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Intramuscular Capillary Hemangioma and Resulting Kasabach-Merritt Phenomenon: A case report

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Abstract

The term “hemangioma” has been incorrectly applied to a wide variety of vascular lesions. Hemangiomas are true vascular neoplasms with evidence of increased endothelial cell turnover. This case had a variety of anatomical positions meaning that every interventionist and orthopedic had a problem in treating and The occurrence of Kasabach-Merit syndrome adds to the complexity of the matter. Case presentation: a 29-year-old man with an eight years history of a round red lesion on his buttock that progressed and become a huge mass so the orthopedic surgeon could not excise the lesion the first time. After the first unsuccessful surgery, there was a disturbance in the patient's blood tests, and the patient was diagnosed with Kasabach Merit.In this case, we had some problems with anatomy and when we occluded the main artery by CT-guided interventions, a small particle detached from the artery and made the pulmonary emboli. After this intervention, the mass lesion had been excised successfully.

Keywords: *Hemangioma, Intramuscular, intervention, kasabach -Merritt*

Introduction

A wide variety of vascular lesions are incorrectly referred to as hemangiomas. Hemangiomas are true vascular neoplasms that exhibit a high rate of endothelial cell turnover(1) There is a subset of intramuscular vascular tumors that consist of capillary-like vessels, generally referred to as "intramuscular angiomas capillary" or "intramuscular hemangiomas small vessels." (2). Angiolipoma infiltrating into the skeletal muscle is another name for the lesion” (3). Capillary intramuscular hemangiomas are sometimes called intramuscular hemangiomas. (4) Among all hemangiomas, 0.8% are internal hemangiomas, which is a rare benign tumors (5). There were 32% of hemangiomas on the lower limb, 27% on the head and neck, 24% on the upper limb, and 17% on the trunk. (6) The vast majority of intramuscular hemangiomas are asymptomatic and are discovered during investigation for other conditions. (5) Symptoms can arise, however, if the tumor becomes enlarged and compresses or pushes the nearby muscles and nerves. Most clinical lesions affect the lower limbs, but they can affect any skeletal muscle. Enlarging intramuscular hemangiomas makes treatment more challenging. In the early stages, intramuscular hemangiomas have no clear symptoms, making them impossible to detect; thus, they are frequently misdiagnosed. In this report, we describe a patient who had intermittent incidental pain in the right buttock and radiating pain in the back of the thigh. He underwent long-term treatment for lumbar disc herniation. And began long-term treatment while her pain persisted and frequently returned. (7)

Case presentation:

A 29-year-old man presented with an 8 years history of a red area on the right buttock(figure1). This patient was referred to the hospital two years after the occurrence of this lesion and underwent MRI at which point the patient was diagnosed with AVM/AVF. The patient underwent angiography. Right lower limb angiography showed right internal artery AVM.(figure2) No invasive procedure or surgery was performed on the patient at that time. The patient had frequent visits to the doctor and no surgery was performed on him. After 8 years the size of the lesion increased dramatically, which suddenly and spontaneously caused severe bleeding and referred to an orthopedic surgeon., The orthopedic surgeon requested MRI and angiography for the patient.MRI showed that” Large abnormal signal in the deep portion of right buttock measuring about 143*140*92mm high on T2 and iso on Tl with the signal void area on T2 mostly due to hypervascular mass involving gluteus maximus(recurrence of sarcoma?) and with abnormal

signal in adjacent posterior subcutaneous adipose tissue which could be due to inflammation/infiltration of subcutaneous adipose tissue by this mass. No extension to bony structures.
Angiography showed that " a mass lesion in the right gluteal region is seen that shows significant multiple feeding tortuous arteries originating from the right internal iliac artery suggestive of severe tumoral neo angiogenesis with significant tumoral blush". The patient underwent surgery for resection of the lesion. During the surgery, due to the vascularity of the mentioned lesion and severe bleeding during the surgery, only the lesion was compressed and the surgery was completed without resection of the lesion, and the patient was referred to a radiologist for angioembolization After the surgery platelet counts reduce to 15000 and the "Kasabach-Merritt" phenomenon happened.

Figure1. A 29-year-old man with a round enlarging area on his right buttock

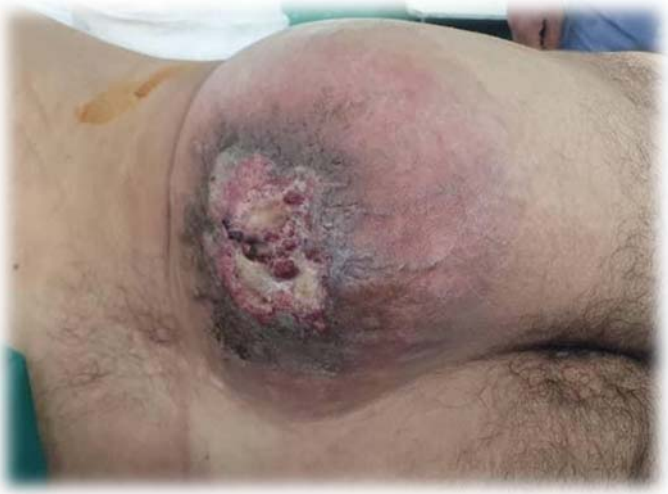
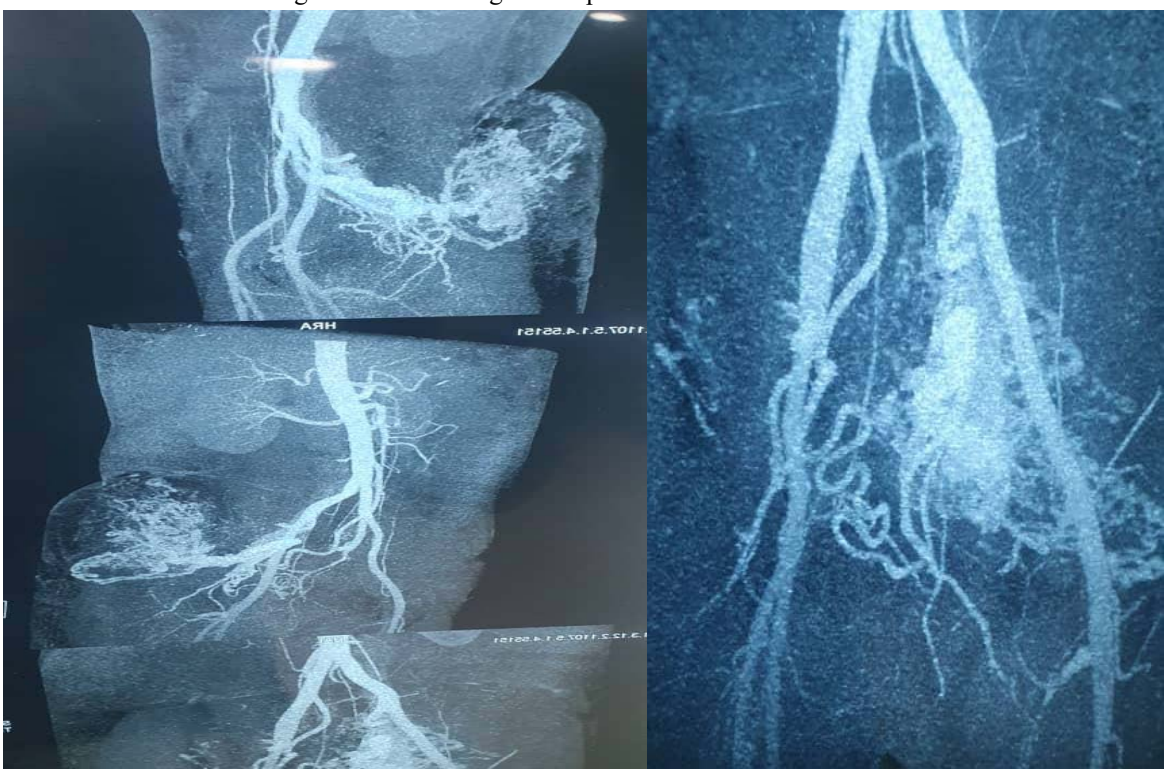


Figure2. MRI findings in the patients show an AVM/AVF



Method:

Interventional radiology:

First, from left femoral access aortography was done. In aortography multiple feeding arteries as the following description show:

1-enlarged tortuous vessels originated from the inferior mesenteric artery

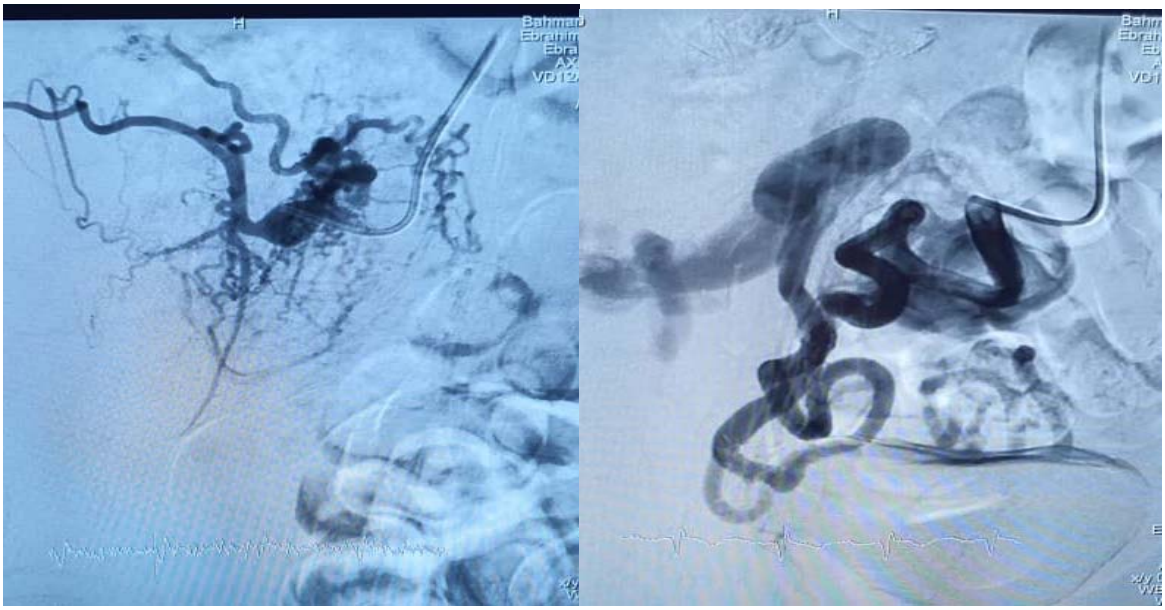
2-multiple tortuous vessels from internal iliac
3-multiple branches from a deep femoral artery(figure3.)

Due to the cephalocaudal orientation of the tumor-feeding vessels, for better access to the target arteries, arterial access was taken from the left brachial artery. After catheterization of the main branch of the tumor (originating from the IMA), a large vascular shunt was observed at the tumor site. Due to the patient's condition and the risk of pulmonary embolism, first, the shunt flow was reduced with a particle (PUV 1500-700), then using a concentration of 50-50 glue (50% history and 50% lipiodol) were injected into the feeding artery through a microcatheter. The flow above the shunt moved some of the adhesives to the lung parenchyma, but fortunately, the patient was not symptomatic and the injection continued. The vascular shunt was completely closed. ,80% of the vascular bed of Other vascular branches were closed with glue and particle.

After angioembolization, the patient underwent resection surgery and the lesion was completely removed and a pathology sample was sent. “Intramuscular capillary hemangioma. The overlying skin is focally ulcerated. Foci of ischemic infarction seen.Peripheral margins are free ، Small foci of proliferated small vascular structures “

After surgery(figure4), we referred the patient to a physiotherapy clinic and he underwent a series of exercises to rehabilitate his muscle strength. Based on a similar case report (4)we advised the patients to only isometric contraction of the lower limb muscles each day without hip movement in the first week. The next week, he was encouraged to move the hip; aquatic therapy was then added and she was thus able to gradually recover her muscle strength.

Figure3. Angiographic view of vessels in hemangioma and resected mass.



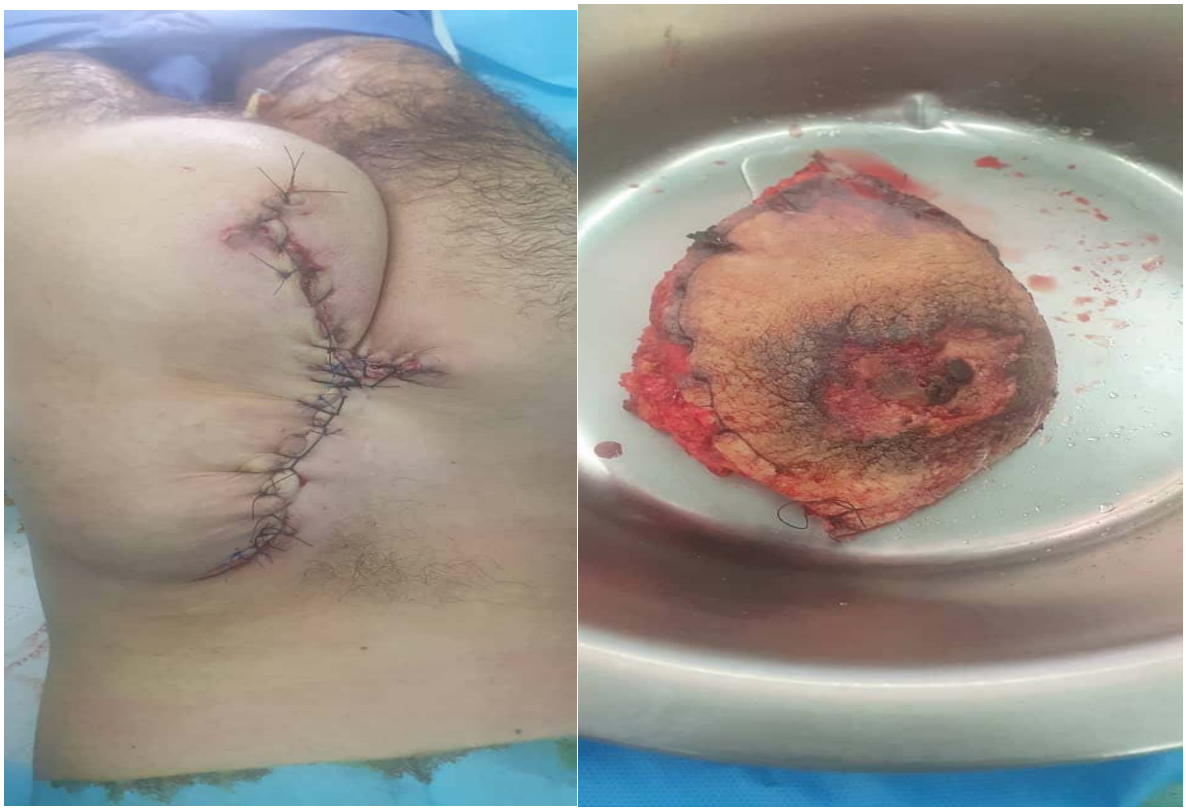
Discussion and conclusion:

An intramuscular hemangioma is referred to as a benign vascular tumor that contains neoplastic proliferations of blood vessels and often occurs in skeletal muscle. In this, tumor vascular endothelial cells proliferated increasingly and this occurs after trauma; in histology aggressive growth and no obvious capsule or boundary are obvious. We don’t know exactly why intramuscular hemangiomas occurred, while both trauma and congenital causes have been suggested to be the main causes. (8) in addition some studies demonstrated traumatic and hormonal influences that may contribute to the etiology or growth spurts of the tumor. (9, 10) Mostly in intramuscular haemangioma different types of mature adipocyte tissues had been shown. That is why these tumors are called intramuscular angiolipomas. Some symptoms of patients include muscle swelling, pain, infiltrative scleroderma involving the whole muscle, fibrosis, calcification, ossification, and other bone damage because of anoxia of myocytes that occurs due to poor local blood circulation and accumulation of metabolites. While most patients with intramuscular hemangioma are asymptomatic in the early stage. In the later stage, patients may develop myosclerosis, contracture, and
muscle and joint deformities and dysfunction. Most patients have a slowly enlarging lesion without any cutaneous change and sometimes they have pain, especially after exercise

without any bruit, pulsation, or thrill (11). Deep location, variable size and shape, lack of specific clinical symptoms, and absence of obvious biological characteristics, make the diagnosis more difficult so the diagnostic rate of intramuscular hemangioma is only 8% to 19%.8 preoperatively. So, it can be missed clinically and must be differentiated from soft tissue malignant tumors and other conditions(7). In the present case, the patient demonstrated the abovementioned slowly developing round red lesion that was typical but nonspecific symptoms, and after 8 years of undiagnosed we take an MRI and angiography and we diagnosed his intramuscular hemangioma and treat it. When encountering similar cases in the clinical setting, the clinician must examine the patient carefully and have the diagnosis of hemangioma in mind. Probably the first modality that clinicians will use will be sonography because of its low cost, availability, and availability that may be able to provide some clues. However, MRI is the most important imaging modality. Based on clinical signs and symptoms, plain radiographs, and MRI results from definitive diagnosis are confirmed. After a definitive diagnosis, most authors suggested resection surgery the tumor completely excised. In the present case, orthopedics excised the whole tumor and resected exactly the tumor entity. Several studies showed that intramuscular hemangiomas can occur in various sites within the muscles of the trunk and the upper and lower limbs, mostly in the thigh muscles (9-11). 45% of hemangiomas occur in the lower extremities, 27% in the upper extremities and others are in the head and neck region and trunk. Because intramuscular hemangiomas are rare vascular

tumors and are not usually suspected based on clinical findings, they are of interest to surgeons as a cause of diagnostic problems. (12) A case report in Taiwan in 2017 reported an intramuscular hemangioma was misdiagnosed as an osteoid osteoma and after resection, the diagnosis became obvious(13). In the present case, the intramuscular hemangioma occurred in the buttock and had no symptoms except a red growing area. Intramuscular hemangiomas rarely occurred and they are easy to misdiagnose because they often have no diagnostic or characteristic symptoms. (14)

Figure4. After excision of the mass-like lesion



In this case report, the patient was initially undiagnosed and left for 8 years without definitive treatment. After the diagnosis, he underwent primary surgery, which was not performed due to the hypervascularity of the lesion, and was referred to an international radiologist. Due to the small obstructing particle, the patients had no symptoms and embolization was completed and the blood supply to the lesion was cut off. About 6 months after the surgery, the pain and limitation of movement have disappeared and the patient is undergone follow-up.

List of abbreviations:

MRI: magnetic resonance imaging

IMA: inferior mesenteric artery
AVF: arteriovenous fistula
AVM: arteriovenous malformation

Declarations:

- Ethics approval and consent to participate

Not applicable

- Consent for publication

Not applicable

- Availability of data and material

Not applicable

- Competing interests

"The authors declare that they have no competing interests”

- Funding

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- Authors' contributions

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- Acknowledgments

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