CASE REPORT

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Intramuscular hemangioma of the arm: ultrasonography and pathology features

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Abstract

Hemangiomas are between the most frequent soft tissue masses and despite the vascular origin, they do not generate metastases and do not have malignant evolution. They are frequent in childhood and female sex is more frequently affected. If deeply located, these tumors are difficult to diagnose and thus an imaging method is often needed, but conventional radiology is not sufficient. If superficially located, it frequently involves the skin and subcutaneous tissues, but in the deeper layer, they are often intramuscular. Clinical findings of intramuscular hemangioma include swelling, pain and sometimes loss of muscle function.

Keywords: ultrasonography, muscle, CD31, CD34, intramuscular hemangioma.

Introduction

Intramuscular hemangioma (IH) is a rare benign vascular tumor, accounting for around 7–10% of all soft tissue tumors [1, 2] and less than 0.8% of all hemangiomas [3]. Skeletal muscles can often be involved as a location [4]. General agreement is that IH might be more frequently found in females and that even if age is not an important predictive factor, IH are is usually diagnosed within the three first decades [2]. As a location, there are papers positioning head and neck on the first place, followed by trunk and extremities [5] but there are others considering the cranial involvement unusual [4, 6–8].

Before surgery, the diagnosis of IH is often difficult due to non-specific clinical and conventional radiology findings [9]. Besides this, fine-needle aspiration biopsy is usually unclear due to excessive blood in the specimen [6]. Clinical findings of IH include pain, swelling and in late presentation, loss of function. As conventional radiology (CR) brings no further information, ultrasonography (US) and magnetic resonance imaging (MRI) are frequently asked.

Differential diagnosis should include other soft tissue and vascular tumors, vascular malformations and sarcomas. Intramuscular lipoma has no vascular involvement and is more indolent, whereas angiosarcoma has no lobular architecture and more endothelial atypia. After diagnosis, IH does not usually need treatment, unless they cause intense pain, function impairment, increase in size or at least cosmetic concerns, situation when embolization or surgical excision might be considered. If surgery decided, extended excision into normal tissue should be taken into consideration, as hemangiomas tend to infiltrate adjacent tissues and incomplete removal could lead to recurrence [7, 10, 11], which has a moderate to high risk, ranging from 18% to 61%. If surgical excision is contraindicated or patient does not agree, embolization, sclerotherapy and radiation therapy could be considered. More than this, as the most common complication of surgery is hemorrhage, embolization prior to excision, could be a solution to decrease blood loss [12–14].

Aim

The aim of this case report was to assess the US and pathology features of an intramuscular hemangioma of the arm, in a 29-year-old woman.

Case presentation

A 29-year-old woman, presented with a mass in the posterior aspect of the arm, since less than two months. The patient had no history of trauma before swelling of the arm. Her biological exams showed nothing particular, with slightly increased erythrocyte sedimentation rate (ESR 11 mm/h) and normal C-reactive protein (CRP). Her complete blood count was as follows: white blood cells count $6.2\times10^3/mm^3$, hemoglobin 12.8 g/dL and platelet count $267\times10^3/mm^3$.

US revealed a lobulated hyperechoic mass, compared to the muscle structure surrounding it, close to humeral bone cortical (Figure 1, A and B). Remarkable was that there was no Doppler signal present (Figure 1B).

As the US came with some information, but failed to clarify the diagnosis, a biopsy was performed, with a piece having an elastic consistency.

Histopathology of the biopsy sample showed a well-

defined, but unencapsulated mass, with skeletal muscle bundles interspersed with variable sized blood vessels, with thick-wall, reduced and fibrous stroma and sinusoidal growth pattern. There were few capillaries filled with red blood cells, others empty or with recent-organized thrombi. The microscopy findings were consistent with the diagnosis of intramuscular hemangioma and CD31 and CD34 immunostaining confirmed their vascular endothelial origin, by revealing positivity in lining cells of the vascular spaces, which proliferate between muscle fibers (Figures 2 and 3).

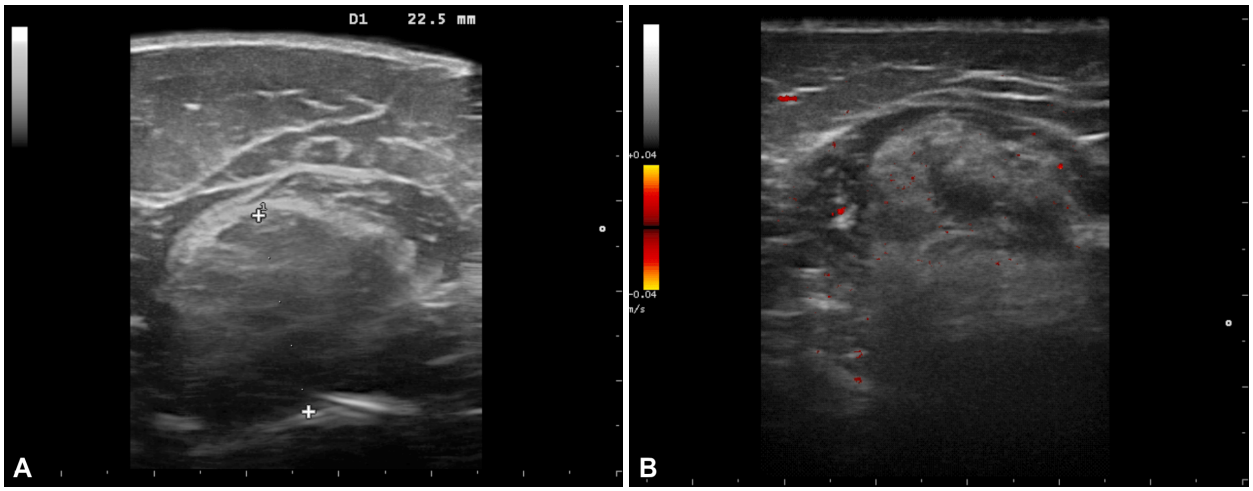

Figure 1 – ***Description. Transverse (A and B) sections, at the level of triceps muscle in the mid-third of the arm: (A) Grayscale image showing unhomogenous area (22.5 mm), deep to the subcutaneous tissue, well-defined, hyperechoic compared to muscle; (B) Power Doppler (PD) ultrasound negative mass, in the same area.***

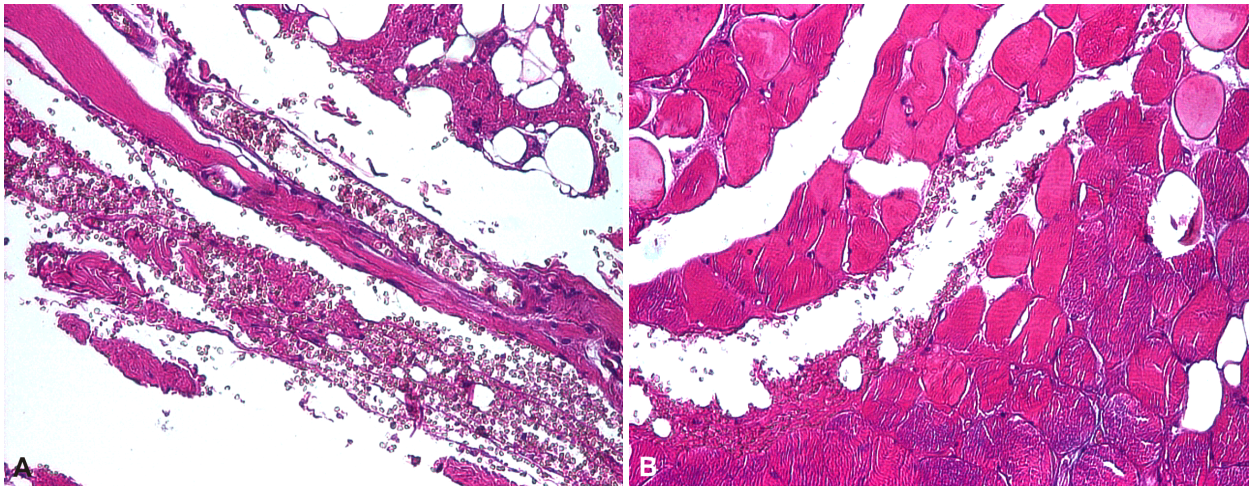

Figure 2 – ***(A) Intramuscular hemangioma, vascular spaces among muscle fibers (HE staining, ×100); (B) Vessels filled with red blood cells, some containing organized thrombi (HE staining, ×200).***

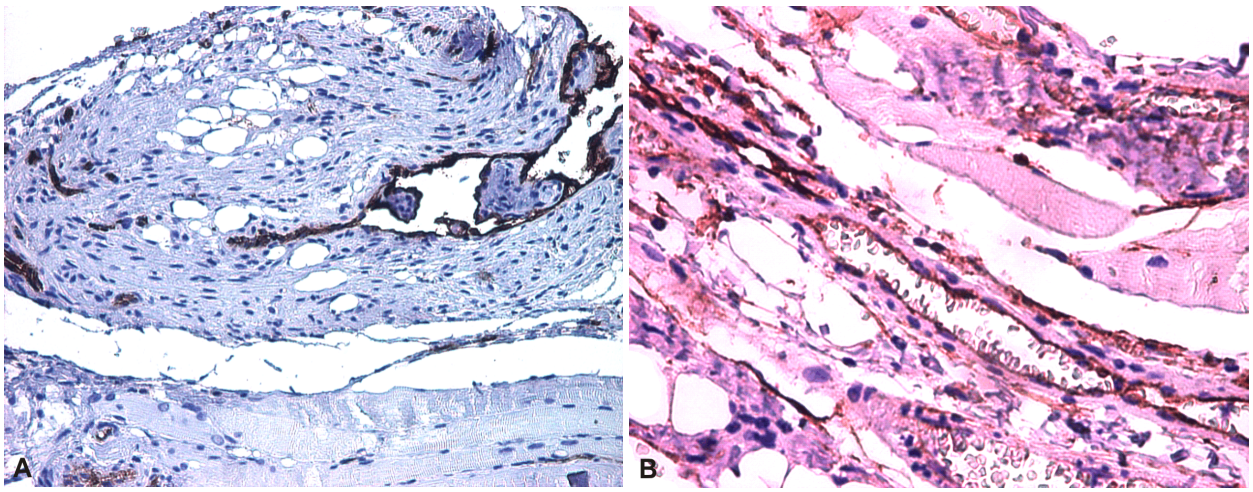

Figure 3 – ***(A) CD31-positive expression in the endothelial cells lining the vascular spaces (CD31 immunostaining, ×200); (B) CD34-positive expression in the endothelial cells lining the vascular spaces (CD34 immunostaining, ×200).***

After biopsy, patient decided for embolization, prior to surgical excision, but unfortunately, the symptoms reoccurred six months later. At this time, patient was pregnant, which made her refractory to any other surgery or needle biopsy. As ultrasonography is a non-invasive method, easily performed by an experienced examiner and well supported by patient, we decided to wait and monthly evaluate the tumor in order to identify any increase in dimensions or vascularization.

Discussion

Intramuscular hemangiomas are benign vascular tumors, which encounter less than 1% of all hemangiomas and may develop in the extremities and the trunk, with a distribution by sex, which indicates predominance in women [15–19]. As the mass develops most frequently in adults, our case is clearly overlapping the literature.

The etiology of these tumors is unknown, but trauma or hormonal changes might be involved in proliferation of embryonic vascular tissue. Intramuscular hemangiomas usually have distinct margins, do not present with vascular signs, such as skin color changes and should be differentiated of neurofibroma, lipomas, dermoid cyst, enlarged adenopathy.

Clinically, those tumors might be described as localized mass, with distinct margins and elastic structure, with little to no compressibility. Usually, it lacks any specific vascular signs, like color changes, specific to superficial hemangioma. The final diagnosis is made by histopathology, on biopsy sample. There is a general perception that needle biopsy is contraindicated because of the high risk of bleeding, but due to the fact that besides vascular structure, the intramuscular hemangioma contains variable amounts of non-vascular tissue, like fat and fibrous tissue [6, 20], which due to pressure from surrounding structure prevent hemorrhage and even prevent free circulation of blood inside hemangioma. This explains the fact that there is no Doppler signal in US, as the blood flow inside hemangioma is low. In the same time, the presence of an amount of fat tissue, could explain the posterior attenuation of the image in ultrasound.

Anyway, the most important imaging method in evaluation and diagnosis of hemangioma remains MRI, but because of its low accessibility, US might increase in importance, as can offer information on location, size and contacts of the mass.

Conservative treatment is usually recommended as the first line of therapy, with regular follow-up and US monitoring of the shape, size and Doppler, especially in situations where surgery is not recommended or patient does not agree. However, treatment for intramuscular hemangioma might be necessary if there are specific symptoms, with non-responsive pain, functional impairment, increase in size and cosmetic problems. Surgical excision, if decided, should include a rim of normal surrounding tissue as a mainstay of treatment, as it reduces the local recurrence rate [21, 22]. Preoperative embolization could be helpful for the control of intraoperative bleeding.

Conclusions

US, a non-invasive, highly accessible and accurate imaging tool, might represent the method of choice in evaluation and monitoring of intramuscular hemangioma prior and after surgery.

Conflict of interests

The authors declare that they have no conflict of interests.

Informed consent

Written informed consent was obtained from the patient for this case report and any accompanying images.

Author contribution

Marius Eugen Ciurea and Ananu-Florentin Vreju contributed equally to the manuscript.

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