Case Report

Surgical management of intramuscular hemangioma of left masseter muscle: A case report

ABSTRACT

Hemangiomas are benign soft tissue tumors which are congenital and occur due to abnormal proliferations of blood vessels. Most common location of hemangiomas is subcutaneous adipose tissue, but skeletal muscle hemangiomas are very rare which make up to 0.8% of all hemangiomas. Usually, the intramuscular lesions are common in thigh region and calf muscles and are relatively rare in the facial muscles. Long-standing lesions results in phleboliths, and this may cause some symptoms. Conventional treatment of these isolated lesions may not yield satisfactory results. Hence, surgical excision of the lesion in toto results in aesthetically pleasing results with low chances of recurrence. In this article, we report a case of a left masseter intramuscular hemangioma in 19-year-old patient which was successfully managed by complete surgical excision.

Keywords: AV malformation, benign tumor, hemangioma, vascular lesions

INTRODUCTION

Hemangiomas are vascular hamartomas which are common in subcutaneous areas. Intramuscular/skeletal muscle hemangiomas are relatively rare. Though it is rare, it can occur in all muscles. Intramuscular hemangioma (IMH) of muscles in facial region is rare and are rarely reported in the literature.[1] These may have varied clinical presentations with minimal or no pain. Usually, patient with IMH of facial region is aesthetically concerned and expects complete recovery after treatment.[2] Well-documented treatment modalities like intralesional sclerosing therapy or steroid injections may not yield completely aesthetically satisfying recovery. Hence, surgical treatment was planned in the present case.[3]

CASE REPORT

A 19-year-old male patient reported with a chief complaint of cosmetic disfigurement of face due to swelling in the left side of the face which had been slowly growing in size since childhood. On extra-oral clinical examination [Figure 1], gross facial asymmetry was detected with a diffuse palpable firm soft tissue swelling in the left middle and lower third of face. The skin over the swelling was normal in color, and no other abnormalities were observed. The patient had previously consulted a physician, and he reported with the ultrasonography (USG) report. The USG examination reported the lesion to be venous malformation with calcifications [Figure 2]. Patient had not undergone any conservative treatment before. Further investigation was advised which included panoramic radiograph and computed tomography (CT) angiography. In panoramic radiograph, there was evidence of multiple calcifications overlapping the areas of left maxillary tuberosity and mandibular

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Received: 03 August 2022, **Revised:** 05 September 2022, **Accepted:** 09 December 2022, **Published:** 19 March 2024

Access this article online	
Website: www.njms.in	Quick Response Code
DOI: 10.4103/njms.njms_140_22	

How to cite this article: Shah R, Venkatesh R, Badi K, Shah K. Surgical management of intramuscular hemangioma of left masseter muscle: A case report. Natl J Maxillofac Surg 2024;15:160-3.

ramus region suggestive of phleboliths [Figure 3]. IN CT angiography, there was a evidence of approximately 5 × 25 × 60 mm (AP × TR × SI) sized well-defined heterogeneously enhancing soft tissue density lesion seen in the superficial and intramuscular planes along the left masseter muscle. Laterally, the lesion was reaching up to the skin. Medially, the lesion was overlying the left ramus of mandible. Postero-laterally, the lesion is causing lateral displacement of the superficial lobe of the parotid gland. On contrast, multiple vessels are seen traversing through it with multiple chunky areas of calcifications [Figure 4]. The features were suggestive of IMH. The patient underwent surgery, direct cheek incision was made on the site of the lesion 10-12 mm behind the Zuker's point, and extracapsular dissection was done until the superior portion of masseter muscle [Figure 5] was identified. 2% Methylene blue dye was injected to identify the lesion. Once the lesion was stained, the stained part was excised. Total 14 phleboliths were identified and removed [Figure 6]. Again 2% methylene blue dye was injected over the lesion to check for residual lesion, and no residual lesion was detected. Closure was done in layers. The excised specimen was sent for histopathological examination. Gross specimen showed soft tissue mass of 3 × 3 cm in size, brownish to reddish in color, firm in consistency and 14 phleboliths. The pathological examination confirmed the intra muscular cavernous hemangioma [Figure 7]. One-month postoperative follow-up showed satisfactory recovery, and presently patient is under periodic review [Figure 8].

Figure 1: Extra-oral view showing left facial swelling

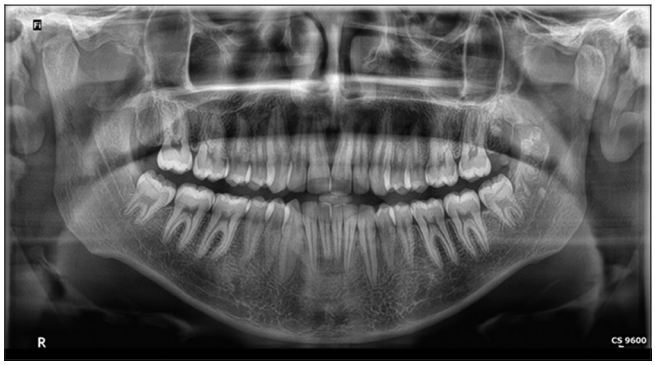

Figure 3: Panoramic radiograph showing phleboliths in left maxillary tuberosity and ramus region

DISCUSSION

A hemangioma is a type of vascular abnormality that usually develops in the trunk and extremities. About 0.8 percent of all hemangiomas are intramuscular; only 14 percent of all sites are found in the head-and-neck region. Intramuscular hemangiomas were initially described by Liston in 1843, and according to the size of the vessel, Allen and Enzinger categorized them in 1972. Three types of intramuscular hemangiomas are recognized: capillary, cavernous, and venous.[4]

Phlebolith formation within intramuscular hemangiomas is present in 15% to 25% of cases, on average. Phleboliths consist of a mixture of calcium carbonate and calcium phosphate salts. The pathogenesis of phleboliths is believed to involve thrombi produced by slowing peripheral blood flow, and then, it becomes organized and mineralized;

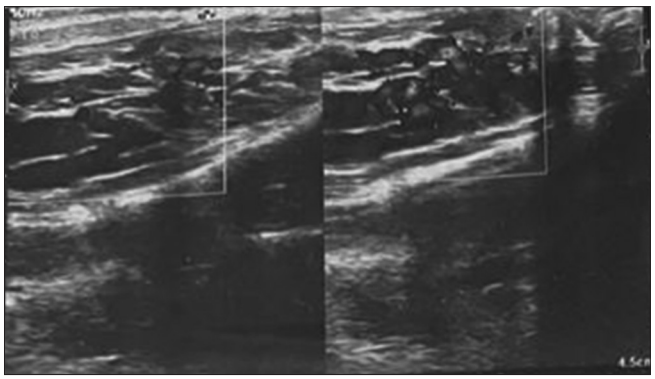

Figure 2: Multiple venous channels with prominent arc-like calcifications were identified in USG

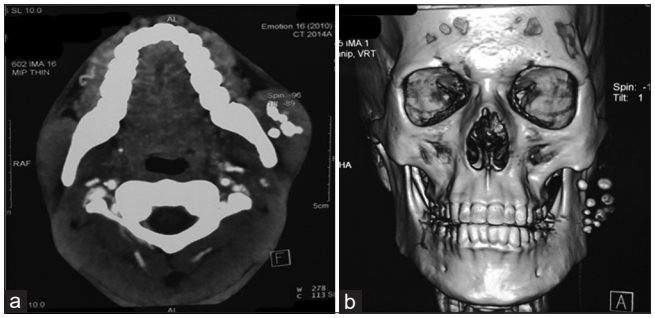

Figure 4: Axial section of CECT showing soft tissue swelling with hyperdense areas (a) and multiple calcifications adjacent to left ramus area (b)

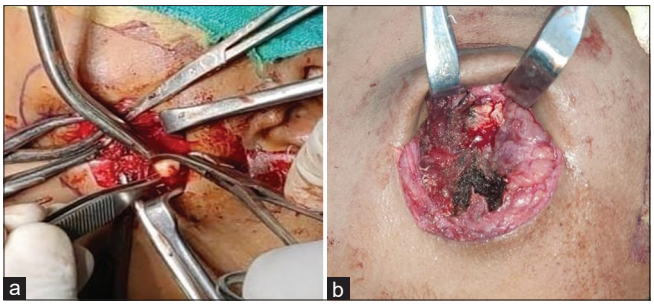

Figure 5: Intra-operative pictures showing extracapsular dissection until the superior portion of masseter muscle (a) and after excision of stained part of soft tissue mass (b)

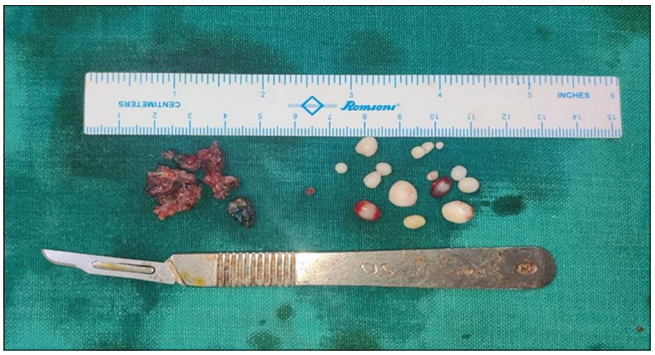

Figure 6: Excision specimen and 14 phleboliths

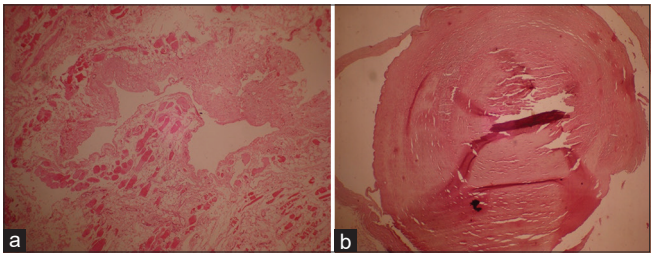

Figure 7: H and E staining shows connective tissue stroma with multiple dilated blood vessels (a); histopathology of phlebolith showing large eosinophilic area with inside basophilic area (b)

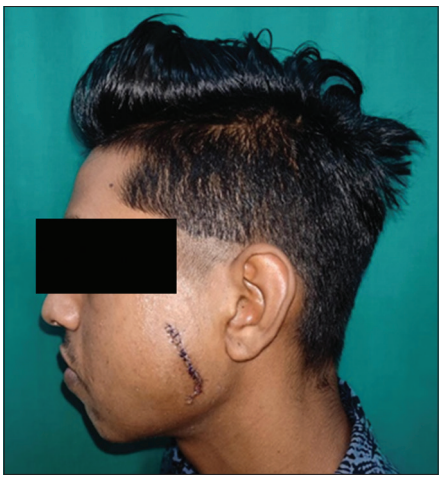

Figure 8: One-month postoperative picture showing satisfactory healing

initially, calcified thrombi occur, forming the core of the phlebolith.[5]

Although the cause of these lesions is uncertain, repetitive trauma or hormonal changes can accelerate tumor growth by causing the creation of embryonic vascular tissue.[6] Due to the high prevalence of hemangiomas in the first three decades of life, a congenital theory has been put up. Before causing any symptoms, intramuscular hemangiomas with the development of phlebolith typically manifest as a slowly expanding, painless mass and do not show any vascular indications or typical symptoms.[6,7] In present case, the history of the lesion from his childhood was given which gradually increased to present size.

As preoperative diagnostic accuracy is only 8%, CT, MRI, and ultrasound examinations are crucial.[8,9] CT scan plays an important role in the identification the size and shape of the tumor and the surrounding tissues. Calcification and no erosion of the underlying bone from the preoperative CT wee appreciated in present case.[8]

Cryotherapy, corticosteroid or sclerosant injection, isolated embolization, arterial ligature or radiation therapies have been tried as non-surgical treatments, but, however, the results of these modalities have been controversial[10] and such methods are currently recommended only where surgery is contraindicated or refused.[1] The majority of hemangiomas involving the muscle are cavernous hemangiomas, and complete resection of the tumor is the ideal to reduce the recurrence rate.[10,11] Recent case reports in the literature show surgical management of masseter muscle hemangioma has given good esthetic results with less recurrence rates.[12-14] Hence, surgical management was planned in this case keeping esthetics in mind where direct cheek incision was used which is a valuable alternative to standard approaches in resecting smaller, benign masses. The benefits of direct check incision are more valuable over the traditional or modified Blair's incision when it comes to eliminating mid-cheek tumors. Also in this, lengthy scar and a wide flap dissection are avoided, it is simpler, quicker and can be done under local anesthesia. The cheek incision can be employed during a brief hospital stay due to the less traumatic nature of the procedure and the postoperative recovery period, which cuts down on operating time, hospital stays, and expenditures.[1,6,10] Methylene blue is a safe, inexpensive, reproducible and highly accurate method of diagnosing lesions. The cationic dye methylene blue can enter the aberrant membranes of the lesions and bind to the negatively charged nucleic acids in the nuclei to enable visualisation of the residual lesion.[1,6] The largest drawback of the direct cheek incision is its visibility. As the

accessory parotid gland is directly associated with Stensen's duct and the zygomatic and buccal branches, which lay more superficially and are frequently draped over or wrapped around the lesion, there is a higher risk of salivary fistula and damage to the facial nerve.[10] The reduced operative viewing area can also make it more difficult to completely remove the mass, perhaps leading to greater recurrence rates due to insufficient excision. There is less flexibility during surgery compared to typical parotidectomy methods. However, Perzik and White claim that in certain circumstances, primary excision through a direct cheek incision may be done successfully while keeping these risks in mind.[6]

The patient reported with no postoperative complications or damage to the nerve post-surgery. Also, the cosmetic outcome was satisfying. The recurrence rates for the capillary hemangioma, cavernous hemangioma, and venous hemangioma are 20%, 9%, and 28%, respectively. Incomplete excision of the lesion and invasion of the surrounding tissue can lead to increased recurrence rate.[10]

CONCLUSION

Advanced imaging modalities like USG, CT scan, and MRI are required to achieve accurate preoperative diagnosis of intramuscular hemangioma, and well-accepted esthetically pleasing results are achieved through surgical management of such lesions.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that his name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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