Case Report

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Intramuscular Hemangioma of the Forearm with Phleboliths: A Case Report

Pathology Section

MUNISWAMY REDDY ARCHANA[1], ARCHANA SHIVAMURTHY[2], SUNIL SHIVANNA[3]

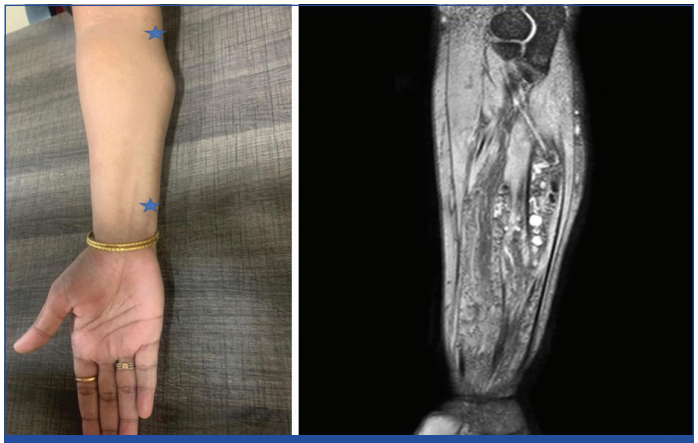
ABSTRACT

Intramuscular hemangiomas of the forearm are rare. Patients typically present with pain, discomfort, and progressive enlargement of the lesion. Diagnosis is often difficult due to their infrequency, deep location, and unfamiliar presentation. In this case report, we present a 47-year-old woman with an intramuscular hemangioma with phleboliths involving the forearm. The patient reported swelling in the right forearm for the past 20 years, which was insidious in onset, gradually progressive in size, and associated with dull aching pain. Upon examination, a soft 20×4 cm swelling was present over the right forearm involving the ulnar aspect, with a positive Tinel's sign at mid-forearm level. Clinical differential diagnosis included lipomatosis, neurofibroma, and intramuscular hemangioma. On Magnetic Resonance Imaging (MRI), a heterogeneously enhancing serpentine lesion was noted involving the flexor compartment muscles of the right forearm. The patient underwent excision of the lesion, and histopathological examination confirmed intramuscular hemangioma. During the postoperative recovery period, all range of movements at the elbow joint and wrist were normal. No recurrence was noted up to two years of follow-up

Keywords: Blood vessels, Swelling, Upper limb

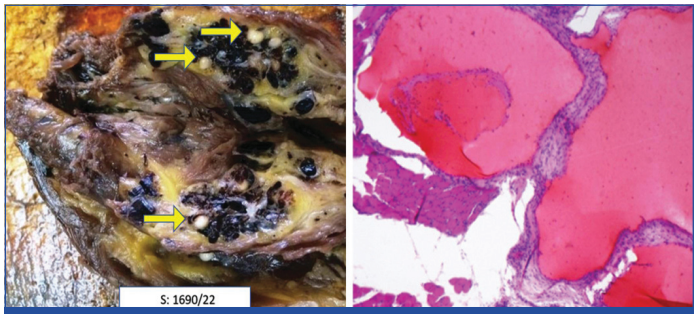
CASE REPORT

A 47-year-old female presented with complaints of swelling in the right forearm for the past 20 years. The swelling was insidious in onset, gradually increasing in size, and associated with dull aching pain that worsened upon holding any object and improved with rest. The patient had no significant medical history. On examination, a soft 20×4 cm swelling was present over the ulnar aspect of the right forearm, with pinchable overlying skin see [Table/Fig-1]. The swelling reduced in size upon muscle contraction, and Tinel's sign was positive at mid-forearm level. A differential diagnosis of lipomatosis, neurofibroma, and intramuscular hemangioma was considered upon complete clinical examination. Routine haematological investigations were within normal limits. MRI revealed a heterogeneously enhancing serpentine lesion in the flexor compartment of the right forearm, insinuating between flexor digitorum superficialis and flexor pollicis longus see [Table/Fig-2]. The lesion encased the median nerve, and phleboliths were present. These findings on MRI supported the diagnosis of intramuscular hemangioma. The patient underwent surgical excision of the lesion with a provisional diagnosis of intramuscular hemangioma, and the specimen was sent for histopathological examination.

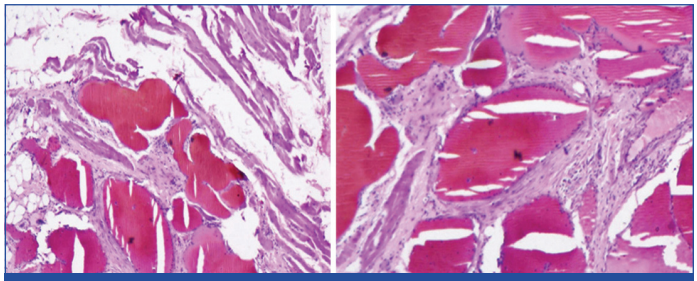


[Table/Fig-1]: Picture shows a swelling on the ulnar side of the right forearm.
[Table/Fig-2]: MRI showing intramuscular hemangioma involving the flexor compartment of the forearm with phleboliths. (Images from left to right)

On gross examination, the excised specimen measured 16×6.5×1.5 cm. The cut section showed bundles of skeletal muscle fibers with areas of haemorrhage and cystic spaces. Phleboliths were also observed see [Table/Fig-3]. Microscopically, the lesion was characterised by skeletal muscle fibers with intervening lobules composed of numerous thick and thin-walled, variably sized dilated and congested vessels lined by flattened endothelial cells see [Table/Fig-4-6]. These features confirmed the diagnosis of intramuscular hemangioma. The patient recovered well after surgery, with normal range of motion at the elbow joint and wrist. No recurrence was observed during the two-year follow-up period.



[Table/Fig-3]: Gross photograph showing the cut section with haemorrhagic areas and phleboliths (yellow arrows).
[Table/Fig-4]: Microphotograph showing large dilated vascular channels with intervening skeletal muscle fibers (Haematoxylin and eosin stain x40). (Images from left to right)



[Table/Fig-5]: Microphotograph showing large dilated vascular channels with intervening skeletal muscle fibers (Haematoxylin and eosin stain x40).
[Table/Fig-6]: Microphotograph showing large cavernous vascular channels with intervening skeletal muscle fibers (Haematoxylin and eosin stain x200). (Images from left to right)

DISCUSSION

In 1843, Sir Liston reported the first case of intramuscular hemangioma. Clinically, these hemangiomas present as a swelling associated with pain [1,2]. The young age of affected patients and the long duration of symptoms in some cases suggest that many of these lesions are congenital tumors that slowly give rise to symptoms during late childhood or early adulthood. Growth spurts or trauma may accelerate their growth, but they can also regress spontaneously. Intramuscular hemangiomas are most commonly located in the lower extremity, with the thigh being the most affected area, followed by the calf muscle. The quadriceps muscle is the most frequently affected muscle. Although these hemangiomas frequently enlarge, they never metastasize [3,4]. Malignant transformation is rare, and the recurrence rate after surgical excision is approximately 9% [5,6]. Some authors suggest that both sexes are equally affected, while others indicate that the incidence is higher in females [7-9]. A review of the literature listing the various reported cases of intramuscular hemangiomas involving the forearm is included in [Table/Fig-7].

Intramuscular hemangiomas are typically diagnosed based on clinic-radiological correlation. X-ray and MRI are the initial investigations of choice. Deep-seated lesions presenting as atypical soft tissue masses can lead to an error in clinical diagnosis. Phleboliths and calcifications are non-specific findings on conventional radiography. Color doppler sonography can detect the vascular structures in and around the muscle and pathological changes such as fibrosis and calcifications [10,11]. Hemangiomas can be distinguished from other soft tissue lesions by their abundant vascularity and high blood flow velocity. MRI provides the best diagnostic information in cases of deeply situated and large intramuscular hemangiomas. The

Case reports	Author, Year, Place	Patients details							Follow-up
		Age	Sex	Location/Muscles involved	Clinical features	Duration of chief complaints	Size of the swelling	Treatment	
1	Sunil TM et al., [1] 2003 Bangalore, India	12	F	Flexor Digitorum Superficialis (FDS)	Painful swelling in the left forearm, h/o trauma; Volkmann's Like Contracture of the Forearm	6 y	Not mentioned	Excision with intensive physiotherapy	Full functional restoration was achieved about 8 months after surgery
2	Nazzi V et al., [2] 2008 Milan, Italy	32	F	Pronator and brachioradialis muscles	Mass Burning pain and paresthesia around the area innervated by radial nerve	2 y	40×36×12.8 mm	Embolisation Surgical excision	2 follow-up examinations at 1 and 3 months postoperatively-No recurrence/surgery related complications
3	Fnini S et al., [3] 7 cases 2013 Casablanca, Morocco	16-39 yrs	5 F 2 M	Flexor carpi radialis (2 cases) Flexor carpi ulnaris (4 cases) FCR+FDS (Flexor digitorum superficialis; 1 case)	Mass Pain Carpal canal syndrome (1 case) Ulnar nerve paraluysis (1 case)	Average consultation time- 13 months	Not mentioned	5 cases-complete excision 2 cases-incomplete excison (due to involvement of ulnar nerve)	4 years average follow-up: Excellent hand movements and no recurrence in 5 patients. 1 patient had persistence of ulnar nerve palsy. 1 patient had postoperative bleeding with recurrence
4	Eyesan SU et al., [4] 2015 Ogbomoso, Oyo State, Nigeria	18	M	Within the common belly of the long flexor group of forearm muscles with the short flexor tendon embedded within it	Pain and swelling of left forearm, clawing of fingers	9 y	7×13 cm	Excision biopsy with ligation of the feeding vessels	Regular follow-up with no recurrence and regular physiotherapy sessions to improve the function of the digits
5	Jeong, E et al., [5] 2015 Seoul Korea	19	F	Pronator Quadratus Muscle	Mass	8 y	5.5×2.4×1 cm	Excision	No evidence of recurrence at 6-month follow-up and all functional limitation of the left forearm and the hand were resolved
6	Hui L et al., [6] 2016 Zhejiang Province, China	16	M	Multiple lesions in the fore arm	Mass	15.7 y	14×12 cm	Sclerotherapy Excision Amputation	After the amputation surgery, the patient gained a functional recovery and the tumour did not recur during the 2 years after the surgery
7	Kamath et al., [7] 2017 Mangalore, India	14	F	Flexor Digitorum Superficialis (FDS) of the ring finger	Painless swelling on left forearm, flexion deformity of ring finger	1 y	Not mentioned	Excision and end to side anastomosis of the FDS to the middle finger	Complete correction of deformity was observed with full extension and flexion post operatively
8	Niempoog S et al., [8] 2019 Thammasat University Thailand	25	M	Flexor Digitorum Profundus (FDP) and Flexor Pollicis Longus (FPL)	Painful flexion contracture of right hand	4 y	2×2 cm	Surgical excision	No recurrence for 1 year follow-up
9	Mishra PK et al., [9] 2019 Madhya Pradesh, India	32	F	Flexor group of muscles	Multiple swellings over left forearm and elbow, diffuse, mild pain and substantial discomfort	5 y	Diffuse swelling	Conservative management with excision biopsy of the most painful area	After 1 year of follow-up, the swelling reduced and pain disappeared
10	Present case 2023 Mysore India	47	F	Flexor digitorum superficialis and flexor pollicis longus	Gradually progressive swelling associated with pain	20 y	20×4 cm	Excision	No recurrence upto 2 years of follow-up

[Table/Fig-7]: Review of literature: Various reported cases of intramuscular hemangiomas involving the forearm reported till date [1,2,4-9].

gross and microscopic appearance of intramuscular hemangiomas is variable depending on whether they are of capillary, cavernous, or mixed type. The capillary type is more common and has a spongy yellow-red appearance, composed of lobules of variable-sized small channels lined by endothelial cells. The cavernous type appears as blue-red masses composed of dilated, thin-walled, large vessels lined by flattened endothelial cells [1,8,12].

Various treatment modalities are available for symptomatic intramuscular hemangiomas, including conservative management, systemic corticosteroids, embolisation, radiation, sclerotherapy, and surgical excision [5,9,12]. The use of each modality depends on the individual characteristics and degree of functional impairment in each case. All management strategies should include regular follow-up, although true intramuscular hemangiomas have no malignant potential [8,10]. The patient in this case was advised active and active-assisted mobilisation. Localisation, circumscription and minimal loculations are the characteristics that make intramuscular hemangiomas good candidates for surgical excision. Owing to the complex and infiltrative nature of hemangiomas, the surrounding normal tissue must be also removed well beyond the gross edges of the lesion to prevent recurrence [7,11,12]. The greatest risk factor for recurrence is incomplete surgical excision. The risk of tumour recurrence after excision is variable, ranging from 18% to 61%. Haemorrhage remains the most common complication. Sclerotherapy has a role in the management of intramuscular hemangioma when excision is not possible.

CONCLUSION(S)

Intramuscular hemangioma should be included in the differential diagnosis especially in a young adult with unexplained deep seated intramuscular swelling. Clinical diagnosis may be challenging owing to the absence of features characteristic of vascular tumours like pulsation, thrill or bruit. It must be investigated with MRI which is the investigation of choice and can be cured with surgical excision in majority of cases.

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PARTICULARS OF CONTRIBUTORS:
1. Senior Resident, Department of Pathology, JSS Medical College, JSS Aher, Mysore, Karnataka, India.
2. Senior Resident, Department of Pathology, JSS Medical College, JSS Aher, Mysore, Karnataka, India.
3. Assistant Professor, Department of Orthopaedics, Chikkaballapur Institute of Medical Sciences, Chikkaballapur, Karnataka, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Archana Shivamurthy,
No. 201, D S Max Smaran Apartment, Vishveshwarnagar,
Bhavani Swamy School Road, Mysore-570008, Karnataka, India.
E-mail: archanashivamurthy@gmail.com

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