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RESEARCH ARTICLE

INTRA-MASSETERIC VENOUS MALFORMATION WITH PHLEBOLITHS – A RARE CASE REPORT

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ABSTRACT

Background: Intramuscular vascular malformations are uncommon tumours in the head and neck region. The masseter muscle is the most common site. Approximately 40% of the venous malformations are located in the head and neck region and occasionally are associated with phleboliths. Because of their rare occurrence, deep location and unfamiliar presentation, these malformations are often difficult to diagnose and treat subsequently. **Objective**: The objective of the study is to formulate a correct diagnosis and treatment plan when this type of lesion is encountered to prevent untoward complications or recurrence. **Case Report**: We present a case of a middle aged lady, presenting with secondarily infected intra masseteric venous malformation with phleboliths. The lesion was excised in toto along with the masseter muscle and the patient was followed up for 6 months. **Conclusion:** Any lesion in the region of the parotid or angle-ramus must be evaluated thoroughly prior to surgery. In cases of Intra masseteric venous malformations, where surgery is the primary option, total masseteric excision should be the treatment of choice as there is negligible cosmetic or functional disability, a better chance of clearance and reduced incidence of recurrence.

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INTRODUCTION

Intramuscular vascular malformations (IMVM) are uncommon tumours in the head and neck region (Agarwal, 2015). Less than 1% of these vascular malformations occur in the skeletal muscle, amongst which around 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 (Wolf, 1985). The etiopathogenesis of these malformations remain obscure. Abnormality in Morphogenesis, genetic abnormalities and causative mutations have been identified as potential causes for several inherited vascular anomalies (Dompmartin, 2010). One hypothesis suggests that differenciated placental cells, such as the trophoblast, may give origin to the vascular malformations. Other possible causes include repeated mechanical and surgical trauma, oral contraceptive pills, puberty and pregnancy. Histopathologically, these are divided into two groups depending on their hemodynamic and lymphodynamic characteristics: low flow and high flow malformations. Low flow lesions include capillary, venous and lymphatic malformations or a combination of these elements. High flow lesions include arterial malformations and arterio-venous malformations (Burns et al., 2009). Approximately 40% of the venous malformations are located in the head and neck region and occasionally are associated with phleboliths. Therefore, these calcifications are considered typical features of venous malformations. Clinically, these IMVM may present with cosmetic and functional deformities. Because of their rare occurrence, deep location and unfamiliar presentation, these malformations are often difficult to diagnose and treat subsequently. We present a case of a middle aged lady, presenting with secondarily infected intra masseteric venous malformation with phleboliths. The lesion was excised in toto along with the masseter muscle and the patient was followed up for 6 months.

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CASE REPORT

A 35-year-old female presented with a mild painful swelling (Fig. 1) over the right facial region since four months. There was no history of trauma, allergy, chronic systemic illness or medications. Head and neck examination was normal except for the presence of swelling over the right angle region of the

mandible (not causing elevation of the ear). Swelling was compressible in nature, as it got reduced in size while applying pressure and reappeared with the same size after leaving the pressure. Diascopy test was positive. It was more apparent on clenching or when the patient bent forward, with the level of her head at a lower position than her heart. No pulsations or bruits were evident on auscultation. Overlying skin appeared to be normal. A small fistulous tract was present at the lower border of the mandible (Fig. 1), which expressed pus on application of pressure. Cervical lymph nodes were not palpable. On aspiration of the lesion, frank blood was obtained. Intra-oral examination revealed normal findings with normal colour and texture of the mucosa. Mouth opening was slightly decreased with maximum inter incisal opening of 28mm. A provisional diagnosis of secondarily infected vascular lesion was made and empirical antibiotics were started to control the infection. The patient was advised a CT scan and MR Angiography to reveal the exact anatomic location of the lesion and to determine the feeder vessels. On axial section, the CT scan revealed significant increase in the muscle mass on the right masseteric muscle region along with the presence of a few foci of hyperintense attenuations suggestive of calcifications scattered intramuscularly, probably phleboliths (Fig. 2).

On MR Angiography, a large altered signal intensity T2 hyperintense lesion was seen in the substance of right masseter muscle with few nodular hyper intensities and center blooming (Fig. 3). No obvious arterial feeder vessels were noted (Fig. 4). Based of the history, clinical presentation and radiographic findings, a presumptive diagnosis of an IMVM with phleboliths was made. The patient was planned for removal of the lesion under General An aesthesia. The lesion was accessed via an extra oral submandibular incision with preauricular extension (Fig. 1), exposing the lateral surface of the ramus and the masseter muscle. Small vessels supplying the lesion were ligated and the entire lesion along with the masseter muscle was excised. Fig. 5 shows the post excision surgical defect and Fig. 6 shows the surgical specimen and the presence of phleboliths within it. A surgical drain was inserted and the incision was closed in layers. The fistulous tract was excised separately and sutured (Fig. 7) Post-operative healing of the patient was uneventful and uncomplicated. The surgical drain was removed on the 2nd post-operative day and the patient was discharged on the 5th post-operative day after removal of the facial sutures. Mouth opening of the patient improved significantly and there was no evidence of any recurrence in the follow up period of 6 months (Fig. 8)

DISCUSSION

Several terms have been used to refer these lesions but the most common and accepted one is ‘‘vasoformative tumours” (Rai, 2006). Hemangiomas are true endothelial cell tumours characterised by rapid endothelial cell proliferation in early infancy, followed by involution phase; all other abnormalities are described as malformations resulting from anomalous development of vascular plexuses (Mulliken, 1982). Vascular malformations, includes high flow and low flow lesions. Venous malformations, a subtype of the low flow vascular lesions that are present at birth and grow commensurately with the child and manifests clinically in late infancy or childhood (Mulliken, 1982). IMVM usually present in the late second or third decade of life without equal sex predilection (Conley, 1977). They usually remain asymptomatic over the years owing to their slow growth and lack of pain, unless secondarily infected. Superficial venous malformations typically present as a soft, compressible, bluish mass. However, these features may not be fully apparent in intramuscular or deeply situated lesions. The presence of pulsations on palpation and thrills or bruits on auscultation is rare because it is a low flow lesion and the blood flow is sluggish in nature (Conley, 1977). Many patients, such as ours, report transient increases in the size of their swelling, when they assume a horizontal position or the head is in a dependent position, held in a position lower than their heart. This feature, also known as the Turkey Wattle sign is a pathognomic feature of vascular malformations. The presence of this sign along with blood on aspiration from the lesion in our patient gave us a cue to our diagnosis of a vascular malformation. Manoeuvres that increase venous pressure such as recumbency, valsalva or jugular vein compression may make the vascular lesion more apparent or it may increase in size.

Differential diagnosis of IMVM may include benign salivary gland tumours, masseter muscle hypertrophy, myositis ossificans, muscle neoplasms, soft tissue sarcomas, sialocele of parotid duct and lymphangioma. After a provisional diagnosis is made, radiologic investigations like ultrasonography, computed tomography and MR Angiography will aid in formulating a final diagnosis. 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 alone (Ahuja, 2003). CT scanning with contrast is extremely sensitive in detecting calcification (phleboliths) and to see the exact anatomic origin of the tumour but provides less soft tissue detail than MRI (Elahi, 1992). MRI provides good soft tissue definition of both normal anatomy and pathology and defines 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 (Rossiter, 1993). In our case, MR Angiography did not demonstrate any significant arterial feeder vessel. Phleboliths associated with vascular anomalies were initially found in the splenic vein by Canstatt in 1843 and later was seen in the maxillofacial region by Kirmission in 1905. Ribbert’s theory suggests that phlebolith formation begins with intravascular thrombus formation which is followed by progressive lamellar fibrosis and subsequent calcification. Because of the sluggish blood flow in venous malformations and occasional stasis, the chances of formation of phleboliths are more. Radiographically, they manifest as multiple round and oval laminated bodies with either radiolucent or radiopaque cores (Scolozzi et al., 2003). Management of IMVM should be patient specific based on tumour location, extent, size, accessibility, depth of invasion, type of flow, patient age and cosmetic considerations. Smaller lesions may be managed conservatively (observation, application of compression bandage, cryotherapy, anticoagulation therapy by a- interferon and steroid therapy) (Legiehn, 2008). Lesions causing significant pain or functional compromise may benefit from sclerotherapy, embolization of the major feeder vessels or surgery which at present remains the treatment of choice. Lesions that attain massive sizes, are diffuse and deeply seated causing severe functional and aesthetic morbidity, show less response to the prevalent treatment options (Deveikis, 2005). In our patient, the lesion was secondarily infected with pain as a presenting feature. Phleboliths were also observed within the lesion. These factors made surgery as our primary treatment option.

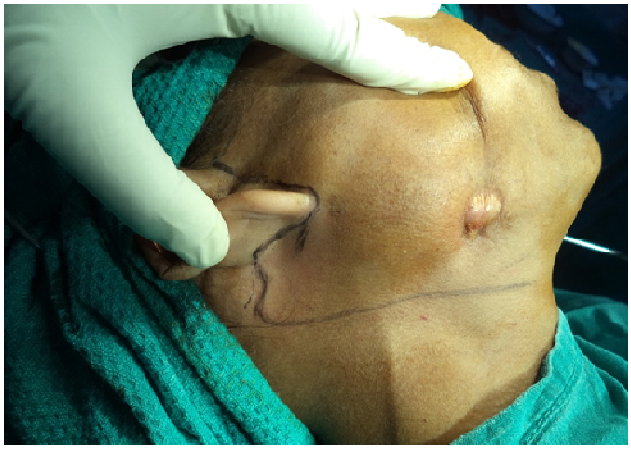

Fig. 1(Pre-Operative image) showing a) Mild swelling over the right angle of mandible with healed fistulous tract (after administration of pre-operative antibiotics). b) Submandibular with pre-auricular extension incision marking

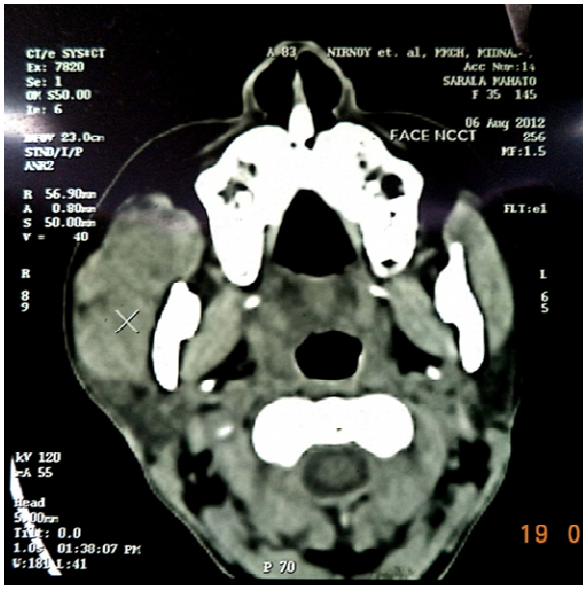

Fig. 2 (CT scan of face) showing Few foci of hyperintense attenuations suggestive of calcified phleboliths

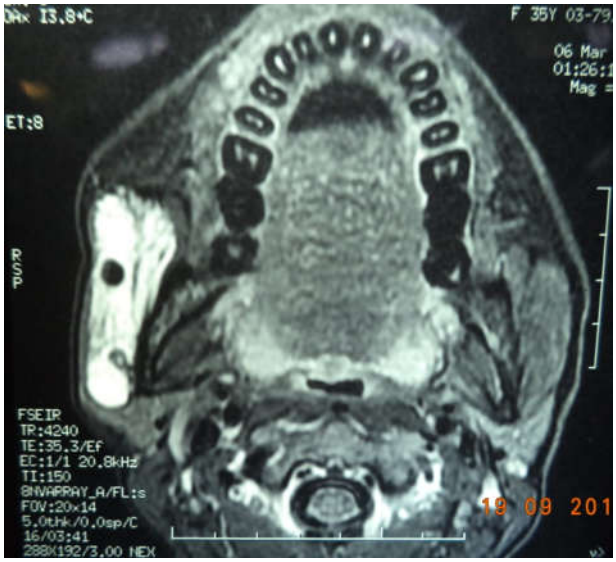

Fig. 3(MR Angiography of face) showing Hyperintense lesion in the substance of right masseter muscle with few nodular hyperintensities

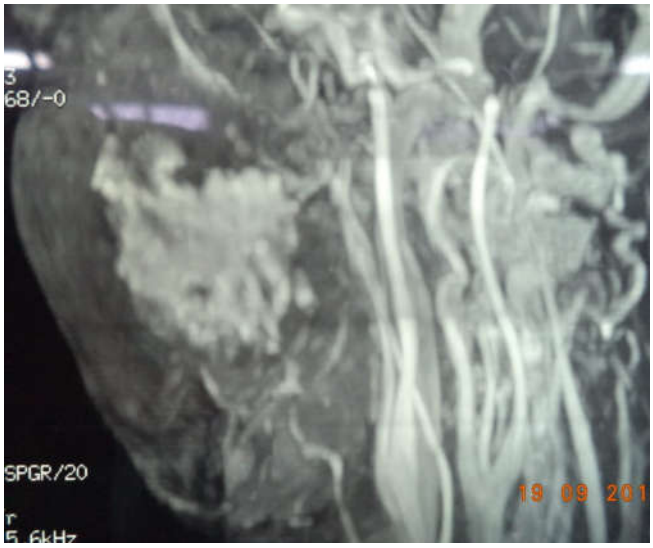

Fig. 4 (MR Angiography of face) showing No obvious arterial feeders to the lesion

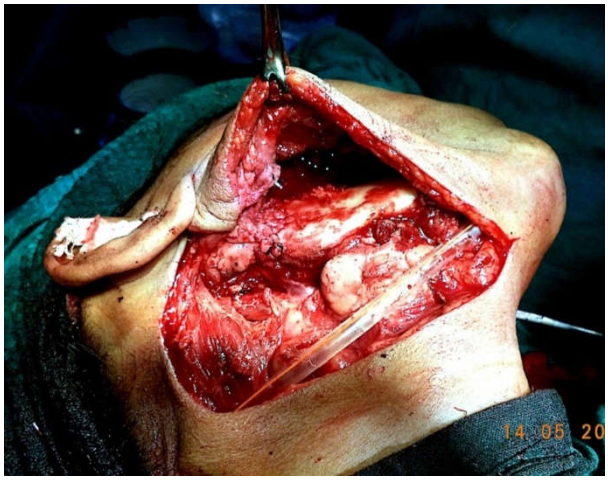

Fig. 5 showing The post excision surgical defect

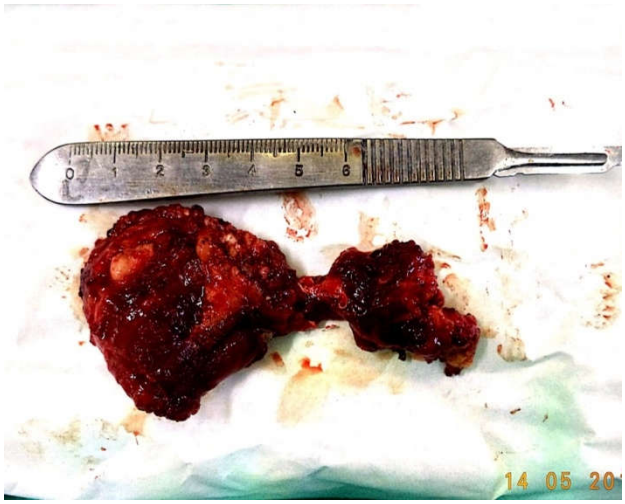

Fig. 6. Showing The excised surgical specimen

Local recurrence ranging from 9 to 28% have been reported even after wide excision (Wolf, 1985), hence we recommend total excision of the lesion along with the masseter muscle to prevent recurrence. This is associated with very little cosmetic and functional disability.

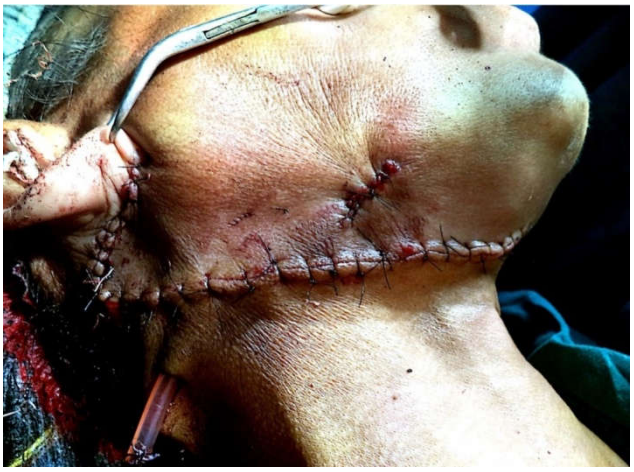

Fig. 7 showing Suturing of the main incision and the fistulous tract separately

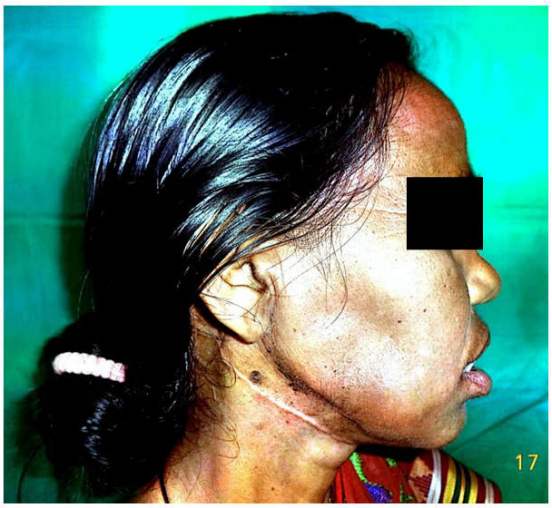

Fig. 8 showing Follow up photograph after 6 months

For ease of accessibility and wider exposure, extra oral incision was given to ensure complete excision. The patient was followed up for a period of 6 months without any evidence of recurrence and post-operative recovery was uncomplicated. Facial nerve function was intact.

Conclusion

Any lesion in the region of the parotid or angle-ramus must be evaluated thoroughly prior to surgery. If aspiration shows a bloody aspirate on repeat sessions, the possibility of a vascular lesion must be kept in mind. Clinically, the diagnosis may be confirmed by the Turkey Wattle sign. Advanced radiography such as CT scan or MRI should be the next line of investigation. Arteriography may be necessary to evaluate the presence of feeder vessels.

If extra-oral incison is given, proximal control of vessels can be obtained via the same incision, if required. Total masseteric excision should be the treatment of choice as there is negligible cosmetic or functional disability, a better chance of clearance and reduced incidence of recurrence.

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