

Case Report

Rare Case of Plunging Ranula with Parapharyngeal Extension and Absent Submandibular Gland: Excision by Transcervical Approach

Abhishek Bhardwaj*, Sudhagar Eswaran, and Hari Shankar Niranjana

Department of Otorhinolaryngology, Vardhmann Mahavir Medical College and Safdarjung Hospital, India

***Corresponding author**

Abhishek Bhardwaj, Department of Otorhinolaryngology, Safdarjung Hospital & Vardhmann Mahavir Medical College, Ansari Nagar, New Delhi-110029, India, Tel: 91-989907792; Email: abhi04stanley@gmail.com

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Keywords

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Abstract

Plunging ranula extending into parapharyngeal space till the skull base with associated absence of submandibular gland is a rare finding. Transcervical approach for its excision is a challenging procedure in view of limited exposure and presence of important neurovascular structures in the field. We present a clinical case of a left sided plunging ranula extending into the parapharyngeal space till skull base in a 19 year old male who presented to a tertiary care hospital with complaints of slowly increasing swelling in neck and oral cavity for duration of six months. Ultrasound neck revealed well defined heterogeneously hypoechoic collection in left submandibular region. Contrast enhanced computed tomography revealed a non-enhancing, cystic mass involving left submandibular space extending into left parapharyngeal space till skull base and absent left submandibular gland. Ranula measuring 10cm*6cm was excised in to by transcervical approach without damage to any neurovascular structure. Histopathology was consistent with low ranula. Patient is in follow up for past six months without any recurrence.

INTRODUCTION

Ranula is a mucus retention pseudocyst from an obstructed sublingual gland. When the salivary gland duct is obstructed, there is building of secretory back pressure which leads to rupture of the duct with mucus being forced into the surrounding tissues. It is a pseudocyst because it does not have an epithelial lining of its own. It can be categorised as simple when it is confined to sublingual space, or plunging ranula when it extends inferiorly beyond the free edge of the mylohyoid muscle or through a dehiscence of the muscle itself. The term ranula is derived from the Latin word *Rana* which means 'belly of frog'. Simple ranula which lies superior to mylohyoid appears as a translucent bluish, soft, well circumscribed, painless fluid containing swelling under the tongue. Plunging ranula presents as swelling in submandibular and submental space [1]. Rarely, it can extend up to the parapharyngeal space. When plunging ranula arises from the ectopic gland, it will not have the typical sublingual part. Plunging ranula is characteristic of sublingual gland because it is the only gland that secretes mucus continuously in spite of absence of any nerve stimulation [2]. The prevalence of simple ranulas is 0.2 cases for every 1000 people. The prevalence

for plunging ranula is unknown [3]. They occur commonly in second and third decade of life [3]. CT scan plays an important role in diagnosis of ranula [4]. It helps in differentiating plunging ranula from other causes of neck swelling. Septations can be seen in plunging ranula when previous trauma or surgery is associated with it. Characteristic 'tail sign' is found in CT image of plunging ranula which suggests that the cystic swelling is plunging from floor of mouth into sublingual space [5], which is absent in case of submandibular mucocele. Surgical excision of the ranula along with sublingual gland is the treatment of choice. In this case report, we present a left side giant plunging ranula extending into parapharyngeal space. The patient also had absent left submandibular gland associated with a sialolith in left Wharton's duct suggesting of obstructive sialadenitis leading to inflammation and eventually atrophy. Very few cases of sialolithiasis with submandibular gland aplasia in a case of plunging ranula have been reported.

CASE PRESENTATION

A 19 year old male patient presented to outpatient clinic with history of painless swelling in left side of neck and floor of mouth.

Patient noticed the swelling six months back which gradually increased in size to attain the present size. Intraoral examination revealed bluish and raised floor of mouth. On palpation, the swelling was found to be soft, non-tender, and cystic. The patient did not experience any prior trauma or surgical intervention. The swelling did not cause any difficulty in swallowing or respiration. Routine blood investigations were found to be in normal limits.

Ultrasound neck revealed well defined heterogeneously hypoechoic collection of size 6.3*4.4 cm noted in left submandibular and sublingual space. Dense internal echoes were seen within this collection. Left submandibular gland was not seen separately. Small calculus of size 3.5*3.2 mm was seen impacted in distal aspect of left submandibular duct. Contrast enhanced computed tomography was done to assess the extent of lesion (Figure 1-3).

Aspirated viscous fluid was sent for cytochemical analysis which revealed mucus and numerous inflammatory cells. Fluid showed increased amylase and protein suggestive of salivary secretion. Depending on the clinical and radiological evaluation, final diagnosis of plunging ranula was made and planned for excision under general anaesthesia.

Under general anaesthesia, the cystic mass was meticulously excised intact via a transcervical approach (Figure 4). Blunt dissection was done to excise the cyst from parapharyngeal space. Precautions were taken to prevent damage to vital structures. Lingual nerve was identified and preserved. Sublingual gland was excised along with the cyst and sent for histopathological examination. The cyst was measuring 10cm*6cm in dimension (Figure 5). Left submandibular gland was found to be absent.

Histopathological examination was consistent with ranula (Figure 6 and 7). Postoperative period was uneventful. Patient was kept under follow up for 6 months and found to have no recurrence.



Figure 2 Coronal CT image showing extension of the ranula into parapharyngeal space reaching almost till skull base.

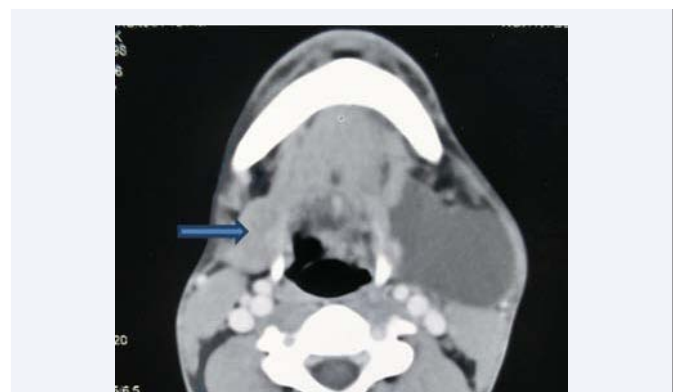


Figure 3 CT picture showing right submandibular gland (arrow) with the giant plunging ranula filling the left submandibular space and no left submandibular gland is seen separately.

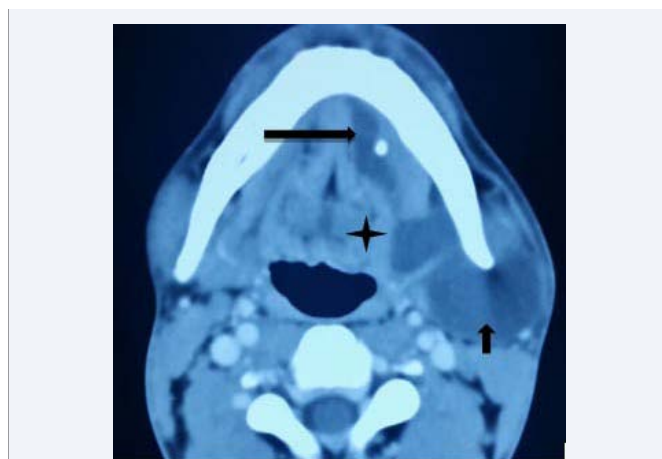


Figure 1 Contrast enhanced axial Computed Tomography shows a non-enhancing, water-density mass from the left sublingual space displacing the tongue to the right (black star). Mass extends posterolaterally into the submandibular space (small arrow). Displacement of the parapharyngeal and mucosal pharyngeal space can be appreciated. Cyst is homogenous, smoothly-marginated, and without internal septations. Anteriorly, the cyst is found tapering smoothly into the sublingual space, forming the "tail sign" (large arrow). Calculus is seen in the left submandibular duct.

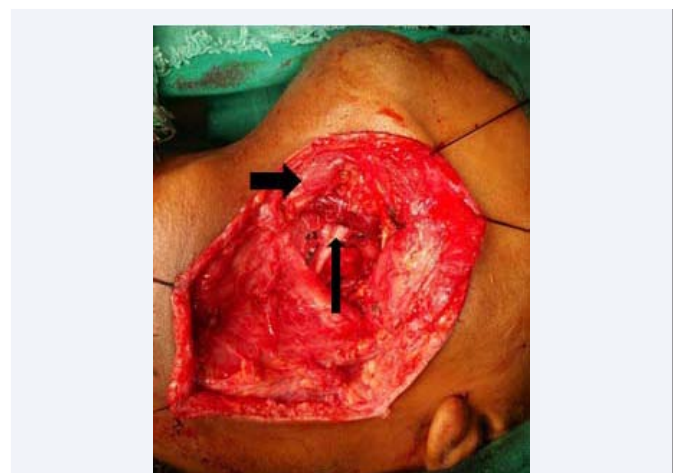


Figure 4 Transcervical approach was used to excise the plunging ranula. Image was taken after the excision. Anterior belly of digastric (short arrow) and posterior belly of digastric along with hypoglossal nerve (long arrow) are seen.



Figure 5 Excised plunging ranula.

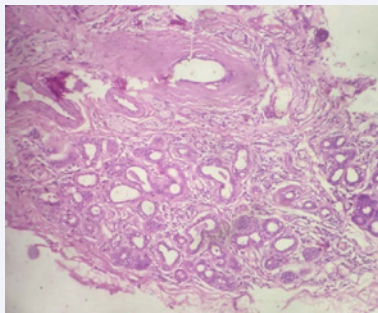


Figure 6 Histology picture of excised sublingual gland.

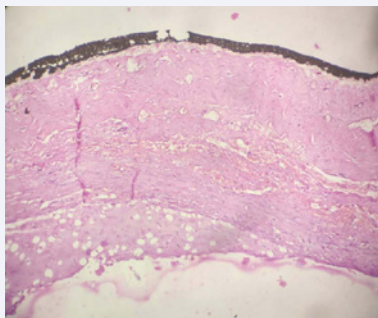


Figure 7 Histology picture of wall of excised ranula.

DISCUSSION

Ranulas are salivary extravasation cysts or mucoceles. Ranula presenting above the mylohyoid muscle are called simple ranula. They present as bluish swelling in the floor of mouth resembling frog's underside. When the ranula involves submandibular space crossing the mylohyoid muscle, it is termed plunging ranula. It can involve one or more adjacent spaces. Male to female ratio is 1:1.3 [6]. There is no significant side preference [6]. On palpation, ranulas are painless anterolateral neck masses.

Giant plunging ranulas are located mostly in the same side sublingual space extending to the parapharyngeal space.

Ultrasound can be used to demonstrate the cystic nature of ranula and its relationship to the mylohyoid muscle and sublingual gland. In present case we did ultrasound neck and CT scan and assessed the extent of the ranula. Diagnosis can be confirmed with fine needle aspiration cytology which will demonstrate mucus and high amylase content.

Left submandibular gland was found absent in this case. It was associated with a calculus in left wharton's duct. Very few cases of unilateral submandibular gland atrophy following obstructive sialolithiasis have been reported. Koo et al., [7] have recently reported about this association. Following ligation of submandibular duct in rats, glandular atrophy was evident after 24 hours in the study conducted by Osailan et al., [8]. The obstruction found in the duct can cause a negative feedback to the gland which could have resulted in reduction of salivary flow. Atrophy can be reversible once the obstruction is released [9]. In this case, absent submandibular gland may be due to the atrophy of the gland following the obstructive sialolithiasis.

Surgical excision is the preferred choice of treatment for ranula. Excision can be done by intraoral or transcervical approach. Involved sublingual gland should also be removed to prevent recurrence. Recurrence rate is 2% if sublingual gland is excised and can go up to 50% if excision is incomplete [10]. Transcervical approach for cyst extending into parapharyngeal space is difficult because of inadequate exposure and needs skilful dissection in order to achieve complete excision and to prevent damage to neurovascular structures in parapharyngeal space.

Other surgical treatment options are marsupialization, aspiration and only ranula excision. All of these procedures are associated with increased recurrence rate. Sclerotherapy with OK - 432 is another option but it needs multiple injections. Fukase et al. [11], reported that the success rate after the first injection was only 45.5% and only after multiple treatments complete clinical resolution occurred.

In this case surgical excision of the giant plunging ranula along with sublingual gland was done by transcervical approach. Transcervical approach was preferred in order to achieve a better exposure in view of parapharyngeal extension of the lesion reaching just below the skull base. Intraoral approach would not have given the adequate exposure.

Complete excision was achieved without damage to any vital structure. The post operative period was uneventful. Patient was under follow-up for 6 months without any sign of recurrence.

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