Intramuscular Hemangioma arising from Frontalis Muscle: An Unusual Presentation

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ABSTRACT

Introduction: Intra-muscular hemangiomas (IMH) are a type of benign vascular tumors which are less commonly seen and constitute a small percentage (< 1%) of all hemangiomas. Due to their infrequency, deep location, and unfamiliar presentation, these lesions are seldom correctly diagnosed clinically. They must be considered in the differential diagnosis of unexplained pain and swelling in muscles.

Case report: Here, we report a case of a 25 year old male presenting with a soft tissue mass in the left frontal region, confirmed as intramuscular hemangioma based on imaging studies and histopathologic examination, treated by surgical excision which had no recurrence after a 1-year follow up.

Conclusion: Intramuscular hemangioma involving the orofacial region is a rare presentation and surgical excision of the tumor as primary treatment modality yields good results.

Keywords: hemangioma, intramuscular hemangioma, vascular malformation.

INTRODUCTION

Vascular anomalies are congenital lesions of abnormal vascular development.1 There are two main categories of vascular anomalies; tumors and malformations.² Vascular tumors are endothelial neoplasms characterized by increased cellular proliferation and account for approximately 7% of all benign tumors, the majority of which develop in the head and neck region.^{2,3} Hemangiomas are the most common vascular tumors. They are rarely apparent at birth, grow rapidly during the first 6 months of life, involute with time and do not necessarily infiltrate but can sometimes be destructive.1 They are categorized into two types: "infantile" or "congenital" with the infantile type being the most common tumour in infancy and occurring in approximately 10 % of the population. They occur most commonly in subcutaneous adipose tissue but may also be found in muscle.^{4,5} In the head and neck region, they occur very rarely in oro-facial muscles (<1%).6

One special form of hemangioma is intramuscular hemangioma, which is a rare benign congenital neoplasm accounting for <1% of all haemangiomas, and <20% of those found in the head and neck area.⁷ It is different from infantile cutaneous hemangioma, in that, no spontaneous regression is seen with IMH and is usually detected in the second or third decade of life. Their location and unfamiliar presentation may require sonography, magnetic resonance imaging (MRI) and sometimes angiography for accurate diagnosis.^{8,9} The masseter muscle is the most frequently involved site in the head and neck area accounting for 5% of all intramuscular haemangioma.¹⁰ The other muscles which may be involved include the trapezius, periorbital, sternocleidomastoid, and temporalis.⁸ The tongue, extra ocular and posterior neck muscles have also been reported to be

involved with less frequency.¹¹ Multiple treatments are available for symptomatic intramuscular hemangiomas and options include conservative management, systemic corticosteroids, embolization, radiation, sclerotherapy, and surgical excision.⁴ Here, we report a rare case of intramuscular hemangioma of the frontalis muscle in forehead of a 25 year old male patient for whom surgical excision was carried out under GA.

CASE REPORT

A 25 year old male presented to the Department of Oral and Maxillofacial Surgery, Institute of Dental Studies and Technologies with a complaint of swelling in the forehead region above the left eye for the past 8 years and a second swelling on the right side of the face since birth (Figure-1). Patient gave history of trauma 8 years ago with subsequent gradual increase in size of the swelling. Patients father had a similar lesion on right side of the face which was not treated. Medical history was unremarkable. Clinical examination revealed a well circumscribed swelling about 4 x 6 cm in dimension, extending medially till midline and laterally upto lateral 2/3 of the left eyebrow. The overlying skin was normal and the surface smooth. Palpation elicited a soft fluctuant swelling which was non - pulsatile, non-mobile and non- tender with normal skin texture, and presence of step on lateral 1/3rd of supraorbital rim. The second lesion extended medially till angle of the mouth and laterally upto middle of the zygomatic arch and on palpation was soft fluctuant with an irregular surface. A clinical diagnosis of vascular malformation and a differential diagnosis of lipoma, hemangioma and dermoid cyst were considered and the necessary diagnostic workup was done.

Routine hematological investigations were within normal limits. PNS view showed a well- defined radiolucency approximately 3x5 cm in dimension in left frontal region (Figure-2). MRI showed a homogenously enhancing lesion in left frontal and periorbital region with scalloping and thinning of underlying frontal bone and getting its blood supply from angular vessels and frontal scalp vessels (Figure-3). No obvious intracranial or intraorbital extension was noted. Based on the diagnosis, surgical excision of the lesion was planned under general anaesthesia. Prior to the incision, circumferential sutures were

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placed all around the base of the lesion inorder to block the feeder vessels supplying the lesion (Figure-4). An elliptical incision was given on the surface of the lesion followed by blunt dissection (Figure-5). Supraperiosteal dissection was done and the entire mass was completely excised from the base with a margin of normal surrounding muscle to prevent recurrence. Flap approximation was done and the wound was sutured close following placement of a drain to prevent formation of dead space (Figure-6). Postsurgically antibiotics and analgesics were prescribed for a week. There was mild postoperative facial edema, which subsided within two weeks with no evidence of pain and significant cosmetic problem. The excised specimen (Figure-7) was sent for histological examination which revealed numerous small, medium sized and few large thick walled blood vessels in a cellular connective tissue stroma showing numerous fibroblasts, abundant muscle fibres and nerve bundles. RBC pooling within the vessel lumen as well as extravasation was noticed in areas and the features were suggestive of intramuscular hemangioma. The patient was followed up for a year and no new symptoms or recurrence of the lesion was seen.

DISCUSSION

Hemangiomas are benign proliferative vascular lesions characterized by increased endothelial turn over.^{8,10,12} They are usually seen at birth, undergo rapid growth followed by progressive involution over a period of years. Intra muscular hemangioma is a rare variant comprising < 1% of all hemangiomas, and < 20% of those involving the head and neck. In the maxillofacial region, the masseter muscle is most frequently involved, accounting for 5% of all intramuscular hemangioma followed by trapezius, periorbital, sternocleidomastoid, and temporalis.

When examined histopathologically, the hemangiomas can be broadly categorized as capillary and cavernous. Allen and Enzinger in 1972 formed a classification system based on the vessel size; capillary, cavernous or mixed small/large vessel types. This classification correlates well with location and prognosis.¹³

In 1843, Liston was the first to report a case of intramuscular cavernous hemangioma naming it as an "erectile tumor." In most cases, the tumor is seen to develop before the age of 30 years; thus requiring consideration of either trauma or congenital origin as a possible etiologic factor.⁸ However the exact cause of intramuscular hemangioma is largely unknown. They generally have no gender predispositions. In the present case the patients history of trauma could be a contributing predisposing factor to the development of the lesion.

On clinical examination, the predominant complaint is presence of a slowly enlarging mass.¹⁵ Intramuscular hemangiomas rarely display any clinical symptoms or signs that reveal their vascular nature.^{12,15} Due to their fibro vascular nature they have a rubbery, firm texture and vascular bruits, thrills or pulsations are usually absent and pain may or may not be present. The lesion is non- fluctuant and largely immobile, although slight side to side movement may be possible opposite to the orientation of the muscle fibres. The overlying skin appears normal and discoloration present in other forms of hemangiomas is rarely seen. These features were consistent with the present case.

Most hemangiomas can be diagnosed on clinical examination



Figure-1: 25 year old male with swelling in left frontal region and on right side of the face; **Figure-2:** PNS showing well defined radiolucency in left frontal region.



Figure-3: MRI showing homogenous enhancing lesion in left frontal and periorbital region; **Figure-4:** Circumferential suturing followed by elliptical incision



Figure-5: Dissection of the lesion; Figure-6: Closure of wound with placement of drain.



Figure-7: Excised tissue specimen.

and do not require any investigation or any treatment as they tend to subside spontaneously. However, imaging is needed in cases of deep seated hemangioma with normal overlying skin or in cases of clinically atypical soft-tissue masses.¹⁶ When imaging is used, it is important to choose the modality based on the specific lesion and clinical situation. Conventional radiographs help in identifying phlebolith and calcifications, but they may not be specific. Ultrasonography and magnetic resonance imaging (MRI) are more accurate in the diagnosis of hemangiomas.

MRI has been shown to provide better detection and delineation of the extent of IMH than Computed tomography.8 MRI is superior because of its multiplanar capabilities and the distinct contrast between normal muscle and the IMH. The MRI findings of an intramuscular hemangioma consist of an intermediate signal on T1 weighted images and an intense signal on T2 weighted images, but it should be noted that not all intramuscular hemangiomas will give a high intensity signal on T2 weighted MRI.^{10,16} If pulsations, bruits, or thrills are evident on clinical examination, arteriography is indicated to identify large vessel communications.¹⁶ Colour doppler sonography is exclusively useful to demonstrate the vascular structures in and around the muscle and to evaluate the pathological changes like fibrosis and to detect calcifications. The presence of a color Doppler signal in a well-defined hypoechoic mass with heterogeneous echo texture should raise the possibility of hemangioma. Fine-needle aspiration cytology has been found to be of poor diagnostic value in most cases. In the present case, MRI was used as the primary diagnostic modality to detect the extent of the lesion.

Management of intramuscular haemangioma should be individualized according to its size, growth rate, anatomic accessibility of the tumor, age of the patient and cosmetic and functional considerations.12 Numerous approaches to the management of the IMH have been advocated such as steroids, sclerosing agents, radiation, cryotherapy, lasers, embolization, and ligation of the feeding vessel have all been used.^{8,18} The first line of medical treatment is corticosteroid, either topical, intralesional, or systemic. If the lesion does not respond to corticosteroid, second-line pharmacologic agents include vincristine or interferon alfa 2b. Sclerotherapy is done if the lesion is too large or too close to some important, structures or organs or when there is a need to debulk the tumor before surgery or cosmetic treatment.8 Recently, there have been reports of preoperative embolization of hemangiomas with muscle fragments as a technique to decrease intraoperative blood loss. However because of the destructive effect on normal tissues and lower rate of complete regression of the tumor, these non -surgical modalities are not as effective as surgical resection. Total wide excision of the tumour along with a small cuff of normal tissue is currently the most accepted treatment modality. Chen et al¹⁷ reported a case of intramuscular angioma in the frontalis muscle of the forehead in which excisional biopsy of the lesion was done. Lakshmi et al¹⁶ reported a case of intramuscular hemangioma in right side massectric region which was treated with surgical excision. Similarly in our case too, surgical excision of the lesion was used as an effective treatment modality. Local recurrences occur in approximately 18% of IMH, usually as a result of incomplete surgical resection. Regional and distant metastasis has not been reported.¹³ In the present case no recurrence was seen at one year follow-up.

CONCLUSION

Intramuscular hemangioma should be considered in the differential diagnosis whenever an isolated muscle enlargement is encountered. An early and accurate diagnosis of the lesion and monitoring the vascularity with MRI angiography is of utmost importance to facilitate proper treatment. Complete resection minimizes the relapse rate of the tumor and results in favorable cosmetic outcomes offering a better life quality for the patient.

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