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Intramuscular hemangioma of the masseter muscle- a case report and review of literature



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ABSTRACT

INTRODUCTION: Intramuscular hemangioma, is a distinctive type of vascular tumor occurring within the skeletal muscle. Most IMH are located in the lower extremity, particularly in the muscles of the thigh and rarely in head and neck region.

PRESENTATION OF CASE: 35 years old male reported with a swelling in the left cheek region since 3 years. Clinical and radiological evaluation leads to the diagnosis of Intramuscular hemangioma. Surgical excision was performed and histopathology confirmed the diagnosis.

DISCUSSION: Hemangiomas of skeletal muscle represent 0.8% of all benign vascular neoplasm Welsch and Hengerer, 1980 [4]. Of these 13.8% occur in the head and neck region, with the masseter muscle being the most common site, followed by the trapezius and sternocleidomastoid muscles respectively. The lesions previously described as deep infiltrating angioliomas have now been recognized by the WHO as intramuscular hemangiomas. Numerous theories proposed for etiopathogenesis of vascular lesions have been discussed.

CONCLUSION: In conclusion, angioliomas are rare in the head and neck region, and it should be considered in the differential diagnosis of masses in these regions. Proper radiological and clinical examination will reveal the type of vascular lesion. Excellent results can be obtained with timely management and good surgical skills.

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1. Introduction

Vascular lesions involving the skeletal musculature is an uncommon tumour characterized by the proliferation of blood vessels, occurring most frequently in the large muscles of the upper and lower extremities and trunk [1]. First described by Liston [2] in 1843, this benign vascular lesion accounts for less than 1% of all hemangiomas [3]. In the head and neck region, the masseter and trapezius muscles are most commonly involved [5]. In the former, they may mimic a parotid neoplasm [6]. The etiopathogenesis of these unusual lesions is not yet clearly understood, although trauma and abnormal sequestration of embryonic tissue have been postulated [5] because they frequently present in early childhood

or early adult life. We report a case of intramuscular hemangioma lesion occurring in the cheek region.

2. Case report

A 35 years old male patient came to the Department of Oral and Maxillofacial Surgery complaining of a swelling in the left cheek region since 3 years. On clinical examination a smooth, oval swelling of size 2 × 3 cm diameter was noted in the left cheek region (Fig. 1). It was diffuse in nature, firm, soft which becomes prominent on clenching. On auscultation mild bruit was heard. On intraoral examination the buccal mucosa was found to be free from the swelling with no evidence of any discoloration. Radiographic investigation including MRI scan and Angiogram was done and it revealed an enhancing well circumscribed intramuscular mass (Fig. 2). Provisional diagnosis of a vascular lesion was made and differential diagnosis of other soft tissue lesions like lipoma, neurofibroma, pleomorphic adenoma was ruled out.

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Fig. 1. Pre Operative.

2.1. Procedure

Surgical excision of the lesion was planned under GA. Sub-mandibular incision was placed (Fig. 3). Skin flap were raised and lower border dissection was done through masseter muscle to expose the well encapsulated lesion (Fig. 4). Once lesion is exposed a blunt dissection was done around the lesion and cauterization was done for any bleeding vessels (Fig. 5). The whole lesion was removed in toto. On the specimen a glistening capsule was seen covering the whole vascular tumour (Fig. 6). The whole exposed specimen was send for histopathology (Fig. 7).

2.2. Histopathology

Histopathology reveals that given soft tissue section shows fibro vascular connective tissue exhibiting numerous large dilated thin walled as well as smaller thick walled vascular spaces filled with RBCs. Some of the larger vessels show fibrin thrombi and basophilic calcification. The intervening connective tissue stroma is densely collagenous comprising of dense collagen fibres with spindle fibroblast, fat cells and muscle tissue (Fig. 8). Focal hemosiderin pigmentation is noted (Fig. 9).

3. Discussion

Various misnomers have been mentioned for vascular lesions involving skeletal musculature. We find the term angioliipoma most suiting to the lesion found in our case as per clinical and imaging findings. Angioliipomas are rare benign mesenchymal tumours that are distinguished from lipomas by proliferating vessels [12] and are categorized as non-infiltrating angioliipoma and less frequent infiltrating angioliipomas. The lesions previously described as deep

infiltrating angioliipomas have now been recognized by the WHO as intramuscular hemangiomas [13].

Hemangiomas of skeletal muscle represent 0.8% of all benign vascular neoplasm [4]. Of these 13.8% occur in the head and neck region, with the masseter muscle being the most common site, followed by the trapezius and sternocleidomastoid muscles respectively [1]. Other possible sites are periorbital muscle, temporalis muscle, geniohyoid and medial pterygoid. Studies show that intramuscular haemangioma mostly present before the age of 30 [4] but some studies report cases in elderly patients also. But in our case the patient is in his early thirties.

Although intramuscular hemangiomas have shown an equal sex distribution, involvement of the masseter has a definite male pre-dominance [17].

Numerous classifications of vascular lesions exist in the English literature. Mulliken and Glowacki classified vascular lesions as vascular malformations and haemangiomas, based on their clinical appearance, histopathologic features and biologic behaviour [8]. Allen & Enzinger [9] classified them histologically as (1) capillary (vessels smaller than 140 micrometer in diameter), (2) cavernous (vessels larger than 140 micrometer in diameter) or (3) mixed (consisting of both small and large vessels). Capillary haemangioma usually presents with a short history [11]. They are highly cellular thus explain the firmness and lack of clinical signs to suggest its vascular nature. Cavernous haemangioma generally present with longer history of symptoms, tend to be larger in size and painful. They are most common in the lower extremity with only 19% occurring in the head and neck [3]. Mixed type is histologically and clinically similar to cavernous type. We find the latter classification most substantiating our clinical and histological findings.

Etiopathogenesis remains unclear although various theories have been proposed to explain its etiology. The most likely explanation is that the intramuscular hemangioma is a congenital mass,

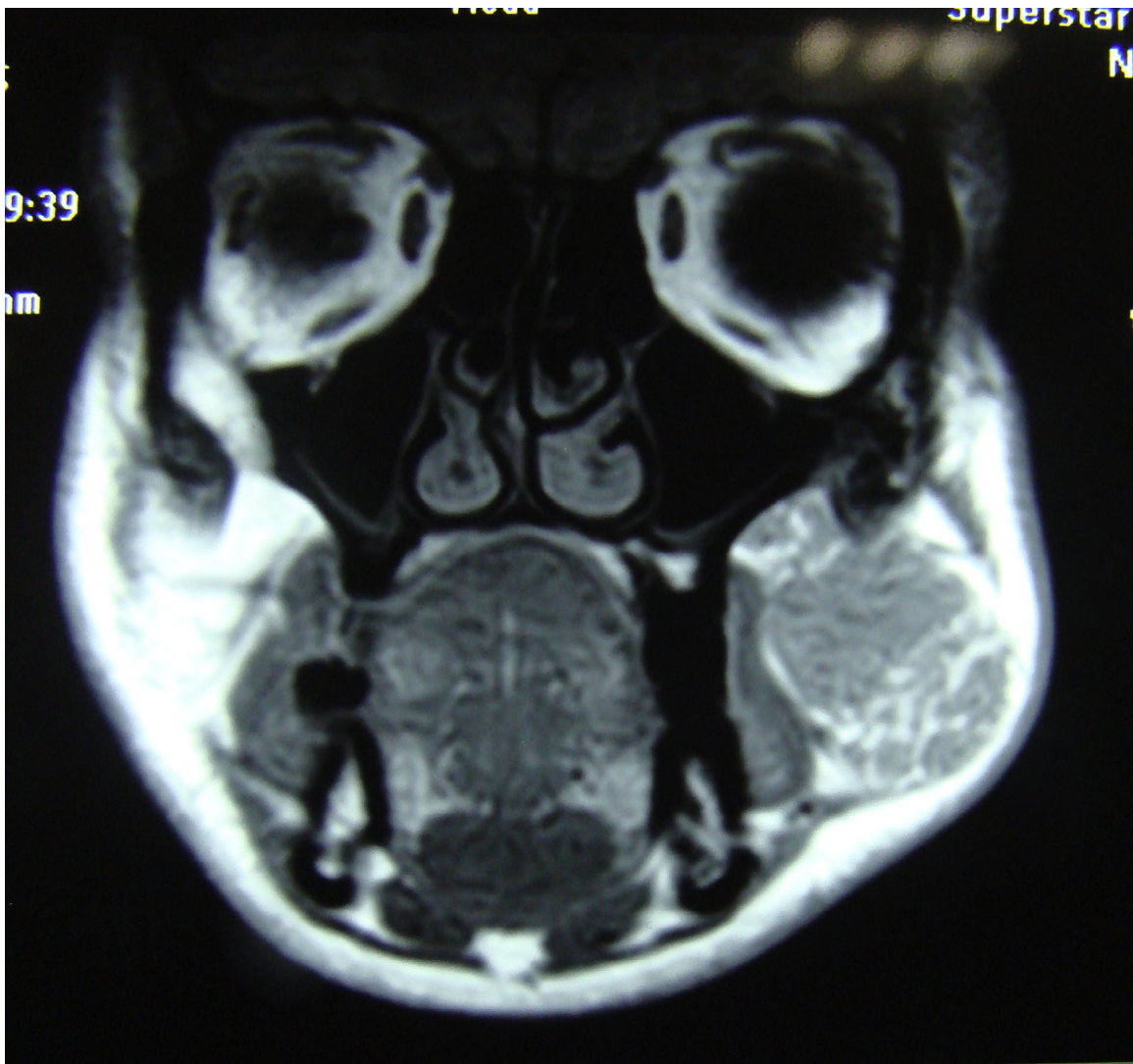


Fig. 2. Pre Operative scan (MRI scan).

arising by abnormal embryonic sequestrations, similar to congenital arteriovenous malformations [7]. Traumatic have been suggested and may contribute to the aetiology or growth spurts [4]. Hormonal role in the growth of intramuscular hemangioma was speculated, but no specific data was available to substantiate this hypothesis [6].

These tumours present as gradually enlarging mass lesions with duration often less than a year. The swelling is normally diffuse in nature, compressible and characteristically deep within the muscle. However, softness and compressibility may be absent due to local fibrosis and overlying musculature or to the prominent cellularity of capillary type tumours. Pulsations, bruits or thrills are uncommon but when present, arteriography is indicated as to identify large vessel communications [6]. There are usually no skin changes. Clenching the teeth could make the lesion to become more firm and fixed⁶.

A variety of tumours can be confused clinically with an IMH. The differential diagnosis includes salivary neoplasms, cysts, lymphangiomas, rhabdomyosarcomas, masseteric hypertrophy, and schwannomas [9].

Diagnosis of such vascular lesions is often challenging. FNAC is inconclusive in arriving at a diagnosis as it yields only a blood tinged

aspirate [14]. Super selective arteriography with subtraction clearly defines the altered vascular pattern and flow dynamics including feeder vessels and opens up therapeutic modalities. Arteriography with pre-op embolization of feeding vessels enhances haemostasis and can facilitate excision. However, it may fail to demonstrate low flow lesions adding to the diagnostic difficulty. Though contrast CT may demonstrate vascular nature of the tumour, MRI shows good tissue delineation and contrast of the lesion from its surroundings due to its multiplanar capability. Intramuscular haemangiomas are characteristically much brighter on T2 than on T1 weighted images [16].

Management of intramuscular haemangioma should be individualized according to the tumour location and extent, tumour growth rate, anatomical accessibility, patient age and cosmetic considerations [15]. Some of the patients can be observed with the accuracy of the MRI especially in young children. Many treatment modalities like cryotherapy, radiation therapy, steroid administration and embolization, sclerosing agents, carbon dioxide snow and blood vessel ligation have been advocated [6]. But the treatment of choice at present remains surgical excision, the indications for surgery being symptomatic but stable tumours, sudden rapid acceleration of tumour growth, gross functional impairment, local skin

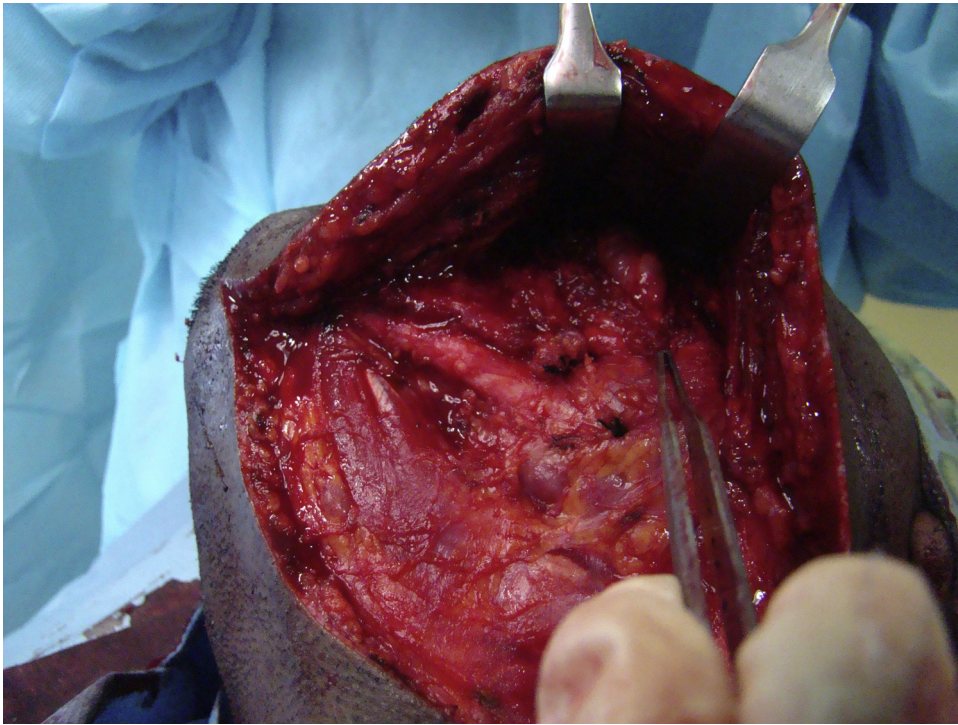


Fig. 3. Incision and Flap elevation.

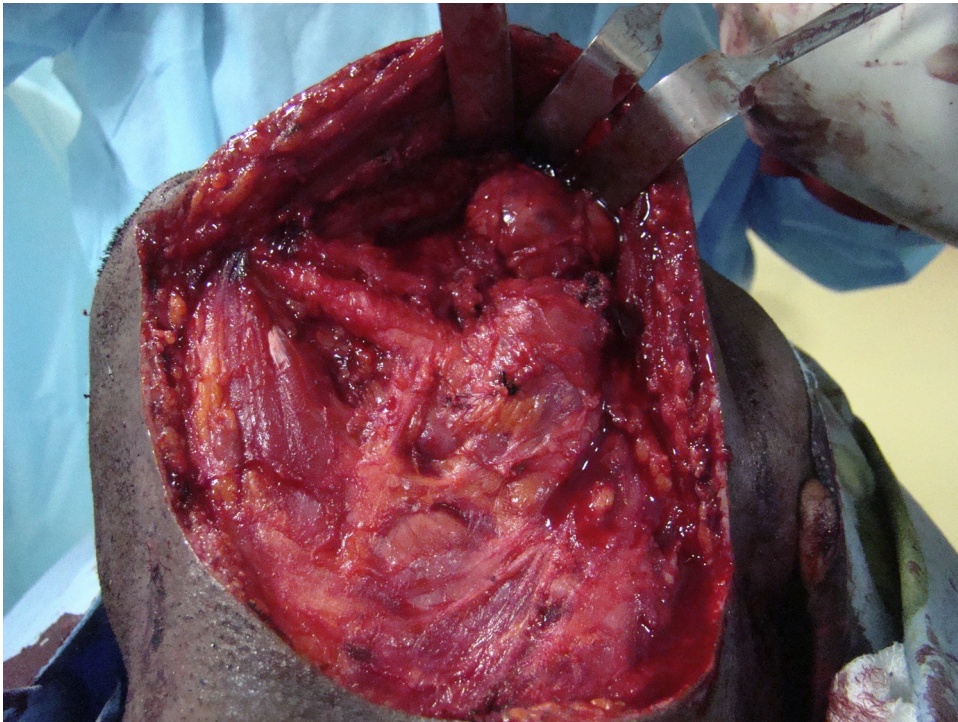


Fig. 4. Lesion exposed 1.

necrosis, thrombocytopenia, cosmetic deformity and suspicious of malignancy [14]. Difficulty in intraoperative localization of the exact extent of the tumour due to its supple nature and the absence of a definite capsule justifies a complete excision of the muscle. The

fibrosis following surgery may render re-exploration and excision in case of recurrence hazardous with more risk of damage to the facial nerve. Cosmetic and functional disabilities after excision have been minimal even after significant removal of surrounding normal

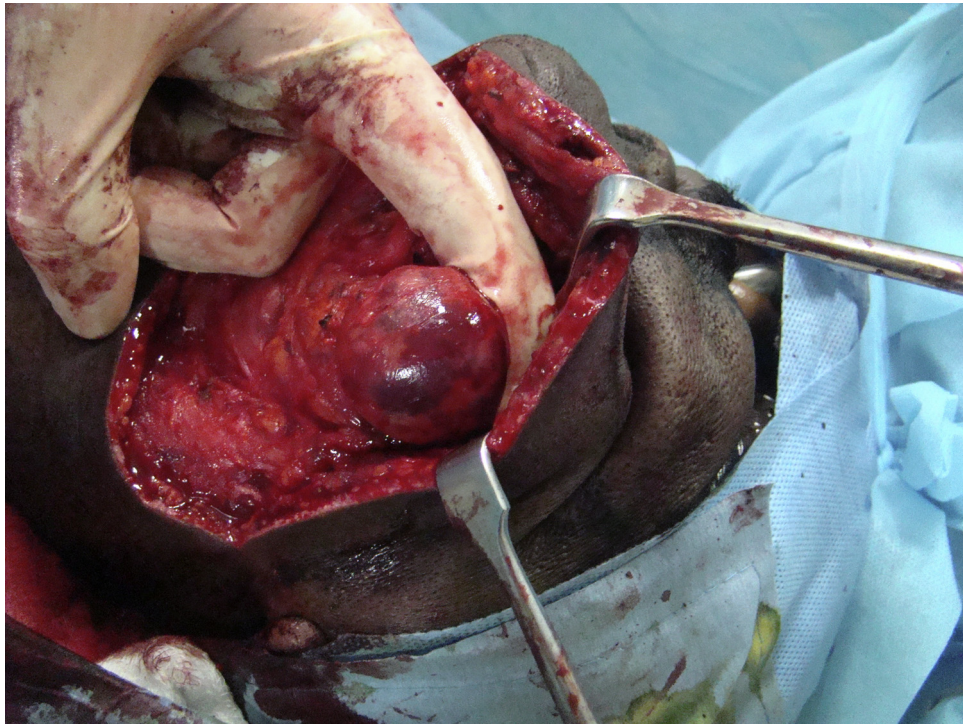


Fig. 5. Lesion exposed 2.

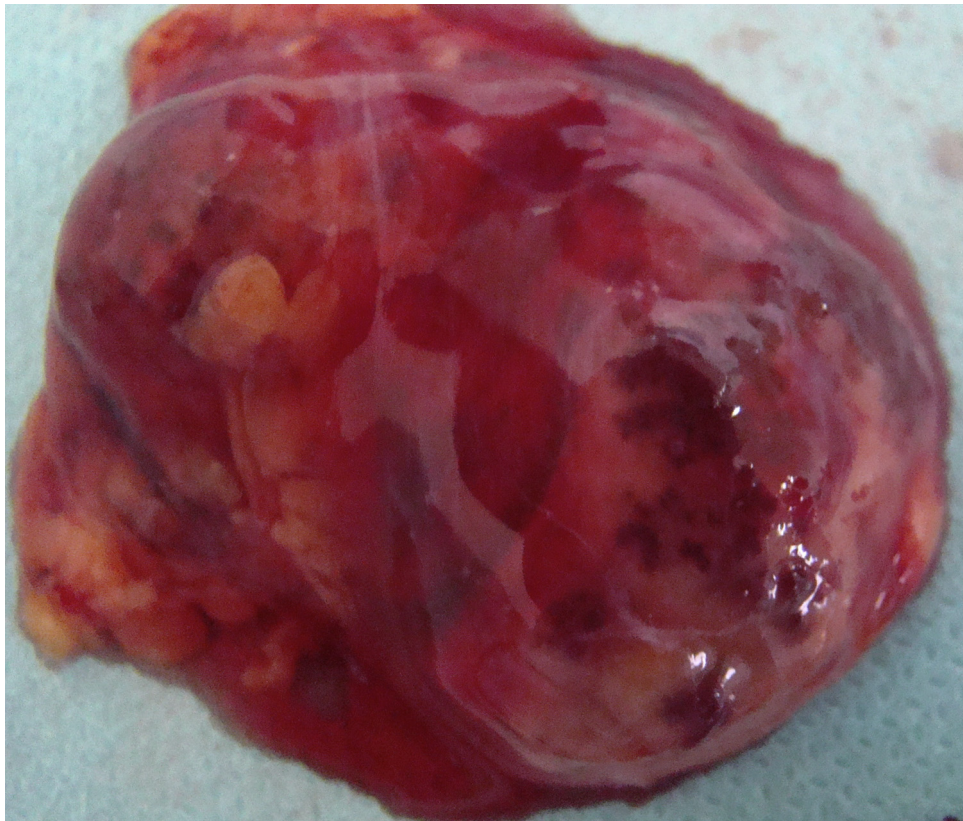


Fig. 6. Excised Lesion (see the glistening capsule).

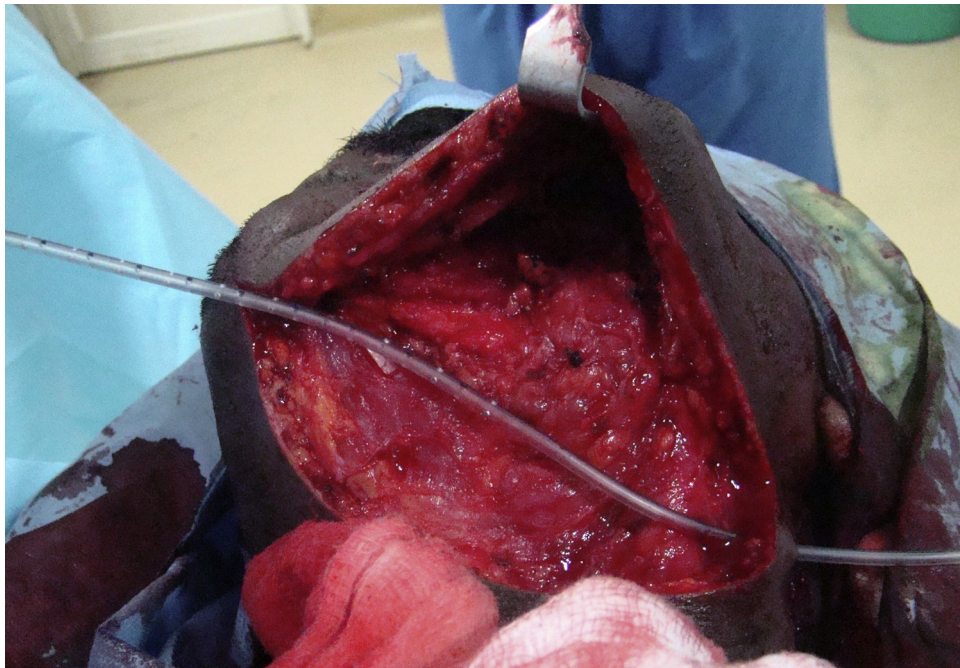


Fig. 7. Surgical defect and Closure.

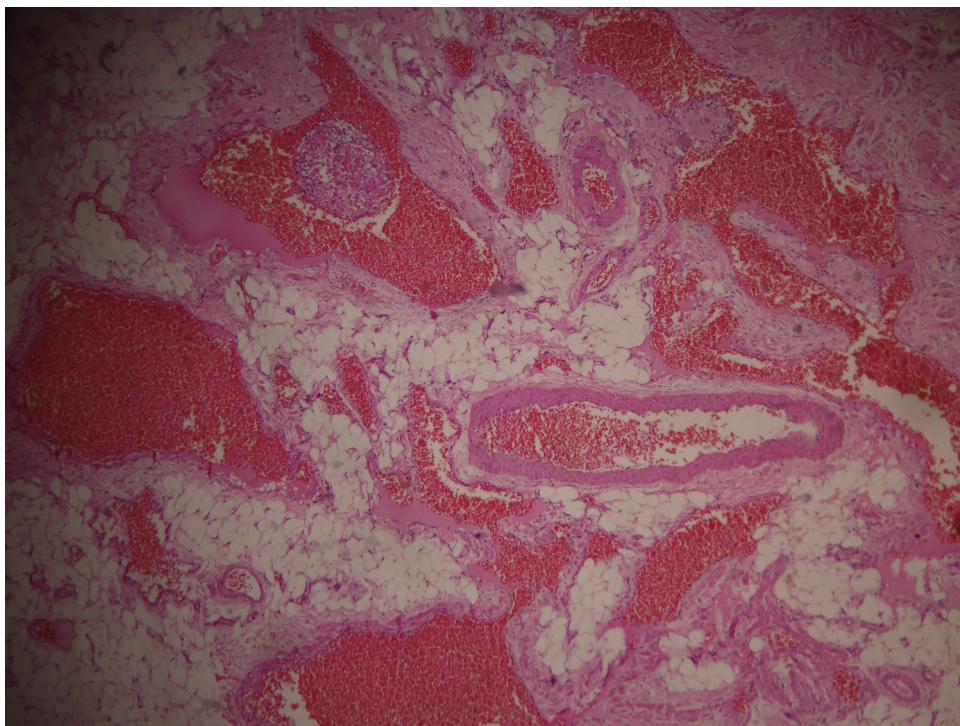


Fig. 8. Microscopic appearance.

muscle [14]. Local recurrences occur in approximately 18% due to incomplete surgical resection [6]. Spontaneous regression does not occur. Regional and distant metastasis has not been reported.

In conclusion, angiolipomas are rare in the head and neck region, and it should be considered in the differential diagnosis of masses in these regions. Its diagnosis is difficult because of its rarity and



Fig. 9. One-year post op.

nonspecific signs. The knowledge of the nature and recurrence rate of an IMH is useful for appropriate management.

Conflicts of interest

None declared.

Funding

None.

Ethical approval

This case report is the surgical treatment of an intramuscular hemangioma (IMH) in the masseter muscle. The treatment of intramuscular hemangioma (IMH) is surgical excision. The treatment plan was approved in the joint discussion by the maxillofacial surgeons and oral pathologists who are co-authors for this paper.

Consent

Obtained written and signed consent.

Author contributions

Dr. Surej Kumar L.K., was the surgeon, Dr nikhil kurian assisted the procedure Dr. Vinod mony was the Oral Pathologist. In addition, Dr Surej Kumar L.K. was also involved in data collection, Dr Surej Kumar L.K., Dr Kannan Venugopal and Dr Parvathi R Nair in data analysis and Dr. Kannan Venugopal and Dr Parvathi R Nair

for review of literature. Dr. Surej kumar L.K, Dr. Kannan Venugopal and Dr Parvathi R Nair was responsible for writing the article. Dr. Surej Kumar L.K. and Dr. Kannan Venugopal were responsible for the manuscript preparation.

Guarantor

Dr Surej Kumar L.K., Dr Kannan Venugopal.

References

- [1] J.E. Chipps, T.J. Weiler, Erectile cavernous hemangioma of the masseter muscle, *Oral Surg. Oral Med. Oral Pathol.* 3 (1950) 1509.
- [2] R. Liston, Case of erectile tumour in the popliteal space: removal, *Med. Chir. Trans.* 26 (1843) 120.
- [3] W.L. Watson, W.D. McCarthy, Blood and lymph vessel tumour, *Surg. Gynaecol. Obstet.* 71 (1940) 569.
- [4] D. Welsch, A.S. Hengerer, The diagnosis and treatment of intramuscular hemangiomas of the masseter muscle, *Am. J. Otolaryngol.* 1 (1980) 186–190.
- [5] G.K. Ingalls, G.J. Bonnington, A.L. Sisk, Intramuscular hemangioma of the mentalis muscle, *Oral Surg. Oral Med. Oral Pathol.* 60 (1985) 476–481.
- [6] G.T. Wolf, F. Daniel, C.J. Krause, et al., Intramuscular hemangioma of the head and neck, *Laryngoscope* 95 (1985) 210.
- [7] J.E.S. Scott, Hemangiomas in skeletal muscle, *Br. J. Surg.* 44 (1957) 496.
- [8] J.B. Mulliken, J. Glowacki, Haemangiomas and vascular malformations in infants and children: a classification based on endothelial characteristics, *Plast. Reconstr. Surg.* 69 (1982) 412–422.
- [9] P.W. Allen, F.M. Enzinger, Hemangioma of skeletal muscle. An analysis of 89 cases, *Cancer* 29 (1972) 8–22.
- [10] M.J. Chan, N.R. McLean, J.V. Scames, Intramuscular hemangioma of the orbicularis oris muscle, *Br. J. Oral Maxillofac. Surg.* 30 (1992) 192–194.
- [11] E. Ozer, D.E. Schuller, Angiolipoma of the neck, *Otolaryngol.—Head Neck Surg.* 135 (2006) 643–644.
- [12] D. Christopher, K. Unni, F. Mertens, Adipocytic tumors, in: *WHO Classification of Tumors. Pathology and Genetics: Tumors of Soft Tissue and Bone*, IARC, Lyon, France, 2002, pp. 19–46.

- [14] M.S. Kenali, P.G. Bridger, Intramuscular hemangioma of the medial pterygoid, ANZ J. Surg. 70 (2000) 462–466.
- [15] G.T. Terezhalmay, C.K. Riley, W.S. Moore, Intramuscular hemangiomas, Quintessence Int. 31 (2000) 142–143.
- [16] J.M. Hawnaur, R.W. Whitehouse, J.P. Jenkins, I. Isherwood, Musculoskeletal hemangiomas; comparison of MRI with CT, Skeletal Radiol. 19 (1990) 251–258.
- [17] J.G. Hoehn, G.M. Farrow, K.D. Devine, Invasive hemangioma of the head and neck, Am. J. Surg. 120 (1970) 495–498.

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