Short communication

Lymphatic malformation of the masseter: A case report

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ABSTRACT

Intramuscular vascular malformations are rare tumours occurring in less than 1% of skeletal muscles throughout the body, about 15% arising in the head and neck. Of these, lymphatic malformations are encountered to a lesser extent, being common in the tongue and the masseter being very rare. We present a case report of a lymphatic malformation of the masseter muscle in a 27 year old male. He presented with a swelling on his right cheek that had developed 1 year ago. Fine needle aspiration (FNA) of the lesion and Computed Tomography (CT) scans were done following which the provisional diagnosis of lymphatic malformation in the masseter was made. Surgical excision of the lesion was performed subsequently and histopathological examination confirmed the diagnosis. It is very rare to find a lymphatic malformation in the masseter and with such late presentation. Follow up should be done to note for potential recurrence.

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1. Introduction

Vascular malformations are not true tumours but congenital abnormalities that demonstrate no endothelial cell proliferation. These are generally present at birth, do not resolve spontaneously and become more apparent as the child grows, unlike hemangiomas.¹ They are further classified into four subgroups based on the type of blood vessel predominantly affected: capillary, venous, arterial, and lymphatic.² Lymphatic malformations can occur on the tongue, floor of the mouth and cervico-facial region, leading to profound facial asymmetry and disfigurement.

These malformations appear as a type of birthmark and are almost completely visible superficially. An intramuscular lesion is comparatively rare.³ Less than 1% of these occur in skeletal muscle throughout the body of which about 15% of them arise in head and neck musculature.⁴ They are most common in the tongue musculature. The masseter is a very rare site of occurrence, especially for a lymphatic malformation, hardly any cases having being reported.⁵

2. Case report

A 27 year old male patient presented with an asymptomatic swelling over his right angle region since 1 year causing him aesthetic displeasure. There was no significant medical history or addiction. Swelling was insidious in growth, had attained its present size in about 2 months and was not associated with pain or discharge. On inspection, a well circumscribed swelling of about 35 × 20 × 20 mm in size, of the same colour as the surrounding skin was visible causing facial asymmetry (Fig. 1). The mass did not become more prominent when the patient clenched his jaw. No facial weakness or pulsations were noted. On palpation, a mass could be ascertained within the buccal tissues, anterior to the right ramus-angle region. It was firm, elastic, mobile and non-tender with no rise in temperature. A solitary lymph node in the right submandibular region was palpable. Intraorally, oral hygiene was good, no tooth was carious and there was no ulceration.

FNA of the mass was performed, which revealed a clear fluid which was cytologically confirmed as lymph. CT of the face (Fig. 2) revealed an ill-defined multicystic lesion measuring about 46 × 20 × 26 mm with minimally enhancing septation in subcutaneous plane around the angle of the mandible, extending in the right cheek superficial to the masseter. There was absence of lytic or erosive changes on the bone. Few lymph nodes about 11.2 mm were noted in right submandibular region. A diagnosis of lymphatic malformation was thus provisionally made.

Subsequent surgical excision of the lesion was done. The patient was positioned supine and general anaesthesia was given through

Abbreviations: CT, Computed Tomography; FNA, Fine needle aspiration.

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Fig. 1. Preoperative photograph and CT scan (Axial section) revealing an ill-defined multicystic lesion around the right angle of mandible.

Fig. 2. Excised specimen with Histopathology demonstrating dilated thin walled lymphatics with adjacent congested vessels.

nasal intubation. Following aseptic measures incision was placed over right submandibular region. Dissection was begun subcutaneously and carried out till the lower border of the mandible. The anterior fibres of the masseter were stripped from the body-ramus region of the mandible. Functional dissection was carried out intramuscularly. The lesion was identified and its excision was performed in totality. Haemostasis was achieved. The excised tissue was subjected to histopathological evaluation. Grossly it was red, had a smooth surface and measured about $25 \times 15 \times 15$ mm. On microscopic examination, thin walled lymphatics, dilated vessels with focal chronic inflammatory exudate, few giant cells and haemorrhagic areas were seen. This confirmed the diagnosis of lymphatic malformation of the masseter.

One year post operatively the patient is relived and doing fine and has been kept on regular follow up for any potential recurrence.

3. Discussion

Lymphatic malformations may arise due to failure of the lymphatic system to connect/separate from the venous system,

abnormal budding of lymphatic structures from the cardinal vein, abnormal sequestration of lymphatic tissue in early embryogenesis or due to acquired processes – trauma, infection, chronic inflammation or lymphatic obstruction.⁶

Lymphatic malformations can reach various depths and when present deeper they become difficult to diagnose, being evident only when they cause functional or aesthetic compromise such as macroglossia.⁷ Their growth may be inhibited by the relatively tougher tissues surrounding the lesion.⁶ The accentuation if the size of the malformation can occur at some point of time due to factors like injury, inflammation, lymphatic obstruction and endocrine changes.⁸ which is what might have occurred in our patient as well. Histologically these can be microcystic, macrocystic or mixed. All types of lymphangiomas occur together in a patient and even lymphatic and vascular malformations can coexist.⁶

Treatment varies according to the age of patient, site of lesion, extensions and presence or possibility of functional compromise. Modalities used include steroids, cryotherapy, sclerotherapy, radiotherapy, CO₂ lasers and surgical removal. Suprahyoid microcystic lymphatic malformations are more difficult to treat than macrocystic lesions in the infrahyoid and posterior cervical regions and warrants surgical excision. Bilateral suprahyoid lymphatic

malformations may require staged treatments in order to prevent complications.⁹

When in a diagnostic dilemma, these malformations should be differentiated from parotid swellings and similar appearing lesions, as management varies completely. Intramuscular haemangiomas, for which masseter is a common site on MRI appear as enhancing lesions in contrast to the non-enhancing lymphatic malformations.¹⁰ In venous lesions, an increase in size with procedures that increase venous pressure such as dependent position, valsalva maneuver, compression of jugular vein can be demonstrated. Arterio-venous malformations can be distinguished on the basis of a solid parenchymal tissue lining. Ultrasonography with Doppler imaging can also be used to demonstrate arterial or venous lesions.³

Recurrence can occur in these lesions since these are a part of a lymphatic system.⁵ It depends upon the patient's age, site of occurrence, histological type and the post treatment surgical margins⁸ so these should be taken into consideration during the surgical removal. The importance of this case report is in the unique presentation of this disease at this site of the masseter as well as the late age of presentation which is quite unusual.

4. Conclusion

Lymphatic malformations if left undiagnosed and untreated can reach enormous sizes, causing various disastrous aesthetic and

functional sequela. Early diagnosis and proper treatment of this infrequent vascular malformation is essential to prevent profound complications later.

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