International Surgery Journal
Bhatia C et al. Int Surg J. 2017 Jun;4(6):2081-2083
http://www.ijsurgery.com
pISSN 2349-3305 | eISSN 2349-2902

DOI: http://dx.doi.org/10.18203/2349-2902.isj20172414

Case Report

Vascular malformation of the parotid gland: a rare case report

Chisel Bhatia*, Satish Dalal, Vundavalli Sattibabu, Jagat P. Beniwal

Department of Surgery, Pt. B.D. Sharma PGIMS, Rohtak, Haryana, India

Received: 28 March 2017
Accepted: 27 April 2017

***Correspondence:**
Dr. Chisel Bhatia,
E-mail: chiselbhatia@gmail.com

ABSTRACT

Vascular malformation of the parotid gland is an extremely rare condition with very few reported cases in the literature. Here we report a case of a 55 years old, female who presented with the complaint of swelling in the right parotid region for one year. Imaging revealed a vascular malformation involving the superficial lobe of the right parotid gland. Superficial Parotidectomy was done and histopathology was consistent with the diagnosis of vascular malformation of the parotid.

Keywords: AV Malformation, Hemangioma, Parotid gland, Vascular malformation

INTRODUCTION

As parotid gland is the most common site for salivary gland tumors, benign tumors of the parotid such as Pleomorphic adenoma are commonly seen in clinical practice. However it is rare to find a vascular anomaly in the Parotid gland in adults. Vascular anomalies remain one of the least well-understood entities encountered in clinical practice. The general term 'Angioma' is still used for both tumors and vascular malformations.[1] Vascular tumors may be benign, locally aggressive or malignant. Vascular malformations can be capillary, lymphatic, venous, arteriovenous malformations or arteriovenous fistula. Here we describe a case of arteriovenous malformation of the right parotid gland. FNAC or cytology or surgical resection without making a radiological diagnosis can give rise to complications in such cases.[2] It is, therefore, important to be aware of the presenting symptoms and a possible diagnosis of vascular malformation of the parotid gland.

CASE REPORT

A 55 year old, female presented to the outpatient department with complain of swelling in front and below the right ear for one year which occurred spontaneously and gradually progressed in size. The swelling was described as painless, not related to meals and increased in size on tilting the face towards the right side (Turkey Wattle Sign). The swelling also increased in size in hot weather. On physical examination, the patient was found to have a soft, non-tender, non-fluctuant, well defined mass in the right parotid region measuring around 3 cm x 3 cm (Figure 1a and 1b). The swelling was compressible and non-pulsatile. There was no evidence of bruit or thrill over the swelling. There was no associated lymphadenopathy.

Figure 1: a) Patient presenting with swelling in front and below the right ear; b) Swelling increasing in size on dependency (Turkey-Wattle sign).

Ultrasonography of the right parotid region reported a compressible swelling comprising of tubular channels showing mixed flow on color doppler suggestive of an arterio-venous malformation. Following this, an MRI of the right parotid region was done which showed enhancing tubular structures involving the superficial lobe of the right parotid gland suggestive of a vascular malformation (Figure 2).

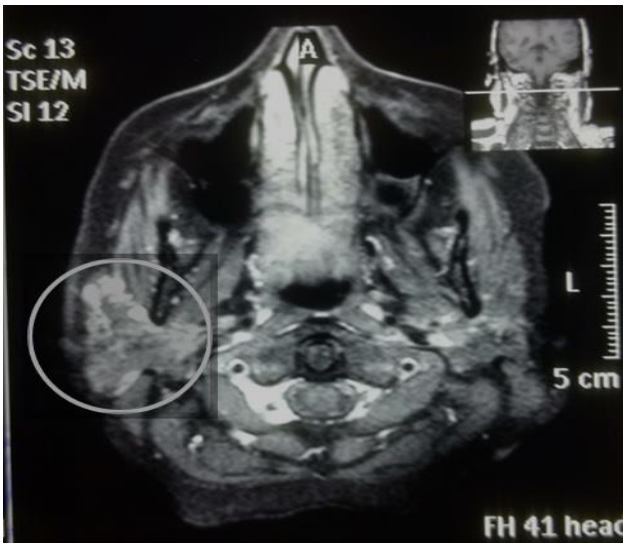

Figure 2: T2 weighted fat suppressed MRI showing Right parotid enlargement with multiple tubular enhancing structures traversing through the superficial lobe of right parotid gland.

Patient was planned for Superficial Parotidectomy. Intra operatively there was a diffuse, vascular enlargement of the superficial lobe of the right parotid gland. No major feeding vessels could be identified during the procedure. There was minimal blood loss during surgery. Facial nerve and its branches were identified and preserved (Figure 3).

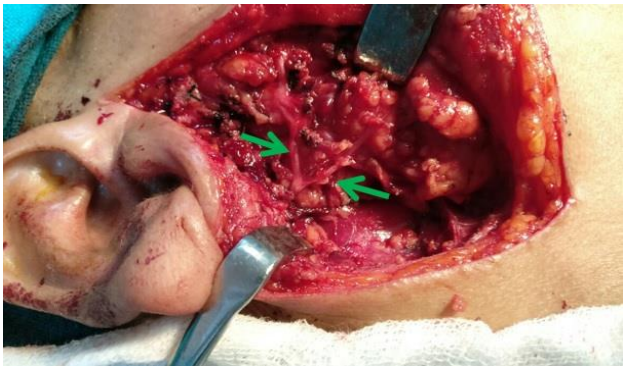

Figure 3: Intra operative photograph showing the upper and lower divisions (Green arrows) of the Facial nerve after excision of the superficial lobe of parotid.

Patient developed neuropraxia of the right facial nerve which improved gradually over a period of two months. Histopathological report of the specimen showed increase in parenchymal fat with focal areas revealing dilated and congested vessels (Figure 4). Post-operative period was uneventful.

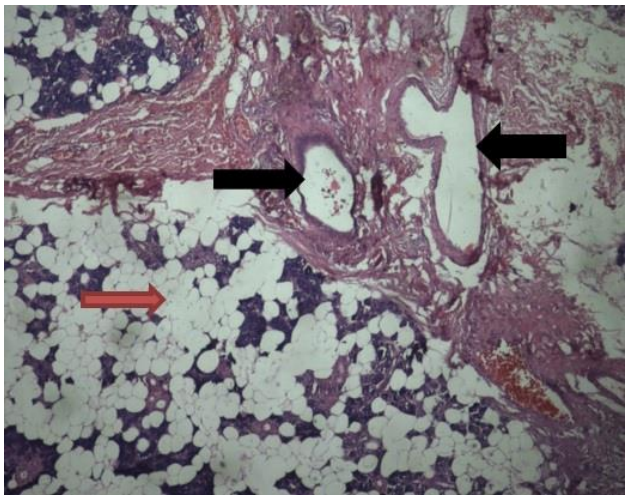

Figure 4: Photomicrograph of Superficial Parotidectomy specimen showing increase in parenchymal fat (Red arrow) with focal areas revealing dilated and congested blood vessels (Black arrows) (H and E, 4x).

DISCUSSION

Classification of vascular lesions has been a topic of debate for many years and the understanding of these disorders is still controversial. Before 1980s, vascular lesions were referred to as Hemangiomas. Mullikin and Glowacki, in 1982, for the first time classified vascular lesions into haemangiomas and vascular malformation based on endothelial characteristics.[3] The terms hemangioma and vascular malformations have been used interchangeably in the past few years. For this reason, the ISSVA (International Society for Study of Vascular Anomalies) recently adopted a classification system that distinguishes vascular tumors (lesions with clear manifestations of cell proliferation) from vascular malformations (due to innately perturbed vascular morphogenesis).

Vascular malformation of the Parotid gland is an extremely rare condition with around 50 reported cases in the literature.[2,3] In the literature, the frequency has been noted at 0.5% for Bears et al, who reported 760 parotid tumors, and at 0.6% for Byars et al, who reported 460 parotid tumors.[4,5] In a more recent study conducted by Achache M et al, parotid malformations accounted for only 1.6% (10 out of 614 patients) of all parotid tumefactions collected over the 10-year period from 1998 to 2008. There was a clear female preponderance in most of the studies with no apparent side predominance. Majority of the cases involved only the superficial lobe of the parotid.[1] Vascular malformations are seen as painless, slow growing soft tissue enlargement. There are usually

no associated symptoms and patient often visit the clinic for cosmetic purpose. There is no evidence of facial nerve involvement or lymph node enlargement or cutaneous infiltration. On clinical examination, 'Turkey wattle sign' may be seen. The turkey wattle sign describes enlargement of a facial mass on dependency of the head and when the sign is present it is pathognomonic of a vascular malformation or haemangioma.[6]

The turkey wattle is a red vascular structure in the neck of the male turkey that can increase in size when filled with blood. The diagnosis can be made clinically but as the condition is very rare, precise clinical imaging is warranted. Most commonly only the superficial lobe is involved but whole of the parotid may also be involved in few cases.

Ultrasonography can detect vascular anomalies but is less sensitive. AVM in head and neck region, reported in the literature, had high T2-weighted signal intensity and were enhanced by gadolinium.[2] Therefore MRI is the investigation of choice in these cases. In cases difficult to diagnose, an MR angiography may be indicated.

The primary goal of treatment is to restore and preserve function, stop bleeding, and improve or restore cosmesis. Treatment options include laser, cryotherapy, embolization and corticosteroids but as the diagnosis is unclear in most of the cases; surgical resection remains the gold standard of treatment.

Histopathological examination shows the presence of dilated ectatic venules lined by flat endothelial lining. Vessels may show congestion, thrombosis or calcifications. Stroma shows the presence of adipose tissue, lymphoid follicles and smooth muscles. IHC shows positivity to vascular markers such as CD31 and CD34 while lymphatics are specifically stained by D2-40 or podoplanin.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Bhatia C, Dalal S, Sattibabu V, Beniwal JP. Vascular malformation of the parotid gland: a rare case report. Int Surg J 2017;4:2081-3.