

A Cervical Ranula: A Case Report

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ABSTRACT

Ranula refers to a collection of extra-glandular and extra ductal saliva in the floor of the mouth originating from the sublingual salivary gland. It may rarely originate from injury to the submandibular gland (SMG) duct. It is a pseudocyst, as it does not contain an epithelial lining. It classically presents as a soft submucosal swelling in the floor of the mouth. A plunging ranula extends into the submandibular triangle of the neck through a defect in the mylohyoid muscle, or less commonly, by passing behind the posterior edge of the muscle. A ranula may also track posteriorly along tissue planes into the parapharyngeal space. The simplest way to clinch the diagnosis is by examining a needle aspirate of the typically thick, straw coloured saliva. However, the diagnosis can be easily missed if the patient does not have any introral lesion. The purpose of this paper is to present clinical and radiographic findings of a rare case of plunging ranula without any intraoral lesion along with the relevant review of the literature.

INTRODUCTION

A plunging ranula is an extravasation of saliva from the sublingual gland due to trauma or obstruction of the duct. Fluid from the obstructed gland dissects between the fascial planes and muscle of the base of the tongue to the submandibular space. The exact prevalence of plunging ranula is not known, however, these lesions are considered uncommon¹. Because most plunging ranulas either accompany a swelling in the floor of mouth or are associated with a history of treatment of intraoral ranula, it is not difficult to diagnose such a lesion. On the other hand, the plunging ranulas in which there is no clinical evidence of an oral connection, diagnosis becomes a chalenge^[11]. Treatment of a ranula includes marsupialization, excision of the sublingual salivary gland, excision of the ranula with or without excision of the sublingual salivary gland, and sclerotherapy^[11]. Excising the sublingual salivary gland is the key to minimize recurrence^[11].

Case Report:-

A 18 years old female reported with a 12-month history of swelling in left submandibular region. The swelling was completely asymptomatic and there was a history of intermittent change in the size of swelling. The patient gave history of increase in swelling during eating and chewing. The patient visited our institution for definitive management.

The patient was in good health and had no history of any systemic disorder. Family history and personal history unremarkable. On examination, general condition was good and vital signs were stable. On examination, a diffuse, soft, fluctuant, non-tender swelling, about 5x4 cm2 in size, was present in right submandibular region. Overlying skin was normal in colour and temperature. No swelling was evident on intraoral examination. Ultrasonography was inconclusive and hence sialogram was performed On injecting a contrast medium in the sublingual space, we could appreciate the extension of the dye in submandibular space. CT scan (figure 1) showed right submandibular lesion above and behind the submandibular gland.

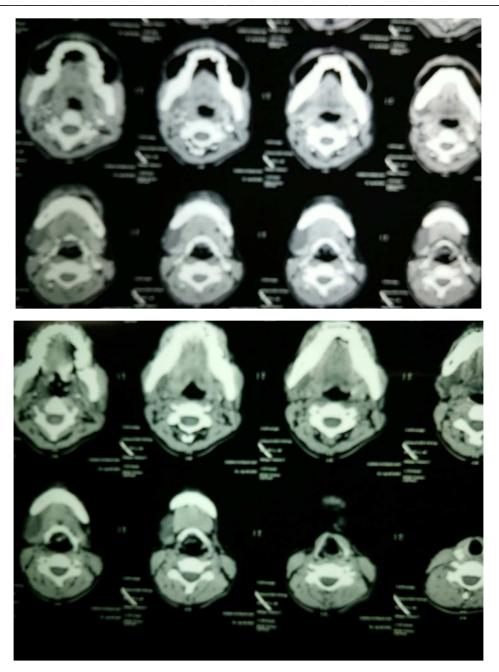


Figure 1: CT Scan : Showing right sided cystic lesion evading submandibular glands

A diagnosis of Cervical Ranula was made as per the clinical and radiological findings and patient was taken for surgery whereby the ranula along with the sublingual glands were resected out was sent for histopathological examination (figure 2).



Figure 2: cystic lesion with Submandibular gland

Histopathological picture of the excised lesion showing mucin collection in the lumen lined by connective tissue with inflammatory cell and confirmed our pre operative diagnosis of Ranula.

DISCUSSION

A ranula by definition is a mucus filled cavity, a mucocele, in the floor of the mouth in relation to the sublingual gland. The name "ranula" has been derived from the Latin word "rana" which means "frog." The swelling resembles a frog's translucent underbelly or air sacs. Ranulas are characteristically large (>2 cm) and appear as a tense, fluctuant and dome-shaped vesicle, sometimes with a blue hue¹. The most common site is the lateral floor of the oral cavity. Plunging ranula occurs when the fluid pressure of the mucin dissects through a perforation in the mylohyoid muscle in the submandibular space.

Ranulas have a prevalence of about 0.2 cases per 1000 persons and accounts for 6% of all oral sialoceles¹. It has been reported from 2 to 61 years of age with a slight female preponderance^[2]. The cervical variant tends to occur a little later in the third decade.

The etiology is unknown, but it has been described in association with congenital anomalies, trauma, and disease of the sublingual gland^[2]. The pathophysiology involved in extravasation is hypertension in the duct due to obstruction leading to acinar rupture in the salivary gland and then extravasation of the mucus^[2]. 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 tissue².

Plunging ranulas arise in the neck by one of the following four mechanisms^[1]. Firstly, the sublingual gland may project through the mylohyoid, or an ectopic sublingual gland may exist on the cervical side of mylohyoid. This may explain the plunging ranulas that exist without an oral component. Secondly, a dehiscence or hiatus in the mylohyoid muscle may occur. This defect is observed along the lateral aspect of the anterior two-thirds of the muscle. Through this defect, the mucin from the sublingual gland may penetrate to the submandibular space. Thirdly, approximately 45% of plunging ranulas occur iatrogenically after surgery to remove oral ranulas or secondary to surgical procedures for sialolith removal, duct transposition and implant placement. Lastly, a duct from

the sublingual gland may join the submandibular gland or its duct, allowing ranulas to form in continuity with the submandibular gland. Therefore, the ranula accesses the neck from behind the mylohyoid muscle.

The cervical ranula appears as an asymptomatic, continuously enlarging mass that may fluctuate in size. Most reported ranulas are 4–10 cm in size¹. Similar to the oral ranula, the mass tends to cause a lateral swelling; however, it may cross the midline. The overlying skin is usually intact. The mass is fluctuant, freely movable, and nontender. In some instances, detecting salivary gland herniation of a portion of the sublingual gland through the mylohyoid muscle into the neck may be possible. Rarely, large-sized ranulas may cause dysphagia or airway obstruction.

In up to 45% of the cases, the patient's first presentation is an oral swelling. Plunging ranulas are associated with oral swelling in 34% of cases. Another 21% of the cases occur without any oral involvement. In the absence of oral swelling, the clinical diagnosis of ranula may be easily missed^[1].

Differential diagnosis of cervical ranula must include thyroglossal duct cyst, branchial cleft cyst, cystic hygroma, submandibular sialadenitis, intramuscular hemangioma, cystic or neoplastic thyroid disease, infectious cervical lymphadenopathy, hematoma, lipoma, laryngocele, and dermoid cyst¹.

Sialogram, ultrasonography, Magnetic resonance imaging (MRI), CT, and aspiration cytology can be helpful for diagnosis².Takimoto suggested a simple radiographic technique for preoperative diagnosis of plunging ranula that involves injecting a contrast media in sublingual space^[1]. A sialogram performed on a patient with a sialocyst reveals smooth displacement of the glandular ducts around the mass. No direct communication with the ductal system is demonstrated. However, the best method of demonstrating a communicating cyst is by sialography^[1]. Ultrasonography is usually inconclusive to study sublingual glands due to their location. CT scan can hep to understand the origin and extent of this cystic lesion. Although a plunging ranula may extend into the submandibular triangle and displace the submandibular gland, it does not intrinsically affect this gland. MRI is the most sensitive study to evaluate the sublingual gland and its states^[1].

Histopathologically, the cervical ranula appears identical to the mucus extravasation phenomenon. Biopsy of the lateral part of the neck may reveal only amorphous material with rare inflammatory cells and predominant histocytes, which stains positive for mucin¹.Biochemical analysis of fluid shows its high amylase and protein content².

In our case, the patient presented with a swelling that was restricted to left submandibular region without any intramural lesion and the patient was completely asymptomatic. On injecting a contrast medium in the sublingual space, we could appreciate the extension of the dye in submandibular space, which was suggestive of a plunging ranula.

Clinicians have been using several different methods for the treatment of cervical ranulas. These include excision of the ranula only, cryosurgery, marsupialization with or without cauterization of the lesion lining, excision of the oral portion of the ranula with the associated sublingual salivary gland or, rarely, the submandibular gland, intraoral excision of the sublingual gland and drainage of the lesion, and excision of the lesion via a cervical approach, sometimes combined with excision of the sublingual gland¹.

The most common complications is the recurrence of the lesion (5.78%) and sensory deficit of the tongue (4.89%), followed by damage of Whartons duct (1.82%). Postoperative hematoma, infection, or dehiscence of the wound were seldom seen³.Excision of the ranula with the associated sublingual salivary gland is the most accepted method with low recurrence rate¹. A biopsy of the cystic wall is recommended not only for histologic confirmation, but also to rule out presence of squamous cell carcinoma arising from the cyst wall and papillary cystadenocarcinoma of the sublingual gland, which may present as ranula.

Besides surgical management, CO2 laser has been used to vaporize ranulas^[3]. In few cases, radiation therapy orintracystic injection of the streptococcal preparation, OK-432, has been used to treat this lesion^[4]. The use of this sclerosing agent as a treatment approach for the cervical ranula is considered experimental. A recent study found orally administered Nickel Gluconate-Mercurius Heel-Potentised Swine Organ Preparations D10/D30/D200, a homotoxicological agent to be an effective treatment modality for ranulas^[5].

CONCLUSION

Though the cases of plunging ranula have been documented with moderate frequency, failure to differentiate the clinical features of oral and plunging ranulas may be a diagnostic pitfall. These lesions may be difficult to differentiate from benign and malignant salivary gland tumors, especially cystadenocarcinoma and mucoepidermoid carcinoma. A case of squamous cell carcinoma in the wall of ranula has also been reported .So, thorough radiological, biochemical, and histopathological investigations should be carried out for all cases of suspected plunging ranulas.

REFERENCES

[1] Loney WW, Jr., Termini S, Sisto J. Plunging ranula formation as a complication of dental implant surgery: a case report. Journal of Oral and Maxillofacial Surgery. 2006;64(8):1204–1208.

[2] Sheikhi M, Jalalian F, Rashidipoor R, Mosavat F. Plunging ranula of the submandibular area. *Dental Research Journal*. 2011;8(Suppl1):S114-S118.

[3] Zhao Y-F, Jia J, Jia Y. Complications associated with surgical management of ranulas. Journal of Oral and Maxillofacial Surgery. 2005;63(1):51–54.

[4] Rho MH, Kim DW, Kwon JS, et al. OK-432 sclerotherapy of plunging ranula in 21 patients: it can be a substitute for surgery. American Journal of Neuroradiology. 2006;27(5):1090–1095.

[5] Garofalo S, Briganti V, Cavallaro S, et al. Nickel Gluconate-Mercurius Heel-Potentised Swine Organ Preparations: a new therapeutical approach for the primary treatment of pediatric ranula and intraoral mucocele. International Journal of Pediatric Otorhinolaryngology. 2007;71(2):247–255.