



Case Report

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Rerouting Wharton's duct - a surgical remedy for intraoral excision of ranula

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Abstract

Ranula is an extravasation pseudocyst that occurs due to trauma or blockage of sublingual glands. The treatment is controversial varying from surgical excision to intralesional sclerotherapy. In this article, the authors have discussed about a case of plunging ranula which was excised intraorally by rerouting Wharton's duct. A 26 years old female presented with a neck and oral swelling to outpatient department. Her fine needle cytology and CT scan were suggestive of a plunging ranula on left side. Because of her cosmetic concerns, intraoral excision of ranula was planned, after duly obtaining consent for the risk of Wharton's duct and lingual nerve injury. Intraoperatively, the cyst was encasing both Wharton's duct hindering further dissection. So, posterior rerouting of Wharton's duct was carried out on both sides. This step helped not only in preserving the ductal integrity but also facilitated easy identification of lingual nerve and sublingual glands. The whole cyst was removed along with the sublingual glands. Postoperative functions of lingual nerve were intact. The authors have discussed about the advantages of rerouting the duct in this procedure along with literature review.

Keywords: Ranula, Wharton's duct, Sublingual gland.

INTRODUCTION

Ranula is an extravasation cyst that occurs due to trauma or blockage of sublingual glands [1]. These extravasation pseudocysts extend into submandibular space and present as a neck swelling, which is known as plunging ranula. The treatment for ranulas is controversial and varies from aspiration of cyst, Marsupialisation, incision and drainage, excision of cyst with/without sublingual gland removal, to sclerotherapy [2-5]. A large cyst requires complete cyst excision along with the sublingual gland and it is widely accepted and practiced by many authors because of its low recurrence rates. Literature describes extraoral/cervical approach for excision of plunging ranulas because of the wide exposure and lesser chances of bleeding and injuring Wharton's duct and Lingual nerve [6]. In this article, we have described a case of plunging ranula, which was removed via intraoral approach by rerouting the Wharton's duct, without damaging lingual nerve. We have also discussed the relevant literature reports about the challenges faced in the management of ranula.

CASE REPORT

A 26 years old female presented to Out Patient Department with complaints of swelling in the floor of mouth for past 6 months. The swelling was insidious and gradually progressive. It was not associated with pain or discharge. Patient also gave history of another swelling in the upper aspect of neck for 4 months. There were no associated systemic comorbidities. On examination of neck, a 3x3 cm nontender swelling was seen in the left submandibular region. It was cystic fluctuant and brilliantly trans illuminating. The skin over the swelling was normal, pinchable without punctum (figure 1). There was no palpable cervical lymphadenopathy. The swelling was not moving with deglutition and protrusion of tongue. On oral cavity examination, a 3x3 cm diffuse bluish swelling was seen in the floor of the mouth. The swelling was fluctuant on bi digital palpation. Rest of the oral cavity examination was normal.

Aspiration cytology from the neck swelling yielded mucin with muciphages and biochemical analysis

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showed increase in amylase and protein content confirming the salivary origin of swelling. CT scan of neck revealed a noncontrast enhancing homogenous swelling with well defined borders extending

from floor of mouth to submandibular space through the posterior border of mylohyoid (figure 2).



Figure 1: Picture showing intraoral and cervical parts of ranula

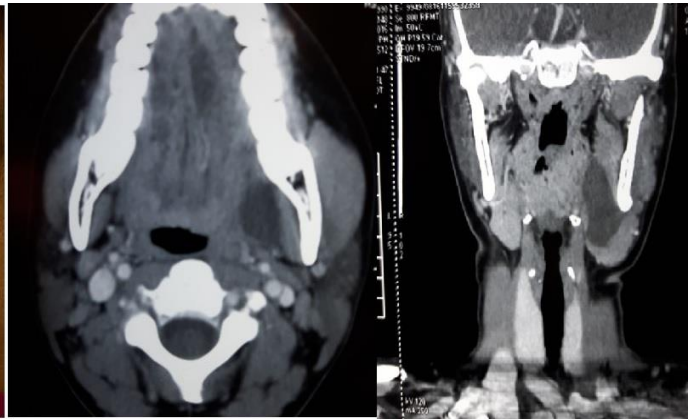


Figure 2: CT showing a noncontrast enhancing homogenous swelling with well-defined borders extending from floor of mouth to submandibular space through the posterior border of mylohyoid

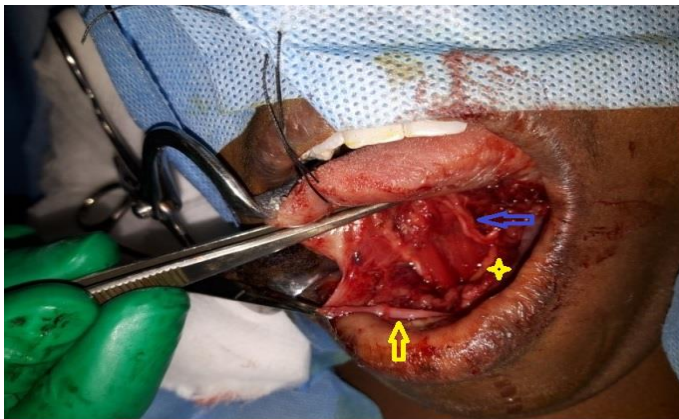


Figure 3: Intraoperative pictures showing rerouting of Wharton's duct. Yellow and blue arrow pointing the rerouted right and left Wharton's duct respectively. Yellow star points the left sublingual gland.

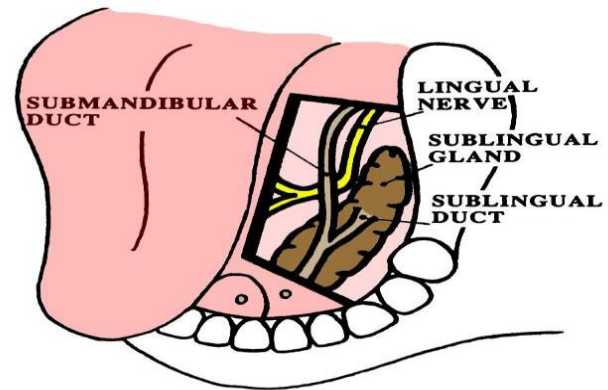


Figure 4: Picture depicting the relation of Lingual nerve and Sublingual gland with respect to Wharton's duct. Note the lingual nerve crossing the duct posteroinferiorly.

Considering cosmesis, intraoral excision of ranula along with sublingual gland excision was planned. However the limiting factors in this approach like limited exposure, injury to whartons duct were considered. A written consent was obtained from the patient. The procedure was done in general anaesthesia. Nasotracheal intubation was performed. After applying Doyen's mouth gag, the tongue was retracted by an assistant after securing it with 1'0 silk. The Wharton's duct on both sides were identified just lateral to frenulum linguae by pressing the submandibular gland and identifying the site of salivary flow. They were cannulated using 22 G intravenous catheter for identification during course of dissection. A curvilinear incision was made over the mucosa of swelling and cyst wall was dissected. During dissection, the ranula was found engulfing both the ducts completely, making the delineation of ducts from cystic wall difficult. In order to prevent injury, both the ducts were dissected from the surrounding tissue, staying as close to cannula without damaging the ductal serosa. This also helps in preventing damage to lingual nerve. Both the ducts were then rerouted posteriorly at the base of the lingual frenulum (figure 3). The cyst dissection was then continued and was removed intoto by retracting the mylohyoid muscle. The sublingual glands were found adjacent to mandibular canine and were removed separately. The whole specimen was sent to histopathological examination. The incision was closed using 3'0 vicryl sutures. Patients recovery from anaesthesia was uneventful. She was discharged on 3rd postoperative day. The patient has been on follow up with us for 1 year and the postoperative period is uneventful at the end of 1 year.

DISCUSSION

The sublingual glands are drained by numerous excretory ducts called the ducts of Rivinus. The largest SLG duct, called Bartholin's duct joins the Wharton's duct and drain in sublingual papillae on either side of frenulum linguae. Rest of the ducts open in the mucosa of floor of mouth through a crest called plica sublingualis.

A ranula develops because of extravasation of saliva from the obstructed sublingual glands. Harrison and Garrett, in 1972 demonstrated that the extravasation of saliva from the sublingual gland induce a fibroblastic reaction that seal the saliva in a connective tissue sac, forming a pseudocyst [7]. It becomes plunging when it crosses the fascial planes and forms a swelling in the submandibular region. As per literature, sublingual glands are more commonly involved than its counterparts in the formation of ranula. This could be due to the continuous secretion of SLG throughout the day, irrespective of food stimulation and relative low resistance of SLG ducts to increased intraluminal pressure during obstruction. The pathway of spread of cervical ranula may be along the deep lobe of the submandibular gland to exit posteriorly between the hyoglossus and mylohyoid muscles, or directly through a dehiscence in the mylohyoid muscle itself [8,9].

The treatment options for plunging ranula is diverse. Repeated aspiration, Incision and drainage, marsupialisation, excision of cyst,

excision of cyst along with sublingual gland and sclerotherapy with OK-432 are the available treatment options [2-5]. Zhao *et al* reported recurrence rates for the treatment of 450 oral, mixed, and plunging ranulas as follows: marsupialization (67%), ranula excision (62%), excision of sublingual gland and ranula (13%), and excision of sublingual gland (3%) [10]. The most effective surgical treatment is excision of cyst along with SLG excision, as the primary gland affected is addressed [6,11]. However, lack of true capsule makes the dissection difficult and places the vital structures like Wharton's duct, Lingual nerve and Hypoglossal nerve at risk during excision.

Plunging ranulas can be excised by intraoral, extraoral/cervical or combined cervical-intraoral approaches. Cervical and combined approaches are preferred generally because of better exposure, ease of tissue retraction and less chances of damaging lingual nerve and Wharton's duct. It is also the approach of choice in recurrent ranulas and ranulas with massive cervical extension, as it is believed that these are due to ectopic SLG below mylohyoid muscle. However, unsightly scar and risk of marginal mandibular and hypoglossal paresis either transient or permanent are few complications associated.

The patient in this case report is a young female and her concern about cosmesis made authors to approach this cyst intraorally. The obvious limitations of this approach are limited exposure and injury to Wharton's duct and lingual nerve. Iatrogenic Wharton's duct injury during ranula excision causing a new extravasation cyst have also been described in literature. These limitations could be overcome by retracting the tongue superiorly through a suture passed through it and rerouting the Wharton's duct.

Rerouting of the Wharton's duct is commonly performed for drooling of saliva in neurologically impaired children. Yates as early as 1994, described a technique to preserve WD and LN during intraoral excision of sublingual gland [12]. The Wharton's duct lies superficially in the floor of the mouth medial to the sublingual fold, and is crossed inferiorly by the lingual nerve which then enters the tongue. He raised a thin flap including the Wharton's duct, which itself led to identification of nerve, which crosses the duct inferiorly (figure 4). Similar technique was followed in this case, by raising a Wharton's duct flap and rerouting it posteriorly thereby preventing its injury as well as easy identification of lingual nerve.

CONCLUSION

Plunging ranulas does not necessitate cervical approach always. Posterior rerouting of the duct can help us preserve the duct and retain the normal physiology without compromising the excision of cyst. Removal of SLG will eliminate the chances of recurrence, though itself is rare after complete cyst excision.

Conflict of Interest

The authors declare no conflict of interest.

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