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CASE REPORT

Pronator Quadratus Hemangioma (PQH): A Rare Case Report and Review

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Abstract
The intramuscular hemangioma is a rare clinical entity in the upper extremity. Pronator quadratus hemangioma poses a considerable morbidity in the functional quality of life of an individual. MRI remains the gold standard modality in diagnosing this rare entity. Additionally, histopathological examination of the lesion corroborates for the same. PQH follows an individualized protocol in the management of the tumour. A 35-year-old female presented with pain over her right wrist from past 1 year which was dull aching, non-radiating, localised to the palmar aspect of distal 1/3rd of the right forearm. There was no history of trauma or infection over the right wrist. Swelling was present over the palmar aspect of distal 1/3rd of right forearm, which was non-pulsatile, non-fluctuant, non-transilluminant, non-compressible, non-reducible and skin over the swelling was pinchable. The movements of the right wrist were painful and restricted. There was no distal neurovascular deficit. The MRI of her right wrist and hand suggested the presence of low flow vascular malformation within pronator quadratus muscle. The patient underwent excision biopsy of pronator quadratus in toto without any neurological complications. No recurrence was noted in the follow-up period for 12 months. The pronator quadratus hemangioma is a rare clinical entity where the natural course of disease and pathophysiology remains controversial. MRI serves as the gold standard in the diagnosis of intramuscular hemangioma. The management of such tumour has to be individualized based on the functional needs of the patient. The natural course of the disease has to be elicited to decrease the morbidity and enhance the functional quality of life of the patient.

Keywords Hemangioma · Intramuscular · Pronator quadratus · MRI · Excision biopsy

Introduction

Among the benign vascular tumours, hemangiomas remain the most common tumour. Hemangiomas are either congenital (80%) or traumatic (20%) in origin [1]. They occur most commonly in subcutaneous adipose tissue and muscles [2]. The natural course of hemangioma shows proliferation of vascular channels, fibroadipose replacement, intravascular clotting, atrophy, and involutionary alterations respectively [3]. Intramuscular hemangioma contributes to 0.8% of all hemangiomas, which follow an indolent natural course of disease with an uncertain etiology [4]. Intramuscular hemangiomas are most frequently seen in the thigh (36%), followed by the calf (17%) and rare in the upper extremity [1]. Such hemangiomas are associated with pain, limited range of involved joint movements and decreased functional quality of life. The management of these cases relies on the functional needs of the patients. After obtaining informed and written consent from the patient, we quote a rare case of pronator quadratus hemangioma and its management in a nutshell manner in view of invoking fundamentals and enhancement of knowledge among all the orthopaedic practitioners.

Case Report

35-year old female presented at our institution with a history of pain over right wrist from past 1 year which was insidious in onset, dull aching, non-radiating, localised to the palmar

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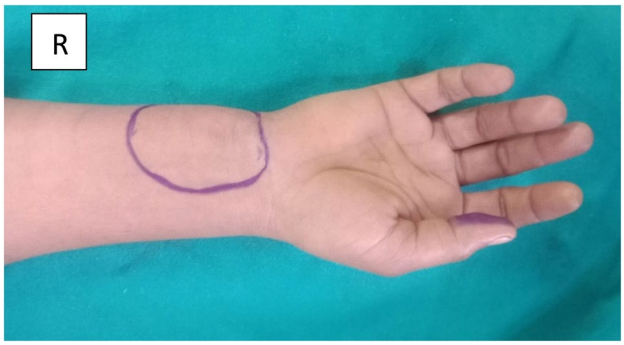


Fig. 1 Clinical image of right hand (marked area) showing the extent of the lesion

aspect of distal 1/3rd of right forearm, aggravated on movements and relieved with rest & medications. There was no history of trauma or infection over her right wrist. On examination, there was a diffuse swelling over the palmar aspect of distal 1/3rd of right forearm, which was non-pulsatile, non-fluctuant, non-transilluminant, non-compressible and non-reducible over distal 1/3rd of the ulnar side of the palmar aspect of wrist (as shown in Fig. 1). The swelling was found to be non-adherent to the skin and underlying structures. Notably, the distal neurovascular status was intact. However, the movements of the right wrist were painful and restricted (active palmar flexion of 30° and dorsiflexion of 20°) .

The plain radiograph of right wrist revealed specs of calcification in the region of distal end of radius (as shown in Fig. 2). The patient was subjected for an ultrasound examination of the right wrist which revealed numerous tubular internal hypoechoic areas that demonstrates the flow suggestive of vascular tumour (as shown in Fig. 3). MRI of right wrist revealed a well-defined, septated and lobulated mass measuring 21 × 24 × 30 mm (approx.) in the distal forearm in the deep anterior compartment in pronator quadratus muscle on T2/SPAIR and hypointense on T1 showing a few small foci of blooming on gradient-echo images (as shown in Fig. 4a–c).

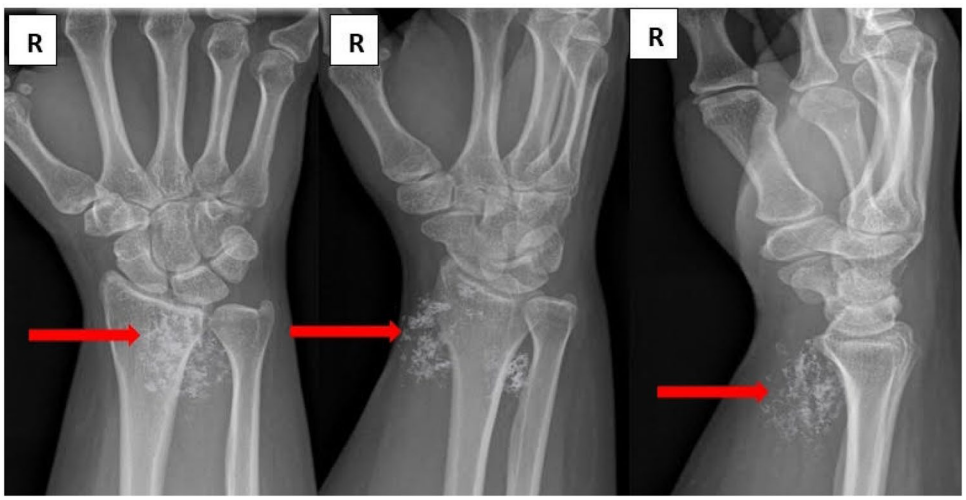


Fig. 2 Radiograph of the right wrist (AP, Oblique and Lateral) (marked area) showing the specs of calcification

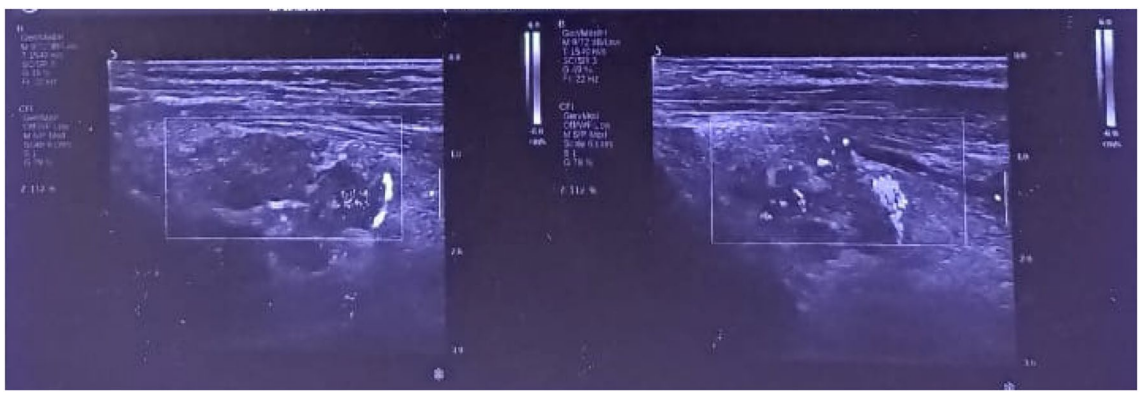


Fig. 3 Ultrasound of right wrist showing hypoechoic areas and low flow vascularity within the pronator quadratus muscular substance in the distal aspect of right forearm



Fig. 4 **a**, **b** (coronal section) and **c** (sagittal section): MRI of right wrist showing a lobulated mass in pronator quadratus muscle with gradient-echo changes

Under general anaesthesia, the patient underwent excision biopsy of pronator quadratus in toto without any neurovascular complications (as shown in Fig. 5a). Though there was an evidence of scalloping effect in the right distal end of the radius (as shown in Fig. 5b) but there was no evidence of encasement of tumorigenic cells over the neurovascular structures. Post-operative period was uneventful. Grossly it was partially circumscribed soft tissue mass measuring 5 × 4 × 2 cm. The cut section of the biopsied muscle showed variable sized vascular channels containing thrombus and haemorrhage along with yellowish solid areas (as shown in Fig. 6). Histopathology of the biopsied muscle revealed infiltrating tumour composed of variable-sized vascular spaces with a predominance of larger sized blood vessels. These are lined by endothelial cells and show RBCs in the lumen. Surrounding area shows mature adipose tissue suggestive of cavernous hemangioma of pronator quadratus muscle (as shown in Fig. 7).

Patient has been followed up at the regular intervals for 6 months post-operatively. At the end of 6 months, the patient showed active palmar flexion of 80º and active dorsiflexion of 70º of the right wrist (as shown in Fig. 8a, b). No recurrence was observed in the follow-up period for 12 months (as shown in Fig. 9). USG of right wrist showed no evidence of inflammatory changes in the pronator quadratus muscle during 12 months follow-up (as shown in Fig. 10).

Discussion

Among hemangiomas, the most common benign soft tissue tumours (7% of all soft tissue tumours), intramuscular hemangiomas contributes to 0.8% of all hemangiomas [2, 5–7]. Though a relative common and benign vascular soft tissue tumour, hemangioma is an endothelial neoplasm with relatively increased endothelial turnover [8]. The etiology of

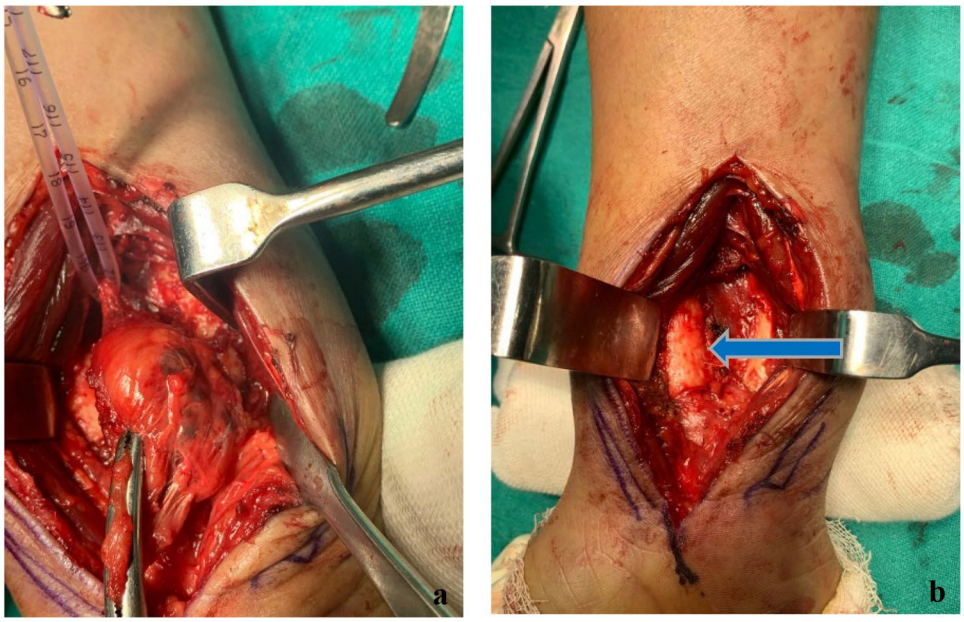


Fig. 5 **a**, **b** Intraoperative images showing the intramuscular (pronator quadratus) vascular malformation and scalloping effect of the distal end of the right radius (as shown in blue arrow), respectively

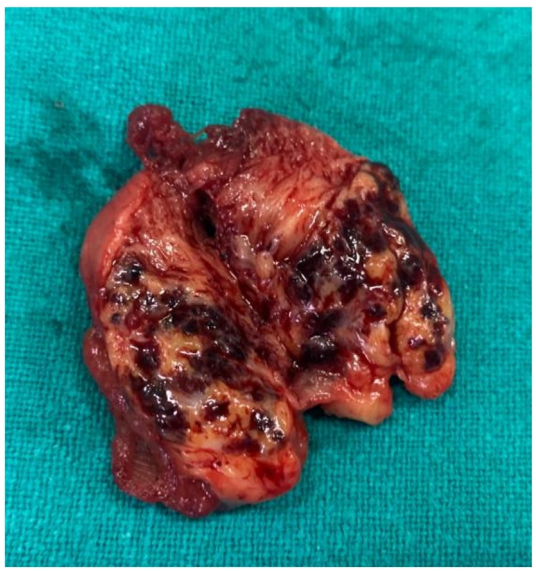


Fig. 6 Cut section of pronator quadratus specimen showing variable-sized vascular channels containing thrombus and haemorrhage along with yellowish solid areas

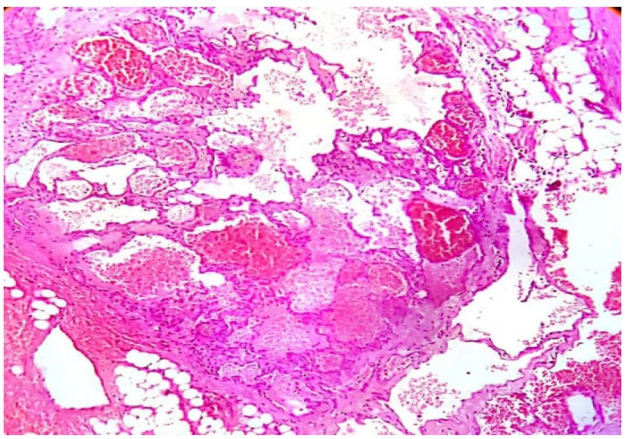


Fig. 7 HPE (× 40) of the lesion showing infiltrating tumor composed of variable-sized vascular spaces with a predominance of larger sized blood vessels and are lined by endothelial cells and show RBCs in the lumen

hemangioma is uncertain but congenital or traumatic causation contributes a little to its pathogenesis [9]. Hemangiomas are more common in infants and children with females being more commonly affected than males. 90% of the hemangiomas occur before the age of 30 years [1, 10]. The most common site of occurrence of hemangiomas are superficial tissues but occasionally it occurs in deep soft tissues such as liver, skeletal muscles, synovial lining, peripheral nerves and rarely bone [11–13].

Intramuscular hemangiomas are also called as intramuscular infiltrating angiolipomas. These lesions have no direct relationship with traumatic etiology. Out of 0.8% of intramuscular hemangiomas, lower extremity contributes for 45%, upper extremity 27%, head & neck 14% and trunk 14%. Wild et al. [7] and Rastogi et al. [5] reported that the quadriceps muscle (36%) represents the most common site of involvement for hemangioma followed by soleus muscle (17%) in the lower limb. Mencke et al. observed that masseter contributes 5% of intramuscular hemangioma followed by trapezius, periorbital, sternocleidomastoid, temporalis muscles, triceps and pronator quadratus [14]. Euicheol et al. reported a case of occurrence of hemangioma in pronator



Fig. 8 **a**, **b** 6 months post-op images showing palmar flexion of 80° and dorsiflexion of 70° of right wrist



Fig. 9 Post-operative surgical scar at 12 months follow up

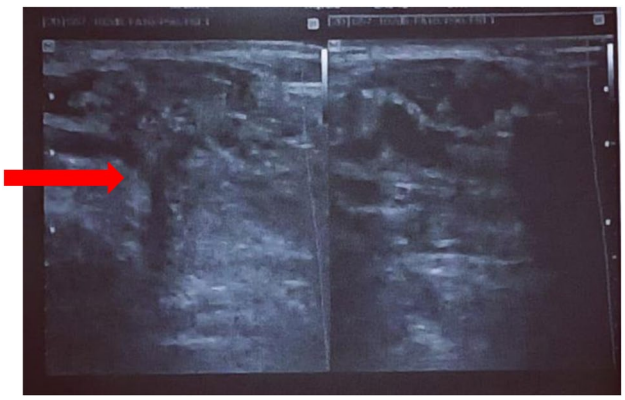


Fig. 10 12 months follow up USG of right wrist showed no inflammatory changes as compared with the left wrist

quadratus [15]. The review of intramuscular hemangiomas were tabulated in Table 1.

Clinically, intramuscular hemangioma presents as a slowly growing mass with pain (approx. 60% of cases) as the most common symptom. In about 80% of cases, it remains intramuscular in nature. The intramuscular hemangioma usually presents as a diffuse swelling, compressible in nature and present deep within the muscle involved. Pulsations, thrills or bruits may be absent since the vascular malformations within the muscle may be due to small vascular communications. In our case, the patient presented with chronic pain of diffuse nature which got aggravated by movements and swelling followed an indolent course to attain the present size. It is postulated that our patient developed a benign and hamartomatous neoplasm in the underlying muscle which gave rise to pain and restriction of movements of the right wrist.

Deep-seated intramuscular hemangioma pose a great diagnostic challenge. The radiographic analysis of right distal forearm with wrist showed soft tissue calcifications which are specific for cavernous hemangioma as mentioned in other studies [2, 5, 8]. USG of the right wrist showed low flow vascular malformation within the pronator quadratus muscular substance and the radial & ulnar arteries showed normal flow in the vasculature and which were not in any continuity with hemangiomatous lesion in the pronator quadratus. MRI remains the gold standard imaging

Table 1 Review of intramuscular hemangiomas from 2011 to 2020

Author ane year of the study	Location of intramuscular hemangioma	Clinical presentation	Imagining modality	Management
Hristov et al. [16], (2011)	Soleus	Pain over left lower leg for 7 months with h/o cavernous hemangioma in the same location with MRI diagnosis and open biopsy after attempted/failed surgery	Phlebography of left leg	Ligation of the feeding and draining veins of the intra-soleal cavernous hemangioma
Kiran et al. [17], (2012)	Vastus medialis	Swelling over left knee for 1 year	MRI of left knee	Surgical removal of hemangioma along with vastus medialis muscle and phleboliths
Doddanna et al. [18], (2014)	Buccinator	Swelling on left side of the face for 8 months	Colour Doppler and USG of the swollen area on the face	Diagnostic biopsy of the lesion
Lakshmi et al. [19], (2014)	Masseter	Swelling on the right side of the face for 6 months	MRI of face	Excision biopsy of masseter
Rai et al. [20], (2014)	Gastrocnemius	Swelling and pain from the back of the left knee for 2 years	MRI of left knee	Excision biopsy of medial head of gastrocnemius
Rastogi et al. [5], (2015)	Triceps	Pain and swelling over the posterior aspect of the left arm for 3 months	MRI of left arm	En bloc removal of triceps along with the mass
Surej Kumar et al. [21], 2016	Masseter	Swelling in the left cheek region for 3 years.	MRI and angiogram of face	Excision biopsy of masseter
Euicheol et al. [15], (2016)	Pronator Quadratus	Painful swelling on left wrist for 8 years	MRI of left wrist	En bloc removal of pronator quadratus with cephalic vein grafting for encased radial artery
Lahrach et al. [22], (2019)	Plantar 1st interossei	Left forefoot pain for 7 years with h/o sclerosant injection over plantar aspect of the foot	MRI of left foot	Excision of 1st plantar interossei in toto
Current study	Pronator quadratus	Diffuse swelling over palmar aspect of distal 1/3rd of the right ulnar border of the forearm for 1 year	MRI of right wrist and hand	Excision biopsy of pronator quadratus in toto

technique in diagnosing soft-tissue haemangiomas [9, 23]. T1 weighted MRI show isointense or hyperintense images with unclear margins whereas T2 weighted images show hyperintense lesions [24, 25]. In our case, MRI show consistent report as described in the literature and the lesion was well defined and intramuscular without capsular breach, no infiltration into the structures beyond pronator quadratus and no features of malignancy were noted. Homogeneous diffuse enhancement was observed in the post-contrast image with low flow vascular malformation within the muscle. No significant solid components were observed within the lesion. The well defined intra-muscular appearance of the hemangioma suggested resectability. Aforementioned features are suggestive of a benign lesion of vascular etiology.

Intramuscular deep-seated hemangioma is associated with neighbouring bone changes due to scalloping effect [26–28]. Ly et al. described bony changes as periosteal, cortical and medullary [26]. Our case had consistent cortical bony changes in the form of a scalloping effect. Hemangiomas neither metastasises nor undergoes a malignant transformation [29]. Management of intramuscular hemangioma follows a personalized protocol according to the tumor location, extent, growth rate, anatomical accessibility, local aggressiveness and cosmetic considerations. The management options vary for hemangiomas such as spontaneous regression [30], excision biopsy in toto [19–22], ligation of feeding vessels [16], radium agents [31], cryosurgery [32], carbon dioxide snow [33], radiation therapy [34–37] and sclerotherapy [38] etc. Intramuscular hemangioma never regresses spontaneously [7, 11, 12]. Hence, excision biopsy in toto resorted as the definite management of intramuscular hemangioma in our case, which provided better functional

outcome in terms of pain. It is imperative to anticipate the recurrence in case of inadequate excision where the researchers reported recurrence rate between 18 and 60% [9]. Patten et al. suggested the usage of interferon-α in case of recurrence [8]. The recurrence is most common in mixed type followed by capillary and then cavernous haemangiomas. In our case, pronator quadratus hemangioma belongs to cavernous type as they have very least possibility for recurrence after surgical excision. There was no recurrence reported in the follow-up period of 12 months.

To date, we found one published literature on pronator quadratus hemangioma. Euicheol et al. [15] reported a case of pronator quadratus hemangioma (8 years of duration) which was found to be encasing distal end of radial artery intra-operatively. Excision biopsy of pronator quadratus in toto had been performed along with cephalic vein grafting for the encased portion of the radial artery. They reported no recurrence with good functional and cosmetic results for the same patient.

Conclusion

Being a benign tumour of vascular channels, MRI is the gold standard in diagnosing cavernous hemangioma. Indeed, intervening at the earliest with meticulous clinical and radiological correlation is required to diagnose such rare clinical entity in view of improvising the functional quality of life. The choice of definitive treatment for pronator quadratus cavernous hemangioma was excision biopsy in toto which provided the better functional results in our patient.

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Compliance with Ethical Standards

Conflict of interest The authors declare that they have no conflicts of interest.

Ethical standard statement All procedures performed in studies involving human participant was in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its alter amendments or comparable ethical standards.

Informed consent Informed consent was obtained from the individual participant included in the study.

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