Plunging ranula with prestyloid parapharyngeal space, masticator space, and parotid gland extension

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Abstract. Plunging ranula with prestyloid parapharyngeal space, masticator space, and parotid gland extension. Introduction: Ranulas develop from mucous extravasation secondary to sublingual gland duct obstruction or trauma. Plunging ranula usually dive into the submandibular space. Methods: This is the first reported case of a plunging ranula with direct extension to the prestyloid parapharyngeal space, masticator space, and parotid gland with avoidance of the submandibular space. Results: The patient presented with a tender parotid mass, of which the differential is broad, including parotitis, parotid malignancy, metastatic malignancy, lymphoma, as well as other infectious etiologies. When an intraoral component is not identified, other differential considerations would be thyroglossal duct cyst, branchial cleft cyst, parathyroid cyst, cervical thymic cyst, dermoid cyst, cystic hygroma, or benign teratoma. Conclusion: The case is unique due to ranula extension into multiple spaces. For optimal treatment, the sublingual gland along with its tract and contents needs to completely removed.

Introduction

Ranulas develop from mucous extravasation secondary to sublingual gland duct obstruction or trauma.1 Plunging ranula typically communicate with an oral ranula, although they can be independent of an intraoral element. Plunging ranula usually arise through three different mechanisms into the submandibular space.2 The most common mechanism has the sublingual gland project through the mylohyoid muscle in an area of dehiscence. These dehiscent sites form a mylohyoid hiatus allowing a path of least resistance. Second, an ectopic sublingual gland can be located on the mylohyoid cervical side. Third, a sublingual gland duct may form a connection with the submandibular gland (SMG) or its duct. Ranula can then form in continuity with the SMG. This connection allows neck access from behind the mylohyoid. This report will detail a plunging ranula with direct extension to the prestyloid parapharyngeal space, masticator space, and parotid gland with avoidance of the submandibular space.

Case report

A 66-year-old male with history of liver transplant, hepatitis C, end-stage renal disease on hemodialysis, hypertension, diabetes mellitus, and severe thrombocytopenia was transferred to the Massachusetts General Hospital for medical evaluation after sustaining a fall resulting in multiple rib and lumbar fractures and altered mental status felt secondary to dehydration. A non-contrast computed tomography noted possible peritonsillar abscess (PTA) and minimal fullness to the left parotid gland (stable from 11 years prior). Evaluation revealed no peritonsillar fullness, fluctuance, erythema, or uvular deviation to suggest PTA. There was no evident mass lesion in tonsillar region to explain imaging findings. The base of tongue was without palpable abnormalities. The nasal mucosa was noted to be dry. A palpable tender mass in region of the left parotid tail was noted. Saliva was extracted from stenson’s duct without purulence. Right parotid was without fullness or tenderness. The submandibular glands were normal
size and shape, with mild tenderness to palpation. Fiberoptic examination was unremarkable.

Head and neck MRI imaging demonstrated a T2 hyperintense lesion centered in the left sublingual space with extension into the left prestyloid parapharyngeal space, the left masticator space, and the left parotid gland, favored to represent a ruptured and inflamed left ranula (Figure 1). Figures 2 and 3 demonstrate the lesion on coronal T2 imaging and posterior to the mandible into the parotid on T1 sequence, respectively. Superimposed infection was suspected due to left facial pain and elevated white blood cell count. There was no cervical adenopathy or discrete mucosal lesion. Asymmetrically prominent left level II, III and V lymph nodes with normal morphology and signal characteristics were likely reactive, and clinical follow-up to resolution was recommended. The lesion was not visible of previous imaging within one-year prior to presentation.

Fine needle aspiration (FNA) was pursued revealing abundant polymorphs (98% neutrophils), moderate red blood cells, and moderate gram positive cocci in pairs and clusters. FNA aspirate was also sent for amylase, culture, acid-fast bacilli (AFB) stain and culture, fungal stain and culture, Epstein-Barr Virus (EBV) serology, and cytology. Intravenous vancomycin (hemodialysis dosing) and aztreonam renal dosing were started while awaiting aspirate culture results. Amylase levels were elevated at 1160U/L, suggesting ranula. Transplant infectious disease recommended discontinuing aztreonam for suspected parotitis. Culture demonstrated methicillin-sensitive staphylococcus aureus, however due to penicillin allergy, a 7-day course of vancomycin was continued. EBV was not detected, and AFB results are in process. Additional otolaryngology recommendations included - salivary gland massage at least three times per day, warm compresses as needed, and use of sialogogues. The parotitis resolved after the antibiotic course. The sublingual gland was completely excised via an intraoral incision and a pathologic diagnosis of ranula was confirmed.

![Figure 1](image1.png)

*T2 hyperintense lesion centered in the left sublingual space with extension into the left prestyloid parapharyngeal space, the left masticator space, and the left parotid gland.*

![Figure 2](image2.png)

*T2 hyperintense lesion in the left sublingual space on coronal imaging.*
Discussion

In the context of transplantation, ESRD, significant thrombocytopenia, the differential for a tender parotid mass is broad, including parotitis (related to either recent dehydration, viral or bacterial process), parotid malignancy including Warthin’s tumor (given patient’s reported history of waxing and waning tenderness of this gland), other malignancy with metastasis to this region (with possible relevance to the ipsilateral peritonsillar fullness seen on CT), lymphoma (given transplant history and ipsilateral tonsillar fullness), as well as other infectious etiologies including tuberculosis/scrofula (negative T spot 7 months prior). A venolymphatic malformation could have the imaging appearance, but this was considered less likely given the lack of corresponding abnormality on prior imaging obtained through the upper neck, the patient’s age, and presence of amylase. Elevated amylase levels have not been associated with lymphatic malformation in the literature, although there are anecdotal reports. When an intraoral component is not identified, other differential considerations would be thyroglossal duct cyst, branchial cleft cyst, parathyroid cyst, cervical thymic cyst, dermoid cyst, cystic hygroma, or benign teratoma. The diagnosis of a plunging ranula can be made by clinical features being a unilateral, painless, cystic swelling beneath the submandibular space, and a positive FNA for amylase along with the above radiographic features. Elevated amylase on FNA confirms salivary gland involvement.

The management of non-plunging ranula remains controversial, varying from incision with marsupialization to sublingual gland excision. Marsupialization may be technically difficult and often results in recurrence. Recurrent ranula make subsequent surgical more difficult, and therefore initial total excision is recommended. For plunging ranula, a variety of surgical techniques are used including sclerotherapy or excision of the sublingual gland with or without removal of the mucous extension. For optimal treatment, the sublingual gland along with its tract and contents needs to completely removed. The SMG can also be excised if thought to be in communication with the ranula. For ectopic sublingual glands, excision of the gland with plunging ranula removal is recommended. In a review of 571 ranula patients and 606 procedures (580 primary lesions and 26 recurrences), 9 involved marsupialization only, 28 had the ranula excised, 356 had the sublingual gland excised, and 213 were an excision of the sublingual gland in conjunction with the ranula. Subsequently, recurrence rates were highest for marsupialization at 66%, 57% for ranula excision, and only 1.2% for combined sublingual gland and ranula excision. These data demonstrate excising the sublingual gland along with the ranula contents provides optimal management.

Conclusion

Our patient is the first described such case of ranula extension into the parotid gland. The case is a unique surgical challenge to completely excise the sublingual along with its tract due to the plunging ranula extending into multiple spaces from the prestyloid parapharyngeal to the masticator space and into the parotid gland. The atypical course of the plunging ranula, combined with chronic immunosuppression, gives this lesion a broad differential. The atypical course might be explained by chronic infection secondary to immunosuppression. For optimal results, the sublin-
gual gland and its contents must be completely excised regardless of extension area.

References


