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Case Report

Intramuscular cavernous haemangioma of the triceps brachii: a rare tumour with good prognosis

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ABSTRACT

Intramuscular cavernous hemangiomas are a sub-type of deep soft tissue hemangiomas most commonly presenting in the lower extremities. This report highlights the rare occurrence of these tumours in the upper extremity (triceps muscle) in which only a handful of cases have been reported in literature. The tumour was treated pre-operatively with embolization and sclerotherapy followed by complete excision of the lesion along with the lateral head of triceps. Following complete removal, the patient has been symptom-free for almost one year with no recurrence reported to date. In conclusion, early diagnosis and timely intervention can help prevent complications like neurovascular compromise, minimise recurrence and lead to excellent functional outcomes with minimal morbidity as exemplified in this case.

Keywords: Intramuscular cavernous hemangioma, Triceps, Surgical excision, Embolization, Sclerotherapy

INTRODUCTION

Hemangiomas are an abnormal proliferation of blood vessels most commonly presenting as benign soft tissue tumours of the skin and subcutaneous tissue, very rarely they may present as intramuscular swelling and even more rarely as bony outgrowths.[1-3] We present the case of a young female who presented with complaints of elbow pain for many years due to a growth in the posterior aspect of the arm.

A search in the literature of the word's intramuscular hemangioma in the triceps revealed only a handful of cases highlighting the rarity of these tumours in the upper extremity.[1-5] Intramuscular hemangiomas are the most common type of deep hemangiomas and constitute 0.8% of all hemangiomas with the most frequent site being the lower extremity.[1-6] These tumours may present as diagnostic dilemmas due to their insidious onset, chronicity and vague symptoms, but they may be diagnosed through a combination of a thorough history and clinical examination, radiological investigations including X-ray, Ultrasound/Doppler imaging, MRI and biopsy for histopathological and morphological diagnosis.[1-3] These tumours do not metastasize or undergo malignant transformation despite their origin from vascular endothelial cells and generally have a good prognosis if excised completely with minimal risk of recurrence.[1,2,4,6,7,10]

CASE REPORT

A 23-year old female presented with complains of left elbow pain and difficulty in extension since 2 to 3 years. She had a history of venous malformation of the affected left upper extremity since birth and underwent excision of superficial dilated veins and forearm seven years ago, but they recurred after a few months. On examination she had

dilated engorged veins over the left dorsum hand, wrist, dorsum of forearm, dilated venous plexus over the posterolateral aspect of left elbow and mass palpable on the posterior aspect of arm deep to the triceps muscle which was slightly tender on palpation and with restriction in elbow flexion and extension (Figure 1). On pre-operative imaging, X-ray revealed soft tissue calcifications with phlebolith (Figure 2), MRI (with angiography -MRA) and Doppler ultrasound confirmed a sizeable intramuscular hemangioma (more than 16 cm in size) in the posterior compartment of the arm within the triceps muscle (Figure 3). So, it was decided to treat her with injection sclerotherapy and embolization of the venous malformations and tumour mass before surgical excision to debulk the tumour.

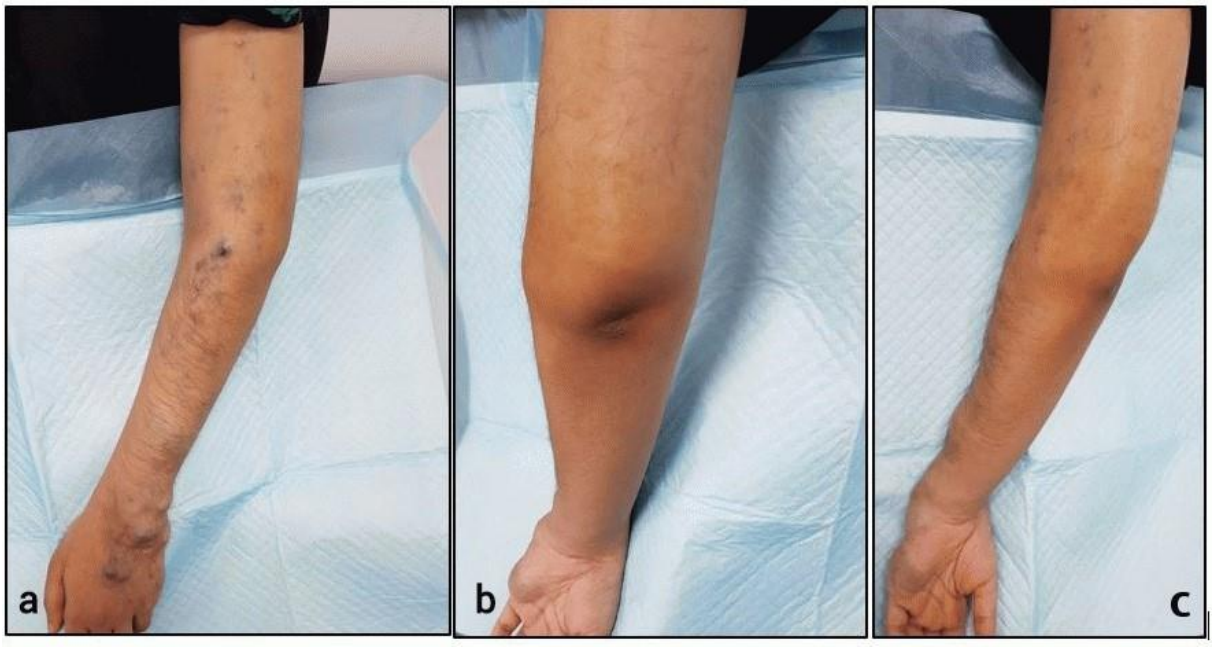

Figure 1: (a) Pre-operative image showing multiple dilated tortuous veins over the left hand and dorsum of forearm extending upto elbow; (b and c) dorsal and lateral view of left upper extremity showing growth involving posterolateral aspect left arm.

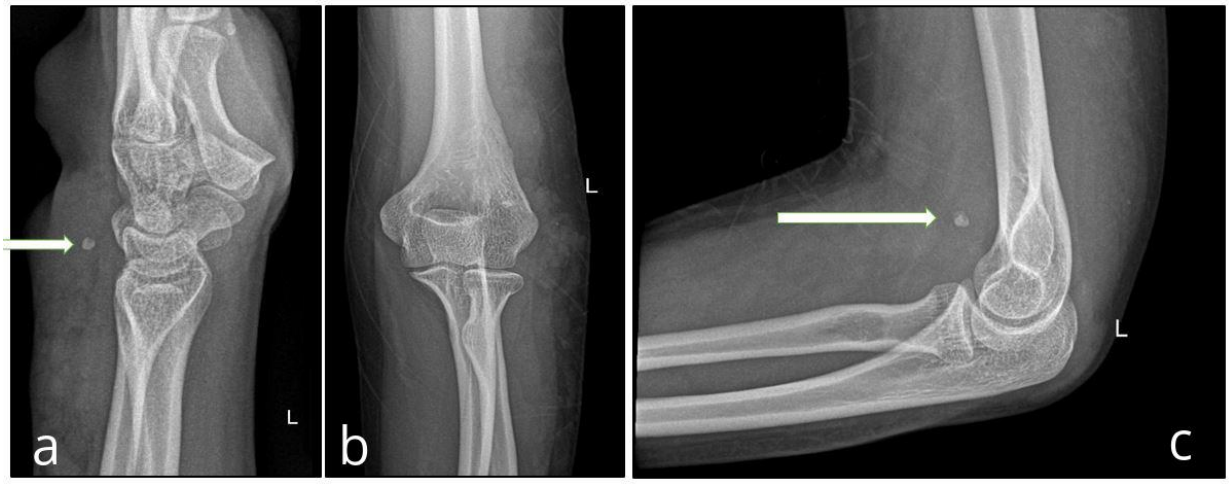

Figure 2 (a-c): Roentgenogram of wrist and elbow showing soft tissue calcifications and phlebolith (white arrow).

During surgical excision, the tumour was excised entirely along with most of the lateral head of triceps (Figure 4 and 5) and specimen sent for histopathological examination which was subsequently reported as a cavernous intramuscular hemangioma. The postoperative course was uneventful, and she refused subsequent intralesional sclerosant injection therapy and surgery for dilated superficial veins of the forearm and hand. She came for regular follow up and 11 months after surgery, she had an almost full range of motion for elbow flexion and extension, with resolution of pain and QuickDASH (disabilities of the arm, shoulder and hand) score of 23% (Figure 6).

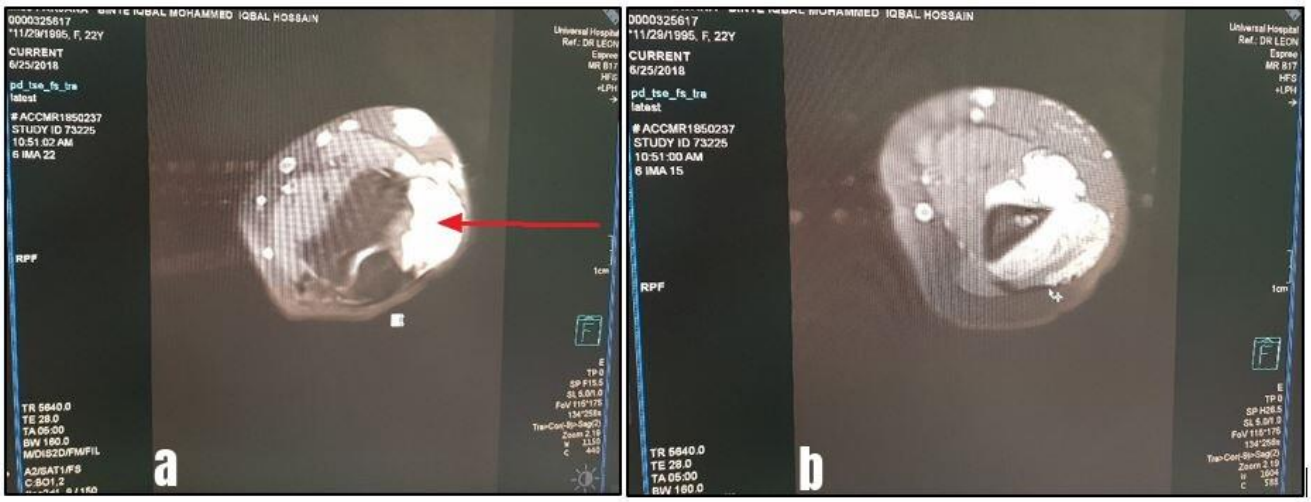

Figure 3 (a and b): MRI showing a large intramuscular hemangioma (red arrow) in the posterior compartment of the distal half of the arm within the triceps muscle.

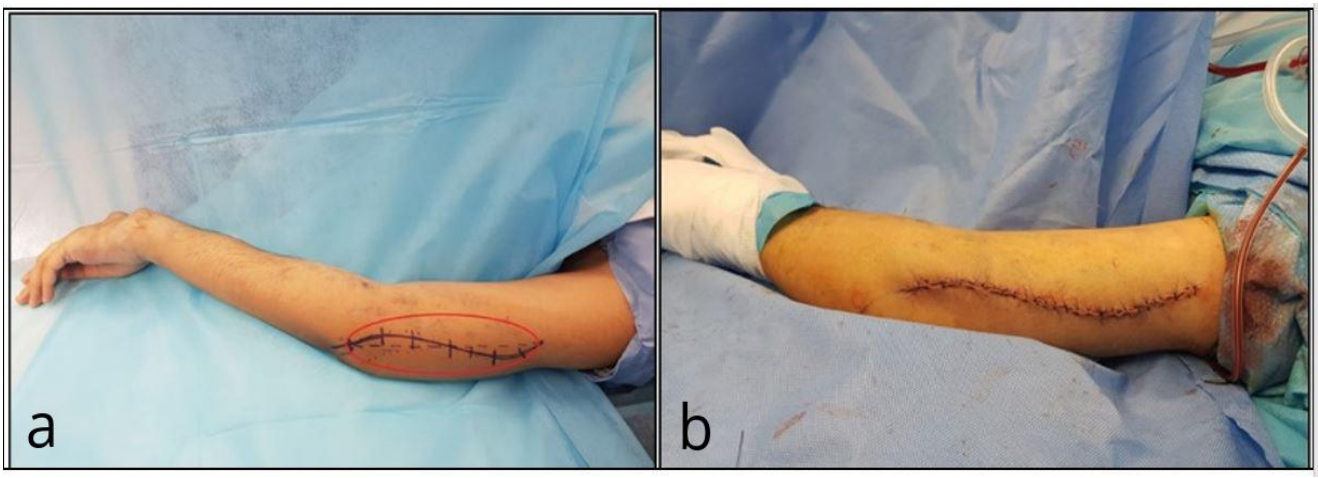

Figure 4: (a) Pre-operative image showing skin incision marking and outline of the tumour (in red); (b) postoperative image showing skin closure with a drain in situ.

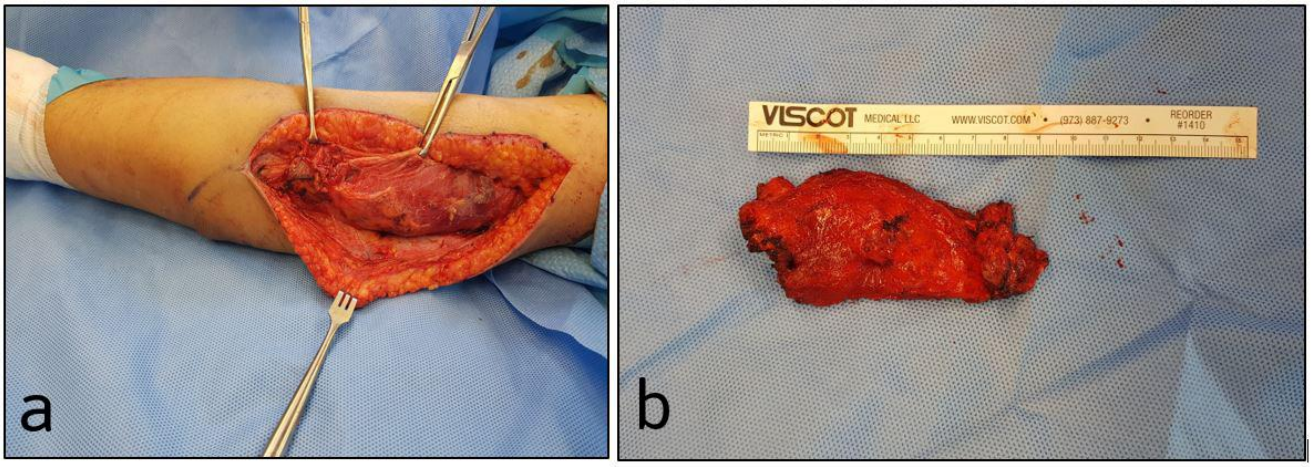

Figure 5: (a) Intra-operative image showing hemangioma involving almost entire lateral head of triceps; (b) image showing specimen excised in toto.

Figure 6 (a and b): Final post-operative outcome at 11 months.

DISCUSSION

This case has been reported because of the rarity of presentation of intramuscular cavernous hemangiomas in the upper limb, as the most common site of such tumours is in the lower extremity (Quadriceps femoris being the commonest site - 36%).[1] These tumours typically occur in infancy or childhood and undergo a period of involution in the adolescent years, but may also occur at any age with causative factors being - congenital or traumatic origin.[1,2,5-7]

The clinical presentation of intramuscular hemangiomas may vary from swelling, pain with limitation of joint mobility and in some cases may even present with symptoms of nerve compression (Pulidori et al, Khan et al) and flexion contracture (Niempoog et al).[4,8,9] In every case, a thorough workup is mandatory and must include conventional radiography, ultrasound with Doppler imaging, MRI with/without angiography and post excision a histopathological analysis should be done. Most of these benign tumours never metastasize; however, there can be recurrence after excision.[10,11]

On X-ray imaging, these tumours will show soft tissue masses with calcifications, phleboliths and sometimes periosteal reaction and bony erosions, which can mimic osteomyelitis or bone tumour. Doppler ultrasound is important to characterise these lesions, whether they are cystic or solid in nature and to know their vascularity, flow patterns, and on ultrasound, these lesions typically present as hyper-echogenicities.

MRI with or without angiography may be the most important pre-operative tool to map these lesions, T1-weighted images appears as a low-to-intermediate signal intensity mass with peripheral high signal intensity due to fat overgrowth. T2-weighted images show areas of high signal intensity due to vascular tissue and intermediate signal intensity due to fat.[2] Other differential diagnoses of intramuscular hemangiomata like arterio-venous vascular malformations and malignant intramuscular tumours such as sarcomas which can lead to diagnostic dilemmas can be reliably distinguished by MRI which is now considered to be the gold standard.[1,7,9,12]

Additionally, MRI can also help in distinguishing these vascular tumours, cavernous hemangiomas can be seen as tumours with large cystic spaces, arteriovenous hemangiomas will show fast flow serpentine vessels, and venous hemangiomas will reveal slow flow serpentine vessels. MRI angiography can be considered if there are symptoms of neurovascular compression.[2]

Treatment options for intramuscular hemangiomas may range from watchful expectancy as some of these tumours may involute with time or spontaneously regress, and for the more aggressive lesion, surgical excision is warranted. The indications for surgery are severe, unremitting pain, cosmetic deformities, distal neurovascular compression, local pressure effects leading skin necrosis, bony/muscular pain, restricted mobility and suspicion of malignancy.[7,12]

Hence the treatment of choice for these tumours is surgery.[1,12] If the tumour is excised completely then chances of recurrence are minimal, sometimes complete excision may entail sacrifice of critical encased structures such as nerves, blood vessels or as in this case the lateral head of triceps had to be sacrificed, hence proper pre-operative documentation, imaging work-up and patient consent before surgery is vital to avoid litigation.

Complications following surgical excision include hemorrhage, delayed wound healing, wound infection, scarring, neurovascular injury leading to ischemic or neurological sequelae and recurrence. If recurrence following surgical excision occurs, then adjuvant therapy with interferon- alpha may be considered. In case of lesions not amenable to surgery, options include therapeutic embolization by an interventional radiologist or radiotherapy.[2]

For complex, high risk and infiltrative lesions, treatment modalities include - sclerotherapy, embolization followed by surgery, embolization alone, radiotherapy and corticosteroids.[11-14]

Embolization is an effective treatment strategy, mainly used before surgery to decrease the vascularity of the tumour, hence decreasing blood loss and it can also be used to control pain and decrease the chances of recurrence following excision. However, for embolization to be effective, it must be combined with surgery, and the tumour is usually embolized 2-3 days before surgery.[1,11-14]

Other treatment options reported but not used frequently, include radium therapy, topical agents (carbon dioxide snow), cryosurgery, and arterial ligation, but these are fraught with uncertainties and recurrence is common following this modalities.[1]

CONCLUSION

Intramuscular cavernous hemangiomas of the upper limb are rare tumours and this possibility must always be kept in mind when faced with chronic, insidious onset tumours presenting with symptoms of joint/muscular pain and limitation in mobility. Their diagnostic workup includes conventional radiography, Doppler ultrasound and MRI. The treatment approach must be individualised depending on patient presentation, but the general consensus is that complete surgical excision gives good results with minimal recurrence.

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