A rare cause of lateral facial swelling

Sujata Mohanty, Ujjwal Gulati, Vandana, Sapna Singh
Department of Oral and Maxillofacial Surgery, Maulana Azad Institute of Dental Sciences, Bahadur Shah Zafar Marg, Department of Radiology, LNJP Hospital, New Delhi, India

Address for correspondence:
Dr. Sujata Mohanty, Department of Oral and Maxillofacial Surgery, Maulana Azad Institute of Dental Sciences, Bahadur Shah Zafar Marg, New Delhi - 110 002, India.
E-mail: drsujatam@hotmail.com

ABSTRACT

A case of chronic, recurrent and asymptomatic facial swelling in a young male is presented. Swelling extended from lower midface to upper lateral neck and right commissure to anterior massectric border. History, clinical signs and symptoms and examination pointed towards the benign nature of the swelling. Fine-needle aspiration cytology tapered the diagnostic possibilities to a salivary cyst or pseudocyst. Ultrasonography identified the lesion to contain echogenic fluid with irregular borders. “Tail sign” was absent on contrast magnetic resonance imaging, excluding the involvement of the sublingual gland. Surgical excision of the lesion was done along with submandibular gland as both were in continuity via a bottle-neck tract. Final histopathological diagnosis was that of the submandibular gland extravasation phenomenon. As per the best of our knowledge, it is the first case report of a submandibular gland extravasation causing swelling in a retrograde direction onto the face.

Keywords: Extravasation phenomenon, pseudocyst, submandibular gland, tail sign

INTRODUCTION

Intraoral extravasation phenomenon of the salivary glands is not an uncommon entity. However, extravasations causing extraoral swellings are a rare phenomenon. These extravasations are present as neck swellings in midline or lateral neck region. In most of the cases, the gland of origin is sublingual gland. Submandibular gland extravasations are extremely rare, and only 12 cases have been reported in the literature as per the best of our knowledge. All of them manifested as lateral neck swellings in the submandibular region except one, which occurred in midline. We report the first case of submandibular extravasation with predominant swelling of the face extending to neck.

CASE REPORT

The 25-year-old male reported to our department with a complaint of an asymptomatic swelling on the right side of the face. Patient gave history of similar swelling, much smaller in size that used to regress on its own. He had experienced such episodes quite a few times during past 3-4 years. Medical history of the patient did not reveal any significant finding. Patient could not relate the occurrence of swelling to any event namely trauma, meals, dental pain, fever, etc.

On examination, a diffuse swelling of about 7 × 3 cm extended from midface to right submandibular region (supero-inferiorly) and from right oral commissure to the anterior border of masseter (anteroposteriorly) [Figure 1]. The swelling was moderately firm, non-tender, fairly mobile and not fixed to the skin or underlying tissues. The color, texture and temperature of the overlying skin were normal. Submandibular nodes could not be examined on the ipsilateral side, and other cervical nodes were not palpable bilaterally. There was no trismus, dysphagia, dyspnea or Dysphonia. Ipsilateral ear lobe was not everted, and masseter was normal in palpation. Neck movements were normal and cranial nerve functions intact. Mouth opening was not restricted, and there were no signs of any intraoral pathology or tooth decay. Flow from all salivary ducts was normal. The differential diagnosis based on clinical findings included buccal space infection, dermoid or epidermoid cyst, branchial cleft cyst, lipoma, vascular malformation, recurrent lymphadenitis, recurrent sialadenitis, cyst/pseudocyst or tumor of salivary origin. Absence of inflammatory signs and normal intraoral findings excluded chances of infection. The recurrent nature of the swelling hinted
more toward recurrent sialadenitis or recurrent lymphadenitis and nullified the tumoral etiology.

Aspiration of the lesion resulted in yellowish fluid of a mucoid consistency that was sent for microscopic and biochemical examination. The clinical picture of aspirate reduced the probability of vascular anomaly, dermoid or epidermoid cyst to a great extent. Other investigations were also ordered (complete hemogram, ultrasonography and thyroid profile) and results awaited. Incidentally, patient experienced increase in swelling while having his meals during this period. The swelling had got painful as the patient was having his meals.

Microscopic examination of the aspirate showed presence of cystic macrophages in mucoid background. Culture was negative for any bacterial growth. Aspirate had high amylase content. This information was more suggestive of a salivary cause for the lesion. Microscopic examination negated the cystic fluid content to be of a branchial, dermoid or epidermoid cyst as there was no evidence of cholesterol crystals, keratin or other relevant contents. Provisional diagnosis was more focused on saline retention or extravasation phenomenon at this stage.

Ultrasonogram of the neck was done with high resolution 7.0 MHz multifrequency linear transducer. The report was suggestive of 7-9 ml of slightly echogenic fluid collection just anterior to right angle of mandible. Margins of fluid collection were irregular. There was no evidence of calcifications. Multiplanar magnetic resonance imaging (MRI) of the face was acquired on a 1.5 tesla magnetic system to determine the precise extent and relation of the lesion to adjacent vital structures. Turbo spin echo, short tau inversion recovery sequences were used to obtain T1-weighted (T1W) and T2-weighted (T2W) images. The study revealed large, relatively well-defined and lobulated 5.5 × 4.5 × 2.5 cm cystic lesion appearing hypointense on T1W and hyperintense on T2W images. It showed few septations and thin peripheral enhancement. The lesion was seen abutting the anterior surface of right submandibular gland and lying lateral to the mylohyoid muscle. The lesion occupied the right submandibular space and extended across the posterior and inferior surface of the mandible over the anterior margin of the masseter muscle. There was no invasion of parapharyngeal or sublingual space. Soft tissue infiltration was also absent. Submandibular gland and parotid gland per SE were normal. "Tail sign," which is pathognomonic of sublingual gland extravasation was absent [Figure 2a-d]. Diagnosis tapered to submandibular gland pathology, most likely being the rare extravasation phenomenon.

The lesion was approached extraorally via submandibular incision [Figure 3]. A layered dissection protecting the marginal mandibular nerve was done to approach the facial extension of the lesion. The pathology in the buccal space extended up to zygomatic buttress superiority lying posteriorly over the masseter muscle. A thin mix of alginate was injected into the cystic cavity to delineate it from surrounding structures and facilitate removal as the lesion was adherent to adjacent tissues superiorly. After dissecting the facial extension of the lesion, the pathology was followed down into the submandibular space [Figure 4]. The cystic cavity was in continuity with submandibular gland via a bottle-neck tract and, hence, it was excised along with the respective gland [Figure 5]. Adjacent vital structures were preserved, and respective duct ligated. Sublingual gland was not involved clinically too. Histopathological analysis revealed loosely arranged collagen fibers, mild chronic inflammatory cell infiltrate and numerous mucinophages, which in correlation with clinical findings was suggestive of the mucous extravasation phenomenon in relation to the submandibular gland [Figure 6a and b]. Patient is under followup with no recurrence [Figure 7].

Figure 1: Preoperative presentation of patient showing facial extension of submandibular swelling

![Figure 2: (a) T1-weighted (T1W) axial magnetic resonance imaging (MRI) showing a well marginated lesion in the right submandibular region. The lesion shows a hypointense signal with no septations. (b) Fat suppressed T2W axial MRI showing the lesion to be high signal. The compressed submandibular gland is seen lying adjacent to the lesion. (c) Fat suppressed T2W axial MRI showing the lesion to have a small projection towards the floor of the mouth. The typical "tail sign" of a plunging ranula was not seen. (d) Post-contrast axial MRI showing the lesion having a thin peripheral rim enhancement]
proposed developmental etiology. Irrespective of the cause, extravasation leads to mucin pool in tissue spaces and elicits inflammatory response. Granulation tissue forms around the mucin and encapsulation occurs to form a pseudocyst. When this extravasation occurs in superficial tissue planes, it clinically presents as a fluctuant, painless and bluish swelling in the region of lower lip, floor of the mouth, ventral tongue or buccal mucosa. Other regions such as the palate, retromolar region and submandibular gland are rare sites for extravasation probably due to low susceptibility to trauma.[1] This encapsulated accumulation may fluctuate in size due to the intermittent drainage of the collected mucin, but recurs due to continued production of mucin.[2] Most accepted treatment option has been the excision of the salivary pool along with the gland involved. Other treatment options have been successfully tried.[3,4] The mucous retention phenomenon occurs as a result of obstruction of the duct by a sialolith or a stricture. The hindrance in the flow of saliva from gland to the oral cavity leads to its accumulation in duct and back pressure. The flow from gland continues, and this causes gradual inflation of ductal diameter to accommodate the salivary volume. The pooled saliva is walled by inflamed duct making the picture resemble a cystic pathology. It is more of a true cyst, as there is the presence of an epithelium lining. Back pressure may inflame the gland and cause acute/chronic recurrent sialadenitis. The signs and symptoms and management vary with the type of gland involved and the anatomical position of obstruction.[1]

DISCUSSION

Mucous extravasation results from rupture of minor or major salivary duct due to trauma. Some authors have also

Figure 3: Exposure via submandibular approach

Figure 4: Facial extension of lesion dissected (circled) and found to be in continuity with submandibular gland

Figure 5: Resected lesion and submandibular gland

Figure 6: (a) Superficial infiltration of chronic inflammatory cells admixed within loosely textured collagen fibers (H and E, ×100). (b) Loosely textured collagen fibers with numerous mucinophages (PAS, ×400)

Figure 7: Six months postoperative photograph
Ranula and mucocele are clinical terms signifying mucous retention or the mucous extravasation phenomenon. However, extravasation is much more commonly seen than true retention. Ranula derives its name from Rana (frog) family as its clinical appearance (translucent blue swelling) in floor of the mouth resembles belly of a frog. It has the capacity to displace the tongue supero-medially on assuming a large size. It may be seen as a midline swelling if the mucin accumulates in the middle of the floor of the mouth. Second possibility is that it may herniate through the mylohyoid into neck spaces, and such appearance is called plunging ranula. An extremely rare possibility is that the mucous may pass through the gaps between the muscle, into the neck spaces and cause extravasation phenomenon extending till there.[1] An extensively infiltrative ranula involving multiple neck spaces and extending superiorly till pterygoid plates has also been reported. The same phenomenon occurring elsewhere in the oral cavity with a much smaller size, for, e.g. lower lip, palate, etc., is called mucocele.

Zhao et al. published an extensive review of 580 ranulas and classified them into three types: oral, plunging and mixed. They reported on three methods of management; marsupialization, excision of the lesion and excision of the lesion with the involved gland. Recurrence seen with these three treatment modalities was 66.67%, 57.69%, and 1.20% respectively. Of the 580 cases, none involved only submandibular gland. However, some lesions were involved only submandibular gland. Patient was taken up for surgery under general anesthesia, and the lesion was excised for surgery under general anesthesia, and the lesion was excised

The authors suggested that an expanding ranula can obstruct Wharton duct and cause its rupture involving submandibular gland in association with both sublingual and submandibular glands. The cases reported have mostly presented as lateral facial swelling. Ann Maxillofac Surg 2014;4:230-3.

REFERENCES

1. Neville BW, Damm DD, Allen CM, Bouquot JE. Oral and Maxillofacial Pathology. 2nd ed. Philadelphia, PA: Saunders; 2002; p. 391-2.
2. Harrison JD, Sowray JH, Smith NJ. Recurrent ranula. A case report. Br Dent J 1976;140:180-2.
3. Amaral MB, De Freitas JB, Mesquita RA. Upgrading of the micro-marsupialisation technique for the management of mucous extravasation or retention phenomena. Int J Oral Maxillofac Surg 2012;41:1527-31.
4. Lai JB, Poon CY. Treatment of ranula using carbon dioxide laser: Case series report. Int J Oral Maxillofac Surg 2009;38:1107-11.
5. Zhao YF, Jia Y, Chen XM, Zhang WF. Clinical review of 580 ranulas. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2004;98:281-7.
6. Hughes WG, Houston GD, Savage MG. Slow-growing midline submental mass. J Oral Maxillofac Surg 1999;57:61-5.
7. Stranc MF, Skoracki R. A complication of submandibular intubation in a panfacial fracture patient. J Craniomaxillofac Surg 2001;29:174-6.
8. Anavi Y, Kaplan I, Calderon S. Lateral neck mass. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2002;94:536-9.
9. Ozturk K, Yaman H, Arbag H, Koroglu D, Toy H. Submandibular gland mucocele: Report of two cases. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2005;100:732-5.
10. Coit WE, Harnsberger HR, Osborn AG, Smoker WR, Stevens MH, Lufkin RB. Ranulas and their mimics: CT evaluation. Radiology 1987;163:211-6.