

Recurrent unilateral submandibular swelling

Ibrahim O. Bello, BDS, PhD,^a Ahmed Qannam, BDS, MSc,^a and Ahmed Sulaiman, BDS, MSc, PhD,^b

Riyadh, Saudi Arabia

KING SAUD UNIVERSITY

(Oral Surg Oral Med Oral Pathol Oral Radiol 2013;115:705-709)

A 27-year-old female patient from the Republic of Yemen presented to the Oral and Maxillofacial Surgery Department, College of Dentistry, King Saud University, with a 2-year history of a recurrent, painful submandibular swelling. She described the pain as an intermittent, dull ache in the right lower jaw region that becomes worse after meals. The pain was relieved with over-the-counter analgesics, such as nonsteroidal anti-inflammatory drugs. Her past history was significant for asthma and anxiety. Clinical examination revealed a somewhat enlarged, nontender, soft right submandibular gland. The gland was milked for saliva. Normal saliva was driven out of the gland with no signs of purulence or decreased flow. No hardened masses or calculi were felt on palpation of the Wharton duct or the gland itself. The overlying mucosa was normal. The overlying skin was nonpulsatile, and of normal color and temperature. A requested orthopantomogram (OPG) showed no radiographic abnormality within the submandibular region. This was followed by an ultrasound examination, which showed an essentially normal-appearing gland apart from dilatation of some of the ducts.

DIFFERENTIAL DIAGNOSIS

Based on the history and clinical presentation, the lesion was suspected to be within the submandibular gland or its duct, although other causes of submandibular swellings were not initially ruled out. The differential diagnosis could be divided into the general categories of obstructive, inflammatory, cystic, and neoplastic (benign or malignant) lesions. Specifically, a differential diagnosis of sialolithiasis, immunoglobulin (Ig)G4-related sclerosing sialadenitis, cervical lym-

phoepithelial cyst, plunging ranula, and a submandibular gland neoplasm was made.

The history of painful, recurrent glandular swelling with an associated increased intensity after meals gave an impression of a chronic obstructive lesion secondary to calculi formation (sialolithiasis). Sialoliths are most commonly found within the ductal system of the submandibular gland owing to the long and tortuous path of the Wharton duct and the more viscid (mucoid) secretions of the gland. Recurrent postprandial pain and swelling are more often seen in patients with sialoliths of the major salivary glands. The radiological and imaging findings were not entirely supportive of sialolithiasis. The absence of calculi within the Wharton duct and the soft consistency of the gland on palpation were also factors weighing against the diagnosis of sialolith-related sialadenitis; however, it is important to remember that during the early phase of calcification, a sialolith may not show on the radiograph, and that firmness and fibrosis of a gland is a feature of only a prolonged and chronic sialadenitis.

Although IgG4-related (sclerosing) sialadenitis should be considered in any suspected submandibular gland swelling, the fact that this patient had a soft consistency of the gland on palpation and pain only related to meals makes this diagnosis an unlikely diagnostic candidate. IgG4-related sialadenitis of the submandibular gland usually manifests as a firm, painless, unilateral, or occasionally bilateral swelling, clinically mimicking a tumor. Pain can sometimes be a feature, although it is extremely uncommon.^{1,2} A cervical lymphoepithelial cyst (branchial cleft cyst) is a congenital cyst that usually presents as a unilateral swelling anterior to the sternocleidomastoid muscle in late childhood or early adulthood. It is a slow-growing, fluctuant mass that becomes apparent in the second or third decade of life. It can occur at any point between the hyoid bone and suprasternal notch.³ It is normally painless except when it becomes infected. It is also usually more posterolateral in location than the swelling of this patient.

Plunging ranulas usually originate as an extravasation of saliva from the sublingual gland and then dissect through the fascial planes of the mylohyoid muscles to reach the submandibular space. They are usually a result of trauma and occasionally show no clinical

^aAssistant Professor, Department of Oral Medicine and Diagnostic Oral Sciences, King Saud University, College of Dentistry, Riyadh, Saudi Arabia.

^bProfessor, Department of Oral and Maxillofacial Surgery, King Saud University, College of Dentistry, Riyadh, Saudi Arabia.

Received for publication Nov 2, 2011; returned for revision Dec 12, 2011; accepted for publication Jan 18, 2012.

© 2013 Elsevier Inc. All rights reserved.

2212-4403/\$ - see front matter

doi:10.1016/j.oooo.2012.01.022

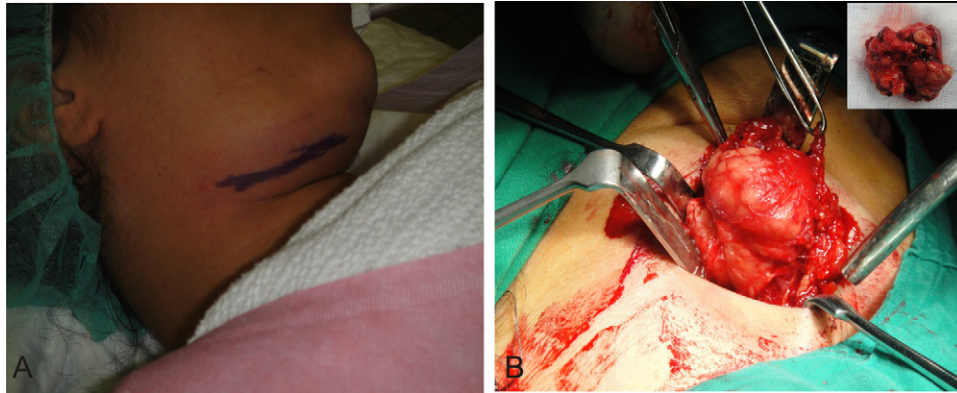


Fig. 1. **A**, Patient being prepared for surgery. A submandibular swelling is seen close to the marked incision site although the extent is not visible because the patient is in a recumbent position. **B**, Excision of right submandibular gland along with the extraglandular tissue. Inset is the gland after removal.

connection intraorally to the floor of the mouth.⁴ They commonly present as a fluctuant, nontender submandibular swelling with an intact overlying skin. The presence of pain in this patient and its association with meals and the absence of fluid collection in the diagnostic ultrasound made it difficult to seriously consider this entity.

The submandibular gland can be affected by a number of benign and malignant tumors. Soft tissue or extraglandular tumors include lipoma, neurofibroma, and hemangioma, and their malignant counterparts. They can involve the gland directly through being located within it, or in close proximity to it. The most common benign tumor of the submandibular gland is pleomorphic adenoma.⁵ Benign tumors of the submandibular glands are usually painless, slow growing, and on palpation appear as firm, occasionally nodular masses that may be freely movable. They do not usually cause pain related to meals or give a long-term intermittent, dull ache. The most common malignant tumors of the submandibular gland are adenoid cystic carcinoma and mucoepidermoid carcinoma.⁵ These tumors are usually firm and nodular and may become fixed at a later stage. They may occasionally cause pain owing to pressure, or owing to perineural invasion in the case of adenoid cystic carcinoma. Such pains are usually independent of meals. Extraglandular tumors of the submandibular space were also considered but were not included in the differential diagnosis of this lesion. They are not common, and their presentations do not sufficiently fit the present complaint of this patient. Despite the wide variety of lesions included in the differential diagnoses, none of them was considered the most likely cause of the patient's symptoms. Chronic sialadenitis secondary to sialolithiasis, IgG4-related chronic sialadenitis, and neoplastic submandibular gland lesions were still in the final tentative diagnoses.

DIAGNOSIS AND MANAGEMENT

With her increased symptoms and her concern of an underlying neoplastic lesion, a discussion of immediate surgical management was made. The patient was admitted to King Abdulaziz Hospital and, under general anesthesia, the submandibular gland was excised (Figure 1). During surgery, no abnormal bleeding was noticed and the resultant bleeding was controlled effectively with cauterization of the small vessels and ligation of the larger ones. On removal, the submandibular gland was soft, not firm or fibrotic, and showed no signs of infection (Figure 1, B). The excised gland was sent for histopathological examination. The wound was sutured in layers, and a drain was inserted to be removed 48 hours later.

Microscopic examination revealed lobules of normal, predominantly serous and a few mucous salivary gland acini with only a few areas of mild, patchy chronic inflammation. No glandular acinar atrophy or effacement was noted in any part of the sections (Figure 2). In the extraglandular tissue, there were endothelium-lined, dilated interconnecting vascular spaces with muscular walls (veins), scattered lymphatics, and scanty arterioles (Figure 2). These were separated by fibrous connective tissue with intermingled lobules of fat. Scattered organized thrombi and phleboliths were also seen (Figure 3). The admixture of different vessel types suggested an arteriovenous malformation, although the lesion was predominantly venous (including the presence of phleboliths and organized thrombi). The clinicopathologic correlates seemed to also favor a venous malformation. A final diagnosis of vascular malformation (VM) of the submandibular gland was made.

The postoperative healing was uneventful. A follow-up was done weekly for 2 weeks, and the patient

Download English Version:

<https://daneshyari.com/en/article/6058461>

Download Persian Version:

<https://daneshyari.com/article/6058461>

[Daneshyari.com](https://daneshyari.com)