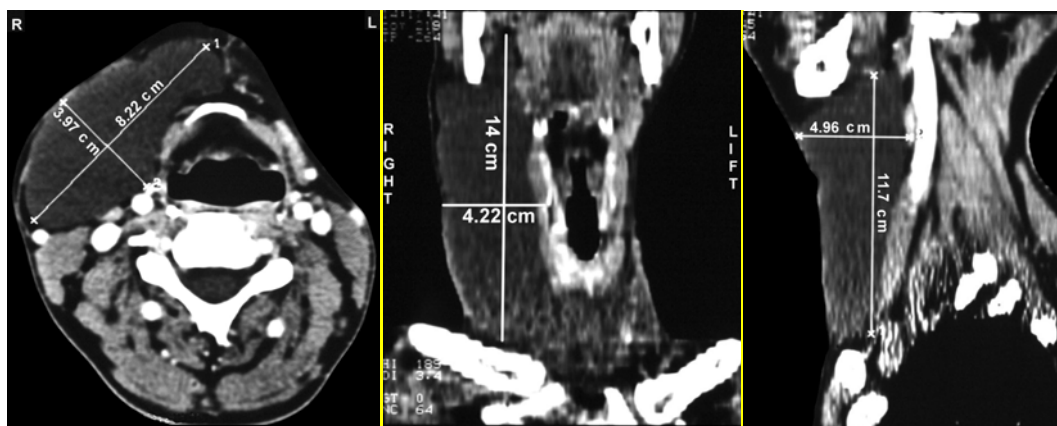


Fig. 4. Direct axial CT section at the level of hyoid bone (A) and reconstructed coronal CT section at the level of mandibular angle and larynx (B) and sagittal CT section at the level of right common carotid artery (C) all with IV contrast agent.

home 2 days postoperatively with the drain in situ. When the patient was seen one week later for follow up, the swelling was completely resolved and the drain was removed. The contour of the neck had returned to normal, and the patient had good tongue mobility and no dysesthesia. Healing was complete and without any signs of recurrence 6 months postoperatively.

Discussion

Massive deep diving cervical ranulas involving multiple tissue spaces and extending to the upper mediastinum are rare (Zhao *et al.*, 2004; Langlois and Kolhe, 1992; Chidzonga and Mahomva, 2007). The clinical diagnosis of this case was difficult because the initial presentation was a large low neck swelling. The floor of the mouth became raised only at a later date. The details of a past surgical operation for a cystic swelling in the right floor of the mouth 2 years previously were unclear, although the operation was most likely to have been removal of the right submandibular gland due to sialolithiasis. Reported plunging ranulas are mostly only 4-10 cm in size and are usually limited to the submandibular space.

Diagnostic imaging of ranulas

Computerized Tomography: ranulas on CT scanning are described as cystic masses in the submandibular or parapharyngeal space that extend into or about the sublingual space. On CT scanning, they are noted to be sharply demarcated lesions of low attenuation that conform to their local fascial boundaries. They are generally unilocular in nature. With the exception of a sublingual epidermoid, the appearance of a simple ranula on CT scanning is distinctive (Shelley *et al.*, 2002). Plunging ranulas are occasionally noted on CT scanning to have a small tail extending into the sublingual space. This finding is almost pathognomonic for plunging ranulas. If this is absent, the presence of a homogeneous cyst in the submandibular or parapharyngeal space that abuts the sublingual space is highly indicative of a plunging ranula (Coit *et al.*, 1987). The extensive nature of our case is well illustrated and documented by CT.

Magnetic Resonance Imaging: MRI is the most sensitive imaging study to evaluate the sublingual gland and its pathologic states. On T1-weighted MRI, the gland appears as an area of intermediate signal intensity, lower than adjacent fat but higher than muscle. T2-weighted images help discriminate cysts from surrounding normal structures (Sumi *et al.*, 1990; Silverstein *et al.*, 1990).

Ultrasonography: Sublingual glands and their

pathologic states are difficult to visualize on ultrasonography because of their location (Shelley *et al.*, 2002).

Diagnostic procedures

Needle aspiration: Analysis of fluid from ranulas demonstrates mucus with prominent histiocytes. The biochemistry of this fluid shows high amylase and protein content.

Differential diagnosis

Lymphadenopathy, cystic hygroma, pleomorphic adenoma, abscess and thyroglossal duct cyst should be considered in the differential diagnosis.

Management of ranulas

Simple marsupialization is the oldest and most widely reported treatment for ranulas. It involves unroofing the cyst and tacking the edges of the cyst to adjacent tissue. Failure rates of marsupialization are high (Baurmash, 1992; Chidzonga and Mahomva, 2007; Yoshimura *et al.*, 1995). Inferior compression on the cyst from the tongue leads to premature closure of the opened cyst. This increases the risk of the cyst recurring. Packing the cyst cavity with gauze for 7-10 days improves the success rate. In one report, 11 of 12 patients had resolution of their ranula with marsupialization and subsequent packing of the cavity.

The criterion standard for treatment of ranulas is excision of the sublingual gland. This removes the source of the mucus and thus significantly decreases the risk for recurrence. A review of 580 patients with ranulas and plunging ranulas found that recurrence rates varied greatly depending on the surgical method chosen. Marsupialization, excision of the ranula alone, and excision of the sublingual gland combined with the ranula resulted in recurrence rates of 66.67%, 57.69%, and 1.20%, respectively (Zhao *et al.*, 2004).

Intraoral excision of the ipsilateral sublingual gland and partial pseudocyst is an effective and safe method for the treatment of the plunging ranula (Yoshimura *et al.*, 1995; Zhi *et al.*, 2009).

Outcome and prognosis

The risk for recurrence when the sublingual gland is not excised has been reported to be in excess of 50%. This rate drops to as low as 2% if the gland is excised (Zhao *et al.*, 2004; Yoshimura *et al.*, 1995; Zhi *et al.*, 2009). Because the risk to adjacent structures is higher for gland-excising procedures (Zhao *et al.*, 2005), a trial of less-invasive procedures is advocated by some such as carbon dioxide laser

(Mintz *et al.*, 1994), low doses radiation therapy (Shimm *et al.*, 1992) and injection Sclerosing agents (Fukase *et al.*, 2003).

Obtaining a specimen for histopathological examination is essential, not only for histologic confirmation but also because the presence of squamous cell carcinoma arising in the cyst wall of a ranula and papillary cystadenocarcinoma of the sublingual gland presenting as a ranula have been reported (Ali *et al.*, 1990; Danford *et al.*, 1992).

Conclusion

We describe a rare case of an extensive plunging ranula. The patient gave a history of a sialolithiasis being previously treated by gland removal, and he subsequently went on to develop a massive plunging ranula. The true extent of this lesion and its relationship to the surrounding anatomy was well illustrated by CT. Despite its extensive ramifications, the lesion was still successfully managed by simple intraoral removal of the sublingual salivary gland, along with drainage of the pseudocyst contents. We feel that this case illustrates well and supports the theory that plunging ranula, however extensive, should be managed by simple intraoral excision of the sublingual salivary gland and drainage of the contents of the lesion.

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. الضفدعية الغاطسة هو كيس من ارتشاح مخاطي يظهر كانتفاخ تحت الذقن وتحت الفك السفلي. نصف حالة نادرة لضفدعية عنقية غاطسة عميقة شملت عدة فراغات نسيجية في العنق وامتدت إلى أعلى منتصف الصدر. تم وصف التسلسل غير المتوقع للأحداث بعد الاستئصال الجراحي للغدة اللعابية تحت الفك. التصوير الطبقي الحاسوبي أظهر الامتداد الفعلي لهذا التجمع وعلاقته بالأنسجة والأعضاء المجاورة. تم مناقشة وسائل التصوير والتشخيص والعلاج لمثل هذه الحالة ومراجعة الأدبيات العلمية ذات الصلة.