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Sublingual-plunging ranula as a complication of supraomohyoid neck dissection

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ABSTRACT

Ranulas are rare cystic lesions resulting from damage or rupture of one or more of the ducts of the sublingual gland, that lead to mucus extravasation or dilatation of the gland's duct. Extravasation cysts are more common than retention cysts. We present a case of a 45-year-old male with a squamous cell carcinoma of the ventral surface of the tongue that was treated with excision of the oral lesion and bilateral supraomohyoid neck dissection without supplementary radiotherapy. A left myocutaneous platysma flap was raised for defect closure. Ten months postoperatively he presented complaining of swelling of the right submandibular region. The diagnosis, based on his medical anamnesis and the CT imaging, was a sublingual-plunging ranula. It is postulated that the ranula resulted from damage to the ducts of the sublingual gland during selective neck dissection. One year postoperatively there are no signs of recurrence either of the ranula or of the cancer. We suggest that sublingual gland excision and intraoral cyst marsupialization is a logical treatment for sublingual-plunging ranulas.

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1. Introduction

Ranulas are cystic lesions originating mainly from the sublingual gland and rarely from the submandibular gland. They often protrude into the floor of the mouth, having only an oral component. Ranulas that are located beyond the mylohyoid muscle, termed "plunging ranulas" and those possessing both an oral and a cervical component, called "sublingual-plunging ranulas", are very rare compared to sublingual ones.¹

These lesions are related to an injury of one or more of the ducts of the sublingual gland that leads to mucus extravasation than to mucus retention due to duct obstruction. The development of ranulas as a complication of surgical interventions was reported in the literature after sialolith removal from the submandibular gland and submandibular gland duct transposition.^{2,3}

We report a case of a sublingual-plunging ranula that developed after supraomohyoid neck dissection. The purpose of the present report is threefold: to provide evidence in support of the extravasation theory for the pathogenesis of ranulas, to provide differential diagnosis and to place emphasis on the possibility of causing damage to the sublingual gland during selective neck dissection.

2. Presentation of case

A 45-year-old male was referred to the outpatient department of the Oral and Maxillofacial Surgery Clinic. He had an

ulcerous lesion on the midline of the ventral surface of the tongue that persisted over the last five years, showing morphological alterations in the last six months. The biopsy sample revealed a squamous cell carcinoma with moderate to high differentiation.

Local cancer resection and bilateral selective neck dissection (SND I–III) were performed. The defect was reconstructed using a left superiorly based platysma flap with arterial blood supply via the submental artery that was preserved during SND. The skin paddle was incorporated in the neck dissection incision. During the procedure attention was paid not to cause damage to the sublingual gland.

Histopathological investigation of the tissue specimens obtained intraoperatively, revealed an infiltrative squamous cell carcinoma with moderate to high differentiation. Lymph nodes were negative for neoplasia and showed signs of lymphadenitis. Muscle fibres, minor salivary glands, vessels and nerves did not show signs of infiltration. Based on the results of the histopathological investigation it was decided that radio- and chemotherapy were not necessary.

Ten months postoperatively, the patient came back to the department complaining of swelling of the right cheek and submandibular region. Physical investigation revealed the presence of a painless, soft, cystic, bulgy mass extending from the right submandibular to the cervical region. Intraoral examination revealed swelling of the right side of the floor of the mouth (Fig. 1). Computer Tomography (CT) imaging confirmed the presence of a large cyst at the right submandibular-lateral pharyngeal area with a maximum diameter of 4.44 cm on the axial view and 5.34 cm on the coronal view, extending into the sublingual space (Fig. 2).

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Fig. 1. Intraoral view. Sublingual swelling protruding into the floor of the mouth.

The lesion was treated with excision of the sublingual gland via a submandibular incision along the scar that resulted from SND. The exploration of the neck showed that the ranula was situated between the platysma and the mylohyoid muscle, becoming narrower when passing through a hiatus of the mylohyoid muscle. The ranula was mobilized from the bellies of the digastric muscle and from the hyoglossus muscle. During mobilization the wall of the ranula was accidentally ruptured and excised simultaneously with the sublingual gland through the cervical incision. A cervical and not an oral approach for the excision of the sublingual gland was chosen in order to ensure removal of even ectopic tissue. The intraoral component of the ranula was marsupialized with iodiform gauze and the submandibular incision was closed in layers. It was decided not to completely excise the ranula because of risking damage to the adjacent structures due to the wide extension of the lesion. It should be pointed out that the ranula was situated on the right side of the floor of the mouth, whereas the platysma flap for the reconstruction of the defect of the tongue and left side of the floor of the mouth was raised on the left.

The histopathological evaluation of the specimens obtained during surgery revealed a cystic wall of 0.5 cm, surrounded by fibroconnective tissue that presented sublingual gland tissue, a dilated salivary gland duct and a minimal degree of chronic inflammatory infiltration (Figs. 3 and 4). These features are consistent with

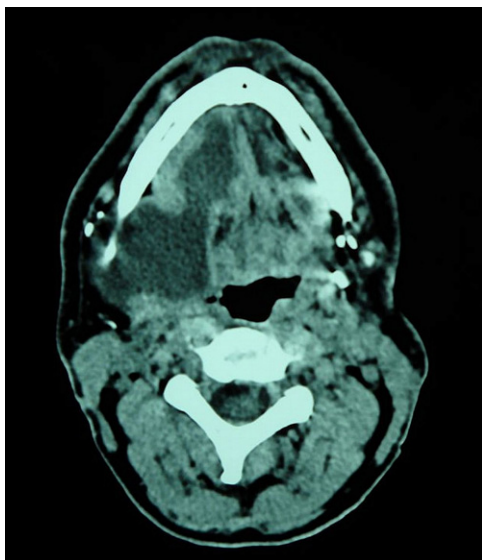


Fig. 2. Computer Tomography (CT), axial view. A low-attenuation collection (mucoid) in the submandibular-lateral pharyngeal region in continuity with the sublingual space.

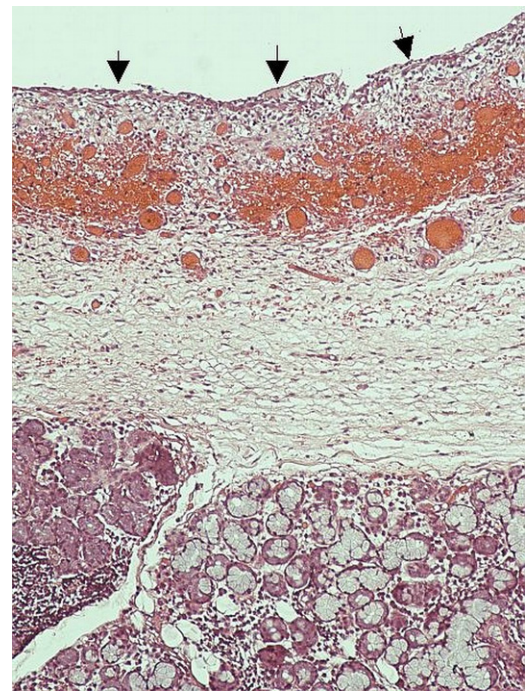


Fig. 3. Cystic wall devoid of epithelium (black arrows), consisting of loose fibroconnective tissue, inflammatory cells and dilated vessels (100x, H + E).

a ranula. Histopathological examination, combined with the CT imaging that revealed extension of the lesion from the sublingual to the submandibular space under the mylohyoid muscle, justifies the diagnosis of a “sublingual-plunging” ranula as a result of saliva extravasation from one of the ducts of the sublingual gland.

After surgery the patient recovered well and showed no signs of paresthesia in the distribution of the lingual nerve. One year after excision of the ranula the patient shows no signs of recurrence either of the ranula or of the tumor.

3. Discussion

The term plunging or cervical ranula refers to a cyst developing from the sublingual gland at the submandibular and upper cervical region.^{1,4,5} The sublingual gland is more vulnerable in developing extravasation cysts, for in contrast to the submandibular and parotid glands it is characterized by permanent mucus excretion, even without the presence of stimuli.⁴ Ranulas are soft cystic

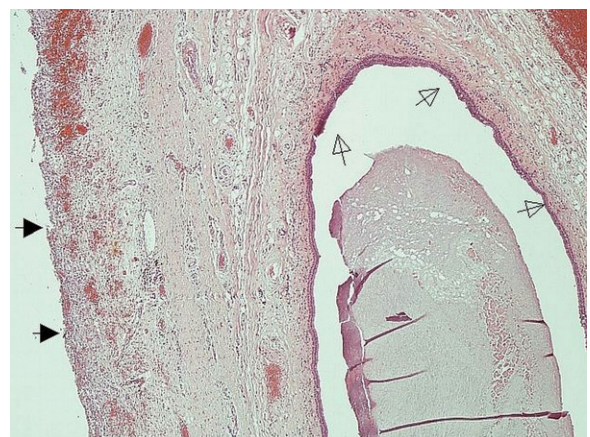


Fig. 4. Dilated salivary gland duct (not filled arrows) and cystic wall devoid of epithelium (filled black arrows) (40x, H + E).

masses that develop mostly unilaterally.¹ Like other benign cysts, epidermoid and branchial cysts that develop in the same region, they are rare.^{4,5} People in their second and third decades of life and those belonging to specific ethnic groups, Maori and Pacific Island Polynesians, are more prone to develop plunging ranulas, suggesting that they may also be congenital in some cases.^{4,5}

Ranulas can anatomically be divided into three main groups: (a) sublingual, above the mylohyoid muscle, (b) plunging, under the mylohyoid muscle and above the hyoid bone (suprahyoid ranulas) and (c) sublingual-plunging with an oral and a cervical component.^{1,6}

There are two main theories for the development of ranulas. One theory states that they are a result of mucus escape from the sublingual gland, whereas the second supports that ranulas result from retention of saliva, both as a result of damage or rupture of a duct of the sublingual gland.⁷ Current opinion supports that ranulas result from mucus extravasation, for they are mainly devoid of lining epithelium.⁶

The extension of ranulas to the submandibular and upper cervical region is related to the herniation of the mylohyoid muscle by the sublingual gland.⁸ Projections of the gland through a hiatus between the anterior and the posterior part of the mylohyoid muscle were reported in 45% of cadaver specimens and it shows the clear involvement of this herniation in cervical extension of the ranulas.⁹ In addition, the presence of ectopic sublingual gland tissue, beyond the mylohyoid muscle may be causally related to a plunging ranula and provides evidence for their possible congenital origin.^{6,9}

Surgical interventions have also been implicated in ranula formation. There are reports of plunging ranulas that developed after the excision of a sialolith or transposition of the duct of the submandibular gland.^{2,3}

The diagnosis of a plunging ranula is of clinical significance for there are many benign as well as malignant lesions that have the same appearance during physical examination. In particular, neoplastic and inflammatory lesions of the submandibular and sublingual glands, of the lymph nodes, granulomatous, vascular, nerve or adipose tissue diseases, branchial or thyroglossal duct cysts, dermoid and epidermoid cysts, cystic hygroma and laryngocele could appear as a soft palpable mass of the submandibular region, complicating the diagnosis.⁹ There are no specific tests for the diagnosis of cervical ranulas. Differential diagnosis should be based on the history of the lesion that shows up as a cystic fluctuating lesion, gradually increasing in size. Additionally, the fluid of ranulas consists of a higher salivary amylase and protein content compared to serum, suggesting an origin from the sublingual gland that produces saliva with a higher protein concentration than the submandibular gland.¹⁰

There are many approaches for the management of plunging ranulas. The excision of the sublingual gland either through an intraoral approach or via a submandibular approach is one option.¹⁰ It is advocated that cervical ramifications could be left for they disappear after the removal of the gland.¹⁰ Other approaches include: drainage of the ranula or marsupialization, drainage of the ranula and excision of the sublingual gland, excision of the ranula and the sublingual gland.^{1,6} The treatment of plunging ranulas must be based on the concept of removing the cause, i.e. the sublingual

gland, in order to prevent recurrence, for the ranula itself does not possess the ability of mucus production. The recurrence rates of ranulas after marsupialization, excision of the ranula and excision of the sublingual gland with or without ranula excision are 66.67%, 57.69% and 1.20%, respectively.¹¹

4. Conclusion

It is postulated that the sublingual-plunging ranula of our patient, resulted from a traumatic injury to the sublingual gland during selective neck dissection. Ranula formation could not be linked to sublingual gland injury during defect closure with platysma flap after cancer resection, for this procedure was contralateral to the side of ranula formation. Ipsilateral sublingual gland excision and intraoral ranula marsupialization is a logical treatment for sublingual-plunging ranulas.

Conflict of interest statement

The authors of this manuscript disclose no financial and personal relationships with other people or organisations that could inappropriately influence (bias) their work.

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Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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