Cavernous Hemangioma In The Submandibular Gland Masquerading As Sialadenitis: A Case Report

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Citation

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Abstract

Submandibular sialadenitis is a common condition encountered in the submandibular gland with typical clinical features but occasionally the condition can masquerade as a benign neoplasm and if proper investigations are not done it can lead to an erroneous diagnosis. We present a case report of cavernous hemangioma in the submandibular gland of a 40 years male, masquerading as submandibular sialadenitis. The rarity of cavernous hemangioma in such a location, its simulation to such a common disorder and occurrence in an adult male led us to report the case.

INTRODUCTION

Submandibular sialadenitis is a common condition encountered in the submandibular gland with typical clinical features but occasionally the condition can masquerade as a benign neoplasm and if proper investigations are not done it can lead to an erroneous diagnosis. One such case of cavernous haemangioma mimicking as submandibular sialadenitis is reported

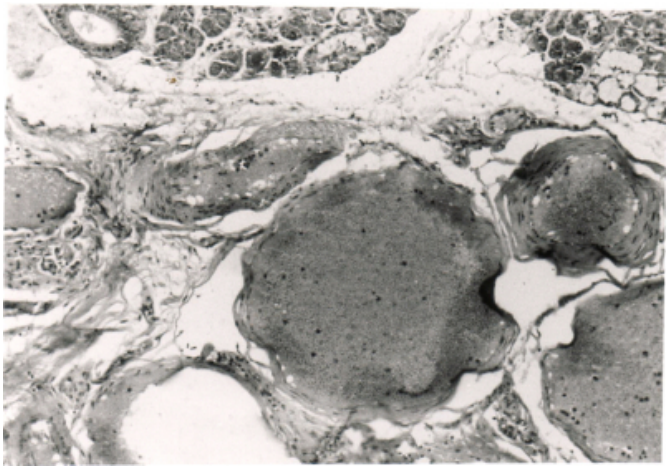
CASE REPORT

A 40-year male reported to us in the Out Patient Section of the Ear Nose Throat Department, Government Medical College & Hospital, Chandigarh with the chief complaint of right-sided swelling in the submandibular region for the past 3 years. Initially, the swelling used to increase in size at the time of meals and used to decrease on its own. Now for the past 3 months the swelling was gradually progressive and causing distinct cosmetic deformity to the patient. It was bimanually palpable and a stone could be felt in the gland. There were no associated features of pain, or fever. On clinical examination, an oblong swelling of 4cm x 3cm in maximum dimensions with diffuse margins, non-tender and firm in consistency was observed. FNAC was done which yielded only blood. Finally a Computerized Tomography (CT) scan was done which demonstrated a large lobulated mass in the right submandibular gland measuring about 3x4.5x3cm, containing punctate calcifications and low attenuation areas. A contrast CT could not be done, as the patient was allergic to the contrast material. Keeping in view the history, clinical examination and CT scan, an impression of submandibular sialadenitis or benign condition was thought and excision of the gland was planned.

At surgery, a bluish coloured lesion was seen involving whole of the submandibular gland .The facial artery, which was feeding the tumour, was ligated and the specimen was removed in toto. Histopathological examination of the specimen revealed lobules of blood filled cavernous spaces lined by flattened endothelium and infiltrating the salivary acinar tissue (Fig 1). Post-operative period was uneventful. The drain was removed on the 4th post-operative day and the patient was discharged on the 7th post –operative day after suture removal.

Figure 1

Figure 1: Photomicrograph showing lobules of blood filled cavernous spaces, surrounded by normal salivary gland tissue. Stain: Hemotoxin & Eosin, Magnification: 100X



DISCUSSION

Haemangioma of salivary gland especially parotid is quite common in paediatric population[1], but is exceedingly rare to find it in sumandibular gland of adult males. After an extensive search of the available literature, we could only find 14 cases of haemangioma of submandibular gland in adults[2,3].

Pathologically, it is debatable whether they are true neoplasms or vascular malformations. In the salivary glands two main types of haemangiomas occur: cavernous and capillary. Capillary type is lobulated, lacks a capsule is purplish in colour and infiltrates the gland involved. Microscopically solid masses of cells and multiple anastomosing capillaries replacing the acinar structure of the gland are seen. The cavernous type is formed by dilated blood vessels or sinusoids lined by endothelium. It is also devoid of a capsule and is infiltrative in nature[4]. It is interesting to note that these benign mesenchymal tumours are common in parotid, probably due to lack of well-defined capsule and presence of neurovascular structures in it[2].

Not much is known regarding the aetiopathogenesis of these mesanchymal tumours. According to Batsakis[5], the cavernous type is either the consequence of trauma or a congenital malformation. Yet another theory states that haemangiomas are generally hamartomatous in nature[6].

No distinct clinical features can be attributed to haemangiomas. A female preponderance of 2:1 is reported in medical literature. Haemangiomas that occur at birth are usually capillary type and involve primarily the parotid gland. They run a benign course and regress. The cavernous ones occur later in life and present as rubbery, firm to soft palpable mass. They are painless and have no bruit. They have a progressive and unresolving course. Haemangiomas may be associated with cutaneous lesions – a strong diagnostic point. Malignant transformation is not reported.

The diagnosis of this lesion in submandibular gland, especially in adults, becomes quite difficult as these lesions are rare and no confirmative non-invasive investigation exists. Plain X-ray may show multiple calcified phelebolith, Ultrasonography reveals heterogeneous, hypoechoic lesions with calcified phleboliths, CT Scan shows tumour with enhancing quality of blood vessels. Magnetic Resonance Imaging demonstrates hyperintensity on T1 weighted images and isointensity with muscle with T2 weighted images. Sialography and angiography have been mentioned and can be done[2,9,10].

As far as submandibular gland haemangiomas are concerned, the treatment of choice is surgical resection. Other modalities of treatment include laser, cryotherapy, embolization and corticosteroids[3].

The case is of importance on two accounts; firstly the submandibular gland is a very uncommon site for cavernous haemangiomas in an adult male and secondly the clinical features were that of submandibular sialadenitis. The stone which was palpable was due to a phlebolith and the clinical picture was that of secondary submandibular sialadenitis.

Contrast injection while doing CT Scan is important. In our case no contrast was given as the patient had demonstrated evidence of allergy to the contrast material. Therefore we could not think of vascular lesions inside the gland and rather thought of a benign condition. Hence, though haemangiomas are rare, they can simulate other benign conditions like submandibular sialadenitis and should be kept as a differential diagnosis of a swelling in the submandibular region.

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