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Case Report

The clinico-radiological spectrum of masseteric venous malformation: Case report

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ABSTRACT

Due to its location, intra-masseteric venous malformation often mistaken for a parotid swelling and clinical examination alone frequently underestimates the deep extension of the lesion and rarely gives accurate pre-operative diagnosis. But once it is diagnosed, the feasibility of the treatment and the plan of approach depend on accurate delineation of the extent, size and location of the lesion. Therefore, complimentary radiographic studies are essential for its management. In this case report, typical features of venous malformation within the masseter muscle, including clinical findings (turkey wattle sign) and imaging are presented. Ultrasonography and CT scan were non-contributory in the diagnosis, while on MRI, masseteric venous malformations have a typical appearance that allowed early identification, patient education and its management.

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1. Introduction

Intramuscular vascular malformations (IMVM) are uncommon tumours in the head and neck region. Less than 1% of vaso-formative tumours throughout the body occur in skeletal muscle; 15% of them arise in head and neck musculature. The masseter muscle is the most common site, which accounts for approximately 5% of all IMVMs in the head and neck region,¹ although the origin of vascular malformations is still not clear. Errors in morphogenesis,

genes and the causative mutations have been identified for several inherited vascular anomalies.² One hypothesis postulates that placental cells, such as the trophoblast, may be the cell of origin for vascular malformations.³ It has also been suggested that they arise from malformed tissue that has been subjected to repeated trauma, surgery or hormonal influences caused by birth control pills, puberty and pregnancy and become more symptomatic. Histopathologically, these are divided into five subtypes based on the involvement of the constituent vessels: capillary, venous, arterial, lymphatic and combined forms.²

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These lesions may also be associated with significant functional and cosmetic deformities. Because of the rarity of these tumours, their deep location, and unfamiliar presentation, inaccurate preoperative diagnosis and inappropriate treatment planning are not uncommon. A case of IMVM of masseter is presented to illustrate the typical clinical presentation and MRI features, which should alert the dental physician to include the possibility of such lesions in their differential diagnosis.

2. Case description

A 30-year-old male presented with a swelling over the right facial region since four months. The patient was apparently alright four months back, and then got tooth 48 extracted due to pain and swelling, which was diagnosed as cellulitis on USG (Ultrasonography). Even after extraction, swelling did not subside and the patient was referred to our hospital for diagnosis and management. There was no history of trauma, allergy or any kind of medical problems.

Head and neck examination was normal except for the presence of swelling on right angle region of the mandible (not causing elevation of the ear). Swelling was compressible in nature (Fig. 1a), as it got reduced in size while applying pressure and reappeared with the same size after leaving the pressure. It was more apparent on clenching or when the patient lay down for several minutes on that side (Fig. 1b). This was the classical “turkey wattle sign,” i.e., a swelling that appears only in the dependent position. No pulsations or bruits were evident on auscultation. Overlying skin appeared to be normal. Scar mark was seen on the ramus region of the mandible, which was present from the childhood as the patient stated. Lymph nodes were not palpable. Intra-oral examination revealed oral sub-mucous fibrosis. Provisional

diagnosis of vascular lesion was made and in differential diagnosis, residual infection with respect to extracted 48 region was also considered.

Orthopantomographic and ultrasonographic findings were non-contributory (Fig. 2a and b). CT scan revealed mild sclerosis on several coronal cuts with significant increase in the muscle mass on right masseteric muscle region (Fig. 2c). On MRI, a diffused multi-loculated lesion was present on right masticator space involving all the muscles of mastication. The lesion was isointense on T1 (Fig. 2d) and hyperintense on T2 weighted image (Fig. 2e). Medially, it was causing partial effacement of the right parapharyngeal space and posteriorly, and was extended up to the right temporo-mandibular joint without its involvement. There was a moderate enhancement on post-contrast (Fig. 2f).

On the basis of the history, clinical presentation and radiographic findings, a presumptive diagnosis of an IMVM was made. Because the lesion was essentially asymptomatic, no treatment was initiated. Periodic observations were advised because of the possibility of continued, albeit slow, growth with symptom development.

3. Discussion

Many terms have been used to refer these lesions but the most accepted one is “vasoformative tumours”.⁴ Hemangiomas are tumours identified by rapid endothelial cell proliferation in early infancy, followed by involution over time; all other abnormalities are malformations resulting from anomalous development of vascular plexuses.⁵ Vascular malformations, which include venous malformations, are vascular lesions that are present at birth and grow commensurately with the child and clinically manifest until late infancy or childhood.⁵

Fig. 1 – (a) Showing swelling on the right angle region of the mandible causing asymmetry of the face. (b) Showing swelling on the right angle region of the mandible, which becomes more prominent on clenching.

Fig. 2 – (a) Panoramic radiograph shows anterosuperior flattening of the right condyle and mesio-angularly impacted 38. There was no evidence of any calcification or phlebolith associated with swelling. **(b)** Gray scale sonography scan showing cellulitis on right cheek with no evidence of pus and phleboliths. **(c)** Axial CT scan showing mild sclerosis of bone and significant increase in all masticatory muscle mass on right side. **(d)** Axial view of T1 weighted MR scan shows diffused multi-loculated homogenous isointense lesion, involving all the muscles of mastication on right side, causes partial effacement of the right parapharyngeal space. **(e)** T2-weighted coronal image showing diffuse multi-loculated homogenous hyperintense lesion involving all the muscles of mastication on right side causing partial effacement of the right parapharyngeal space. **(f)** Contrast-enhanced axial T1-weighted image showing homogenous moderate enhancement of the lesion.

IMVM usually present in the third decade of life and both sexes are equally affected.⁶ Their typical slow growth may cause them to be asymptomatic for many years. Pain is a variable symptom, which depends on the size of the lesion. Discoloration of the overlying skin is unusual because of its depth. The presence of pulsations, thrills or bruits is rare because of the sluggish nature of the blood flow.⁶ IMVM can vary in consistency from a soft, compressible mass to very firm mass. Many patients report transient increases in the size of their swelling, when they assume a horizontal position or the head is in a dependent position, served as a key clue.⁷ It was one of the chief signs in indexed case.

Clinically, an IMVM can be confused with salivary gland tumours, benign masseter muscle hypertrophy, myositis ossificans, benign and malignant tumour of muscle, osteogenic sarcoma, chondrosarcoma hemangiopericytoma, congenital cyst, lymphadenopathy, sialoceles of parotid duct, parotid neoplasm, schwannoma and lymphangioma. Manoeuvres that increase venous pressure such as recumbency, valsalva or jugular vein compression may make the tumour more apparent. Pulsations, bruits or thrills when present indicate the vascular lesion. The use of sialography can be used to differentiate the parotid from extra-parotid masses.¹ Once the clinical diagnosis is made, radiologic investigations like plain radiography, ultrasonography, computed tomography and MR imaging will aid in diagnosis.

Plain radiographs help in evaluating bony involvement by the lesion and phleboliths (calcified calculi). Although phlebolith are very uncommon in the oral region, their presence suggests an IMVM, but it can occur in the absence of vascular lesions also.⁸ In this case, no evidence of calcification was found on imaging (conventional, USG, CT and MRI).

Ahuja et al., in their study on 30 patients, concluded that ultrasound with high-resolution transducers can confidently suggest the diagnosis of IMVM in up to 90% of cases, when compared with MRI.⁸ In indexed case, the lesion is deeply seated in the masseter muscle and could not be seen on USG.

Fine et al. reported that the CT scanning with contrast is extremely sensitive in detecting calcification (phleboliths) and to see anatomic origin of the tumour, but provides less soft tissue detail than MRI.⁹ MRI not only provides images with good soft tissue definition of both normal anatomy and pathology, but also sensitive to blood flow within the vessels and more easily defined the nature and extent of vascular malformations. Angiography is another imaging modality, which may delineate the vascular nature of an IMVM and the feeding artery.^{1,10} Depending on the areas of involvement, despite being benign lesions, venous malformations may be associated with significant functional and cosmetic deformity. Management of IMVM should be individualized based on tumour location, accessibility, depth of invasion, patient age and cosmetic considerations. The majority of venous malformations can be managed

conservatively (observation, compression, anticoagulation α -interferon and steroid therapy).¹¹ Lesions causing significant pain or functional compromise may benefit from sclerotherapy or in some, embolotherapy and surgical debulking or removal are the helpful procedures. Lesions that are diffuse, large and deep are generally less responsive to all treatment options.¹²

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Conflicts of interest

The authors have none to declare.

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