

CASE REPORT

PLUNGING RANULA: A CASE REPORT AND REVIEW OF LITERATURE

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<p>¹Senior Resident ²Second Year Resident ³Professor and HOD ^{4,5}Professor</p>	<p>ABSTRACT</p> <p>Ranula is a diffuse swelling in the floor of the mouth caused by a mucous retention cyst derived from the major sublingual or submandibular salivary glands. The word was taken from rana; a Latin word meaning frog because the typical swelling in the floor of mouth resembles the underbelly of a frog. They are most common in the second decade of life and in females. We are presenting a male patient with plunging ranula, patient had under gone for excision of the sublingual gland treatment. Due to rare incidence of male patient with plunging ranula and the diagnosis of a plunging ranula has clinical significance for there are many benign as well as malignant lesions that have the same appearance during physical examination</p> <p>Key words: ranula, plunging, sublingual, glands, salivary, swelling, surgery, cyst.</p>
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INTRODUCTION

Periodontitis refers to the inflammation of the supporting tissues of the teeth which cause progressive destruction leading to bone loss and ultimate tooth loss¹. Periodontitis is of multifactorial origin with bacterial plaque as primary etiological factor².

Periodontitis is caused by an imbalance between pathogenic microbes and the local and systemic host defence mechanisms. This imbalance results in alteration of synthesis and degradation of collagen fibrils and alveolar bone by the release of various inflammatory markers³.

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INTRODUCTION

Ranulas are benign pseudocystic lesions filled with salivary secretions that present as painless mobile masses in the floor of mouth or neck. First described centuries ago by Hippocrates as a local inflammatory process, the etiology of ranulas was not made clear until modern advancements in microscopy. Extralymphatic theories of origin were entertained as late as the 1960s, as many still thought that ranulas were embryologic in origin and similar to branchial cleft cysts. Suzanne and von Hippel concluded ultimately that ranulas arose from the sublingual gland and therefore treatment should involve removal of the sublingual gland and the ranula. The pathogenesis remained controversial until the 1950s when it was found that ranulas were produced by the extravasation of mucous from damaged sublingual ducts into the surrounding connective tissues and the ranula sac lacked a true epithelial lining. In 1962, Whitlock and Summers-Gill treated one case of a plunging ranula with removal of the sublingual gland alone as it was the underlying source of salivary extravasation. Despite recognition that the sublingual glands are the source of ranulas, a plethora of treatments have been described in the literature over the past 50 years including incision and drainage, surgical or laser-assisted marsupialization, sclerotherapy, removal of the sublingual gland and ranula, and removal of the ranula sac alone. Simple ranulas are restricted to the sublingual space and are surgically managed with simple intraoral techniques. Plunging ranulas, however, extend beyond the sublingual space and can be more challenging as many head and neck surgeons have attempted resection of the ranula through deep intraoral dissection through the mylohyoid or external

dissection through a cervical incision. Moreover, directly excising a plunging ranula without removing the sublingual gland is associated with a high rate of recurrence. Sublingual glands (SLGs) are the smallest of the paired major salivary glands, weighing about 2 g, and shaped like a flattened almond measuring about 2.5 cm antero-posteriorly, each gland has a row of about 12–20 short ducts that open independently along the summit of the sublingual fold in the floor of the mouth, obstruction of one of these ducts results in formation of a mucous retention cyst in the sublingual space, termed simple ranula, ranula is a blue-domed, cystic swelling of the floor of the mouth, further accumulation of secretions with time results in extension along sublingual space anteriorly and posteriorly.

CASE REPORT

In 2018 a 40-year male patient from Saurashtra (Gujarat) reported to the department of maxillo-facial surgery with the complaint of swelling at the neck since 1 month. On extraoral examination, inspeitory findings show there is a well-defined oval shape approx. 2*1.5 cm in size swelling at the submandibular region overlying skin is normal, no any sinus fistula present. All the inspeitory findings are conformed by palpation, swelling is non-tender, fluctuant, soft in nature and no any fluid discharge is present on pressure.

On intraoral examination, inspeitory findings show there is a well-defined oval shape approx. 2X2 cm in size swelling present on the floor of the mouth overlying mucosa is red, frenum attachment is deviated on the left side. All inspeitory findings are conformed by palpation, swelling is nontender, fluctuant, soft in nature and no any fluid discharge present on pressure.

On USG report – there is a presence of hypoechoic lesion of size of 57 X 38 mm with deeper extension seen in midline of neck with moving internal echoes and few foci of calcification.



Fig 1: frontal view



Fig 2: Submentovertex view



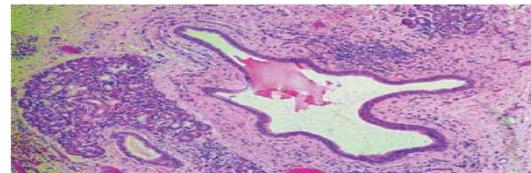
Fig. 3: Intraoral photograph showing swelling on ,sublingual and submandibular area accompaied by floor of mouth



Fig 4: Excisional mass

Based upon the clinicoradiological and USG findings, a tentative diagnosis of plunging ranula was made with a differential diagnosis of dermoid and epidermoid cyst, thyroglossal duct cyst,

cystic hygroma, and lymphadenopathy. The excision of the lesion was done via cervical approach under general anaesthesia and the tissue was subjected to histopathological evaluation Patient was planned for surgery. He was operated under general anaesthesia for excision of sublingual gland. Patient followed uneventful post op course and was discharged after 5 days. Patient is followingup monthly for period of 2 years. Histopathological picture of the excised lesion showing mucin collection in the lumen lined by connective tissue with inflammatory cells.



DISCUSSION

Ranula develops from extravasation of mucus after trauma to the sublingual gland or obstruction of the ducts.[6] Ranula can occur at any age. It has been reported from 2 to 61 years of age with a slight female preponderance.[8] Regarding the Patel *et al.* study a total of 26 ranulas were identified at their institution over an 18-year period. There were 54% male and 46% female patients with an average age of 25.6 and a median age of 26. Of the 26 ranulas identified, 16 were oral (62%) and 10 were plunging (38%).[9]

The etiology is unknown, but it has been described in association with congenital anomalies, trauma, and disease of the sublingual gland.[8]

The initial stage is a traumatic rupture of the excretory duct and the second stage is the extravasation and subsequent accumulation of saliva within the tissue, as shown by experimental studies.[4]

When these extravasation cysts extend into the submandibular or submental space, they are called plunging ranula.

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. 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. Surgical interventions have also been implicated in ranula formation.

According to Gupta *et al.*, Kalra *et al.*, and Zhao *et al.* studies the cervical ranula appears as an asymptomatic, continuously enlarging mass that may fluctuate in size. Most reported ranulas are 4–10 cm in size. The overlying skin is usually intact. The mass is fluctuant, freely movable, and nontender.[12] The mass is not associated with the thyroid gland or lymph node chains. In some instances, detecting salivary gland herniation of a portion of the sublingual gland through the mylohyoid muscle into the neck may be possible. The mass may not be well defined, but should follow the facial planes of the neck and may extend into the mediastinum. Similar to the oral ranula, the mass tends to cause a lateral swelling; however, it may cross the midline. They have been reported to extend into the submental region, the contralateral neck, the nasopharynx up to the skull base, the retro pharynx and even into the upper mediastinum.[15] Rarely, large-sized ranulas may cause dysphagia or airway obstruction. Sialogram, ultrasonography, Magnetic resonance imaging (MRI), CT, and aspiration cytology can be helpful for diagnosis. Aspiration cytology will show

mucin with muciphages and biochemical analysis will show increase in amylase and protein content. This is diagnostic of the salivary origin.[4]The microscopic appearance of a ranula is similar to that of a mucocele in other locations. On microscopic examination, the mucocele shows an area of spilled mucin surrounded by a granulation tissue response. The inflammation usually includes numerous foamy histiocytes (macrophages). In some cases, a ruptured salivary duct may be identified feeding into the area. The adjacent minor salivary glands often contain a chronic inflammatory cell infiltrate and dilated ducts.[17]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 lymphnodes, 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. 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.[11]Surgery is the main stay for the management of ranulas. These include incision and drainage, excision of ranula, marsupialization, and marsupialization with packing or complete excision of the sublingual gland.[4]Many methods have been used for the treatment of cervical

ranulas and this includes surgical removal of lesion, use of cryosurgery, marsupialization, excision of oral portion and associated sublingual gland or, rarely, submandibular gland. Others are excision of sublingual gland via intraoral approach, and drainage of lesion, and use of cervical approach to excise the lesion sometimes this is combined with excision of sublingual gland [1]. Recurrences have been known to occur even after these treatments [6]. However, when the sublingual gland together with the associated lesion is excised, a low recurrence rate is recorded [16, 17]. The oldest and most widely used therapy for oral ranula is marsupialization; this involves the removal of the roof of the cyst and attachment of its borders to surrounding tissues. This procedure recorded a very high recurrence rate of 61–89% within 6 weeks to 12 months of surgery. Early closure of the surgery site due to compression of the tongue on the cyst has been known to increase the recurrence rate. Therefore, packing the cavity with gauze for 7–10 days reduces the recurrence rate. Marsupialization and filling the cavity with gauze improves the success rate. Micromarsupialization involves the placement of a silk suture (Seton) for a minimum of 7 days during which an epithelial tract forms to allow for mucus drainage between the surface and the underlying salivary glandular tissue. This procedure is simple with minimal to nonexistent minimal to nonexistent; however, its recurrence or treatment failure is the primary complication [11]. Although considered experimental, sclerosing agents have been employed in the treatment of PR [18, 19]. Bleomycin and OK-432 have been successfully used to manage ranulas [19]. Even so, OK-432 has been shown to

collapse and cause adhesion of the pseudocyst wall of a PR [18]. In a study of 32 cases of PR, 31 (97%) achieved a marked decrease in size of lesion with injection of OK-432. While about 50% of all cases experienced local pain or fever, this, however, resolved after some days [11]. Rho et al. thus advocate that sclerotherapy is a safe and potentially curative procedure that may be used as a primary treatment before considering surgery for PR. Carbon dioxide laser has also been used as a treatment options for PR; this has shown good success with a decrease in recurrence rate [20]. In very few patients who cannot endure the stress of surgery, a worthwhile substitute would be the used of radiotherapy, and low doses, from 20–25 grays (Gy), have been known to be effective. The attendant complications of radiotherapy especially xerostomia can be circumvented using of low doses and protecting the contralateral parotid gland with a shield. The danger of malignancies developing as a result of treatment with radiation is very low [5]. In the management of PR, the sublingual gland could be excised either be via the intraoral or the transcervical technique. Excision of the gland through the intraoral method and drainage of the associated cervical content is enough to give cure and is presently considered as the treatment of choice [6]. With the transcervical approach, total excision of the sublingual gland is challenging because this method requires the painstaking division of the mylohyoid muscle in the floor of mouth [11]. However, this method is recommended for review cases and when the PR is large in size [21]. Removal of the gland by a transoral approach after drainage of the cyst has also been advocated. Should this method fail, then the cyst should be excised

via a transcervical approach. Cases restricted to the neck are also best treated by the transcervical approach [11]. Other treatment modalities have also been utilized. Sclerotherapy with OK-432 is a good substitute for surgery.[15] Recurrence was noted in 14.3% and the patient had an average of 1.7 injections. Fukase *et al.*, [16] used a higher concentration in partially regressed cases and had 100% cure rate.

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