CASE REPORTS IN PLASTIC SURGERY AND HAND SURGERY
2024, VOL. 11, NO. 1, 2393819
https://doi.org/10.1080/23320885.2024.2393819

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CASE REPORT

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# Intramuscular hemangioma of the infraspinatus muscle: a rare presentation

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**ABSTRACT**

Intramuscular hemangiomas (IMH) are extremely rare, accounting for 0.8% of all hemangiomas. IMH must be included in the differential diagnosis of soft tissue masses, and unexplained muscular pain. We herein describe the case of a patient who presented with an atypical localization of IMH in the infraspinatus muscle.

**ARTICLE HISTORY**
Received 2 April 2024
Accepted 11 August 2024

**KEYWORDS**
Intramuscular hemangioma; infraspinatus muscle; vascular malformation; chronic pain; surgical excision

## Introduction

Hemangiomas are rare, benign, proliferative vascular neoplasms caused by endothelial cell hyperplasia and should be differentiated from vascular malformations, which are not true neoplasms, but rather localized defects of vascular morphogenesis caused by dysfunction in embryogenesis and vasculogenesis [1, 2]. According to the latest World Health Organization (WHO) classification of soft tissue tumors, vascular tumors are classified based on their malignant potential as benign, intermediate (locally aggressive) or malignant [3]. Furthermore, the International Society for the Study of Vascular Anomalies (ISSVA) follows a similar classification system [4]. Despite similarities between the two systems, discrepancies exist with regard to the classification of certain lesions. Such is the case of the Intramuscular hemangioma (IMH), classified by the WHO as a benign vascular tumor, in contrast with the ISSVA, which currently places IMH as a provisionally unclassified vascular anomaly, suggesting IMH may be more akin to a vascular malformation, rather than a true endothelial neoplasm.

The first description of Intramuscular hemangioma is generally attributed to Liston in 1843. Since then, numerous cases of IMH have been reported in the literature [5]. Interest in these vascular lesions lies both in their rarity (<0.8% of all hemangiomas [5]) and location, which can span any muscle, but often show a predilection for the lower limb, followed by the head and neck regions [6].

Imaging modalities, especially ultrasonography (US) and magnetic resonance imaging (MRI), are pivotal in the work-up of both vascular tumors and vascular malformations [7, 8].

Treatment of IMH involves surgical excision, sclerotherapy and arterial embolization. Surgery is the main treatment. Partial excision is associated with recurrence rates of up to 18% due to the infiltrative growth pattern of the lesion, in spite of which, IMH doesn't produce metastasis [5]. When excision is not feasible, or the patient does not wish to undergo surgery, or debulking is required prior to surgery, compression sclerotherapy [9, 10] with sclerosing agents (sodium tetradecyl sulfate, ethanol, polidocanol, hypertonic

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saline, and sodium morrhuate) or arterial embolization [11] can be valid therapeutic options.

Reports of IMH are plentiful among the literature, but to the best of our knowledge, there are very few descriptions of IMH located within the infraspinatus muscle: Yetişgin A, et al. described the case of a hemangioma presenting in the left infraspinous fossa [8], and a French multicentric study reported on two intramuscular capillary-type hemangiomas (out of 66 cases) located in the infraspinatus muscle [12]. IMH of the infraspinatus muscle are therefore extremely rare. Lesions located in this region pose a challenge for the surgeon and can limit adequate excision due to limitations in exposure. We herein describe the case of a patient presenting with chronic back pain irradiating to his left arm, with concomitant reduced range of motion. Initial diagnostic suspicion was that of an elastofibroma dorsi, but later imaging with MRI revealed the lesion to be an intramuscular hemangioma located within the left infraspinatus muscle. Surgical excision of the lesion was carried out without complications and completely resolved the patient's pain.

## Report of the case

A 34-year-old male patient, with a prior history of arterial embolization of an IMH in the left infraspinatus muscle, was referred to our department with a two-year history of persistent pain over the left lower scapular region. Pain was present at rest and exacerbated with movement of his left arm, limiting his range of motion. Physical examination did not reveal any masses or overlying skin changes, but palpation over the infrascapular region did exacerbate the pain.

Two years prior the patient had an ultrasonography of the lesion carried out, which revealed a 2.2 cm intramuscular hypoechoic lesion in the scapular area initially compatible with the diagnosis of an elastofibroma dorsi (Figure 1). This was followed by a computed tomography (CT) scan which further characterized the lesion as a well-defined hypodense mass with a maximum diameter of 1.8 cm located deep within the left infraspinatus muscle (Figure 2). In addition, MRI imaging further revealed a heterogenous mass of 1.7 × 1.8 × 2.1 cm in size within the left infraspinatus muscle in close contact with the scapular bone (Figure 3).

The patient underwent a core needle biopsy that revealed a vascular lesion infiltrating the muscular tissue with capillary proliferation and a lobulated architecture. Immunohistochemical analysis was also performed and positivity for CD31, CD34 and ERG was found. The histological diagnosis of intramuscular hemangioma was made.

Embolization was carried out through feeding arteries branching from the subscapular artery. Post embolization angiography showed no residual lesion.

Despite the initial improvement in pain, reported by the patient, recurrence in symptoms was observed shortly after. Three months after the embolization an MRI revealed recurrence of the vascular lesion as well as a slight increase in its size, measuring 2.4 × 2.0 × 1.7 cm (Figure 4). The patient now had referred pain radiating from his left shoulder and loss of strength in his left arm. Treatment with nonsteroidal anti-inflammatory drugs (Dexketoprofen 25 mg TID) was ineffective at pain control.

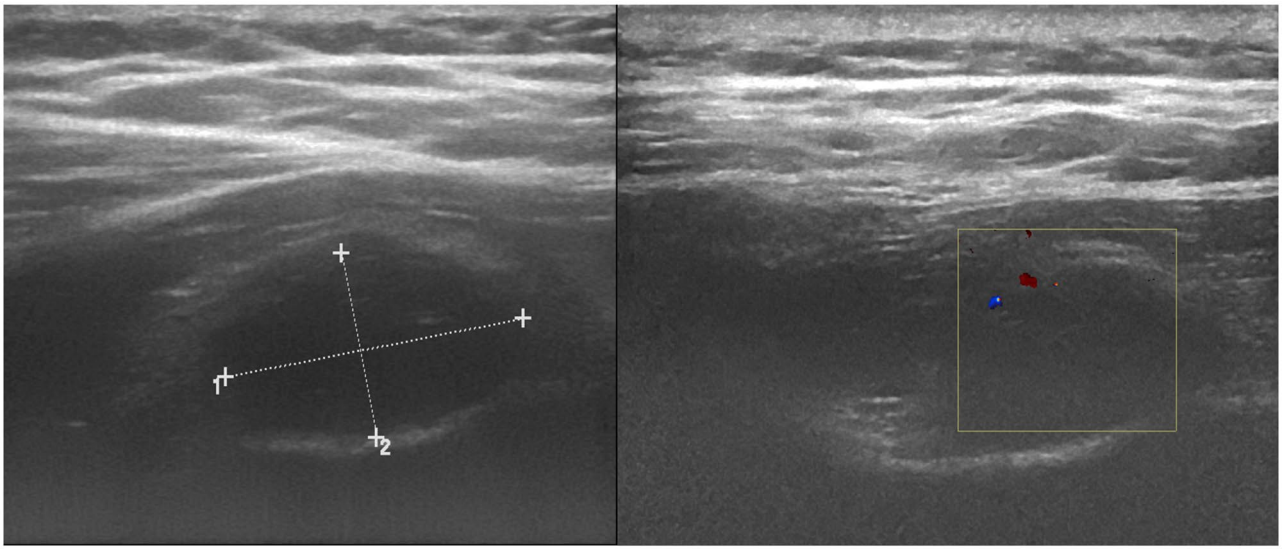


Figure 1. Ultrasonography showing a well-defined, hypoechoic lesion measuring 2.2 x 1.3 cm. Peripheral vascular flow is observed on Doppler imaging.

Surgical excision of the lesion was performed as follows. Under general anesthesia and with the patient in a prone position, an S-shaped incision was made just inferior to the spine of the scapula in the left scapular region. The infraspinatus muscle was exposed, and an intramuscular, firm mass was observed. Only partial excision was achieved likely due to the scar tissue developed after embolization. Neither bleeding nor any other intraoperative complications occurred. The postoperative period was uneventful. Histological examination confirmed the diagnosis of intramuscular hemangioma and revealed a microscopic residual resection (R1) (Figure 5).

At 8 months follow-up, the patient had referred pain resolution and a complete recovery in range of motion of his left arm. An expeditious return to work without the need of analgesia was achieved. The resulting scar healed uneventfully.

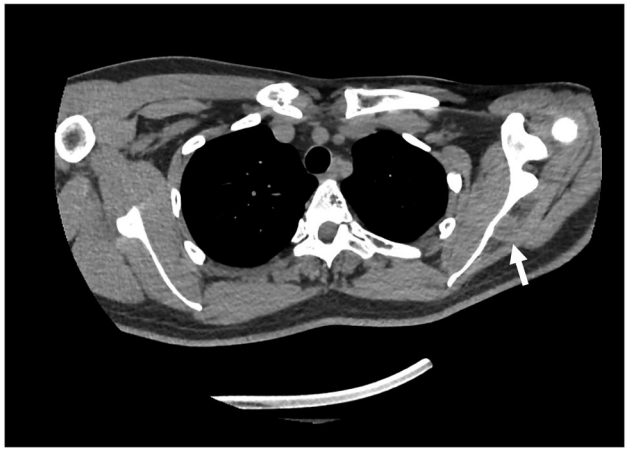

Figure 2. (arrow) Computed tomography showing a well-defined, round hypodense mass measuring 1.8 cm in diameter, immediately posterior to the left scapula and deep to the left infraspinatus. Less dense adipose tissue can be observed surrounding the lesion.

## Discussion

The etiology of IMH is unknown, and despite many patients attributing their initial symptoms to past trauma, a congenital origin seems far more likely. However, unlike infantile cutaneous hemangioma, IMH does not regress spontaneously and is usually detected in the second or third decades of life. Initially, the lesion may be asymptomatic, but continual growth exerts pressure on nearby structures leading to non-specific soft tissue complaints over time, such as pain, swelling, and a palpable firm or fluctuating mass. On some occasions, the mass can be pulsatile or be accompanied by a bruit [13]. Due to the non-specific nature of these symptoms, IMH is frequently misdiagnosed, and a high index of suspicion is generally required [14]. Furthermore, the correct diagnosis of IMH is paramount, and a differential diagnosis must be made with other masses of the musculoskeletal system, especially those with malignant potential, such as angiosarcoma. Hence, surgical excision is warranted in these patients, both to relieve symptoms and to exclude malignancy.

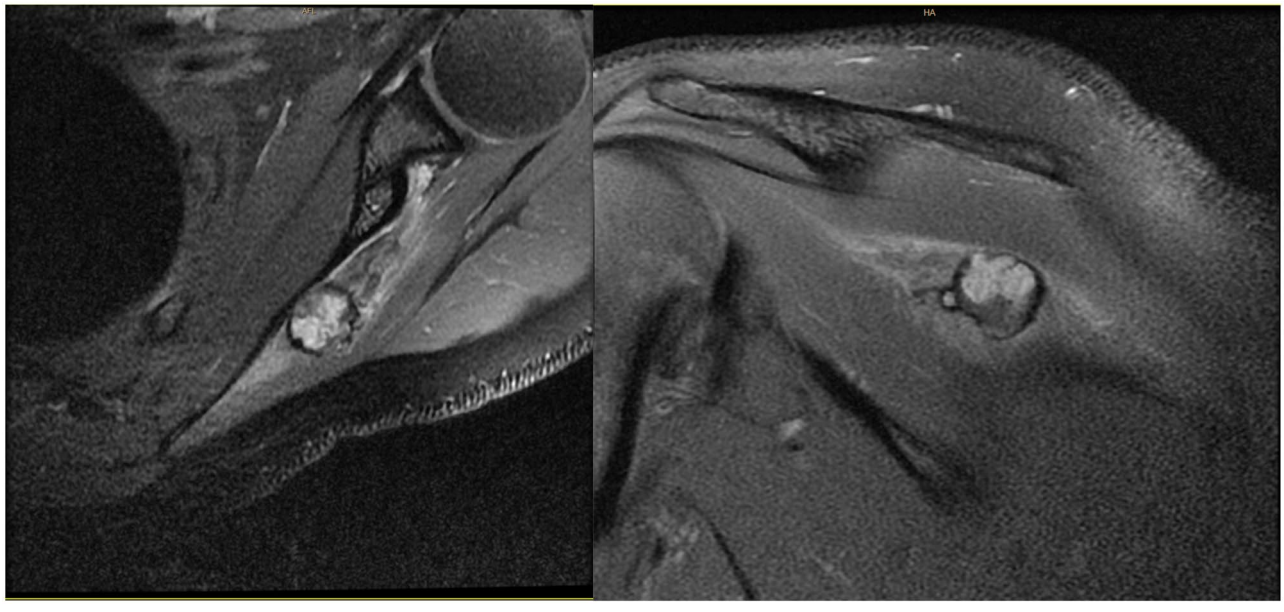

Figure 3. Magnetic resonance imaging shows a well-defined lesion measuring 1.7 x 1.8 x 2.1 cm with small areas of signal hypointensity suggesting fibrosis and large areas of hyperintensity on T2 weighted sequences, which persist upon fat suppression.

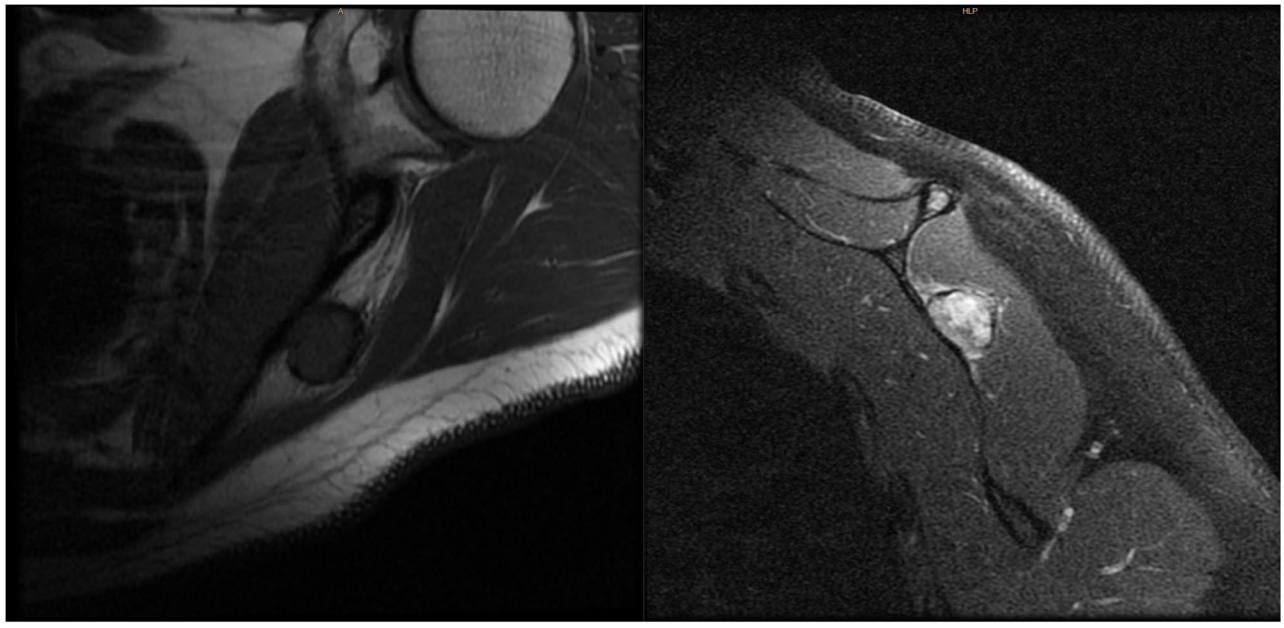

Figure 4. Magnetic resonance imaging 3 months after arterial embolization, showing a persistence/recurrence of the vascular lesion, having slightly increased in dimensions to 2.4 x 2.0 x 1.7 cm.

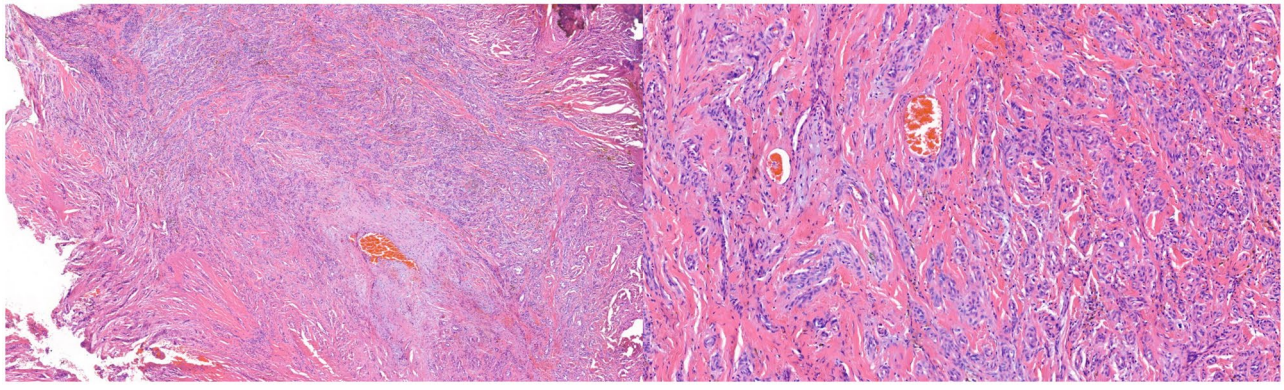

Figure 5. Skeletal muscle tissue infiltrated by numerous small blood vessels without features of malignancy. LEFT (Hematoxylin & eosin, x4) RIGHT (Hematoxylin & eosin, x10).

In our case, we presented a 34-year-old male with no past history of trauma to the region, but with pain and loss of strength in his left arm. Physical examination also did not reveal any skin or subcutaneous lesions.

US including Doppler Imaging allows the clinician to assess the flow pattern of the vascular lesion, while follow-up with MRI including three-dimensional (3D) contrast enhanced-magnetic resonance angiography sequence (CE-MRA) allows further characterization of the US findings as well as delineating the extent of the lesion [6]. Findings on MRI consistent with IMH include an intermediate signal on T1 weighted sequences and an intense signal on T2 weighted sequences, though the latter may not necessarily be present in selected cases [15]. In addition, MRI also provides a valuable roadmap for preoperative planning. Interestingly, the findings on imaging correlate well with histopathology results, upon which a definitive diagnosis is eventually made on histopathology [7]. Allen and Enzinger described a classification system for IMH based on the predominant vessel size observed on pathology; small-vessel, large-vessel, or mixed types [5]. This classification correlates well with location and prognosis [16]. Findings on histopathology are usually consistent with blood vessels proliferation within muscle fibers, and positivity for CD31 and CD34 [17].

Since some imaging findings can be reliably distinguish IMH from other lesions, a preoperative core needle biopsy is rarely required [18]. However, due to the unusual location of the IMH presented in our case, we did perform a core needle biopsy to confirm the diagnosis.

Treatment options for IMH are surgery, embolization, or sclerotherapy; surgery and embolization can be combined together to reduce the risk of bleeding and the rate of recurrences [10, 19]. A retrospective multicentric French cohort regarding intramuscular hemangiomas treatment, reports three major complications: bleeding requiring transfusion during surgical removal of the lesion, bilateral cerebellar ischemia after embolization and intraoperative hemorrhagic shock within the first 24 h after embolization [12]. We did not experience any complications. Both embolization and surgery were performed uneventfully.

Bella and Cheng et al. found a recurrence rate of 14% at 2 years, 27% at 5 years, 38% at 10 years, with surgical margins and tumor size being the only independent variables predicting local recurrence rates [20].

On 8 months postoperative follow-up, our patient has not show any signs or symptoms of recurrence, however longer follow up periods will be needed to rule out any relapse.

## Conclusion

Intramuscular hemangioma can very rarely present in the infraspinatus muscle. In such cases, surgical excision of the lesion can achieve complete remission of pain symptoms. In the scenarios where the patient is not a suitable candidate for surgery, sclerotherapy and arterial embolization may be considered. Ultrasonography and magnetic resonance imaging are useful adjuncts in the work up and preoperative planning of these lesions.

## Disclosure statement

No potential conflict of interest was reported by the author(s).

## Consent

The patient agreed and provided written informed consent to publication of his data.

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