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Case Report

Venous malformation of the foot: Spontaneous regression postpartum on MRI ☆,☆☆

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ABSTRACT

Venous malformations (VMs) are present at birth, grow proportionally during childhood, and usually do not regress. We report the imaging appearance of a VM of the foot found during pregnancy, which regressed spontaneously postpartum. A 35-year-old, 8-month-pregnant woman presented with a 6-month history of painful swelling of the left foot. MRI demonstrated a well-defined, intricate-shaped mass measuring 38 × 36 × 28 mm between the muscles and tendons of the third, fourth, and fifth toes with subcutaneous extension. Dynamic CT taken a month after delivery revealed gradual enhancement of the lesion. Gray-scale ultrasonography (US) showed a heterogenic hypoechoic mass containing thrombi with venous waveforms on Doppler US. A second MRI obtained 15 months after delivery showed a remarkable reduction of the lesion size (16 × 20 × 15 mm). Symptomatic VMs found during pregnancy can be observed conservatively without treatment.

Introduction

Venous malformations (VMs) are one of the vascular anomalies classified by the International Society of the Study for Vascular Anomalies (ISSVA). They are the most common vascular malformations and often misdiagnosed as hemangioma or cavernous hemangioma [1]. VMs are present at birth and grow commensurately during childhood without involution. They often enlarge during puberty and pregnancy due to hormonal influence [2,3]. Two case reports described MRI findings of VMs in the orbita related to pregnancy and childbirth [4,5]. However, no report was found in the extremities. We report a case of a VM of the foot that onset during pregnancy and markedly decreased in size on MRI postpartum without treatment.

Case report

A 35-year-old, 8-month-pregnant woman presented with a 6-month history of progressive pain and swelling in the left foot between the fourth and fifth toes. The patient had no

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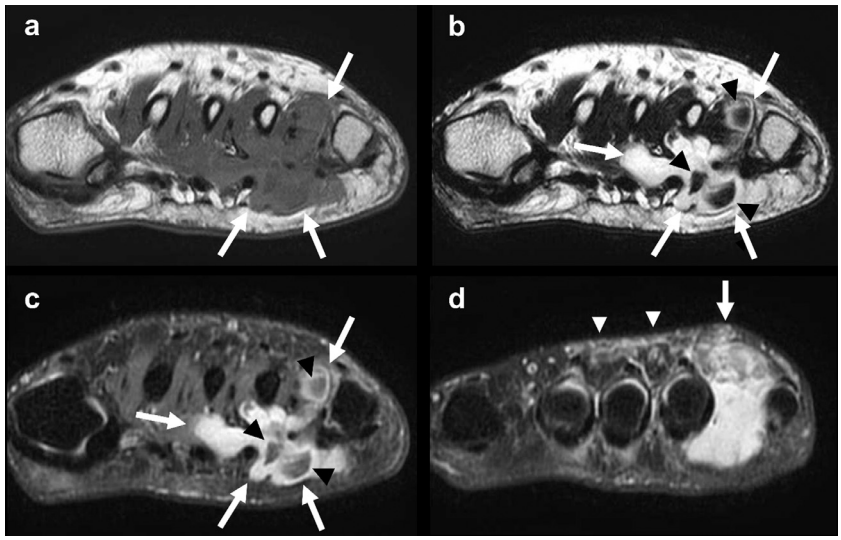

Fig. 1 – A 35-year-old, 8-month-pregnant woman with a VM in the left foot. Coronal MRI shows a lobulated mass in the plantar aspect of the third to fifth metatarsal bones (a-c, arrows), extending dorsally between the fourth and fifth metatarsal bones to the subcutaneous tissue (d, arrow). It measured 38 × 36 × 28 mm in size. The mass was isointense to the muscles on T1-weighted image (a) and hyperintense on T2-weighted image (b). It contained amorphous hypointense areas on T2-weighted and STIR image (b, c, arrowheads). Note the hyperintense area in the dorsal subcutaneous tissue indicating edema (d, arrowheads).

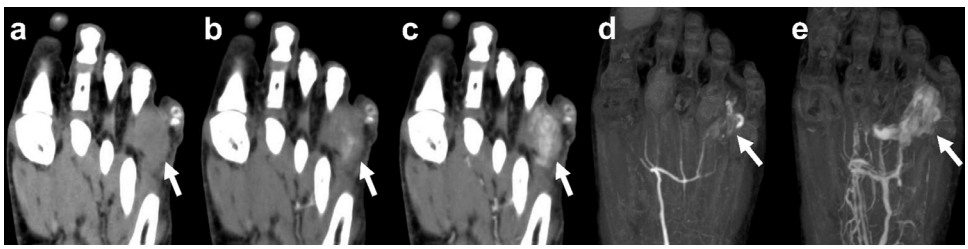

Fig. 2 – Noncontrast CT taken a month after delivery shows no calcification (a, arrow). Dynamic CT and maximum intensity projection images reveal gradual, heterogeneous enhancement of the lesion (b-e, arrow).

traumatic episode, past medical history, or medication. The patient's blood tests were normal. MRI demonstrated a well-defined intricate-shaped mass measuring 38 × 36 × 28 mm between the interosseous muscles and tendons of the third, fourth, and fifth toes with subcutaneous extension. The mass had intermediate signal intensity on T1-weighted images and high signal intensity on T2-weighted and short TI inversion recovery (STIR) images. It contained amorphous hypointense areas on T2-weighted and STIR images. There was no abnormality in the phalangeal or metatarsal bones adjacent to the mass. STIR images showed soft-tissue edema in the dorsum of the foot (Fig. 1). A complicated cyst was suspected and punctured a month after delivery. Fine needle aspiration revealed pale blood, indicating a vascular lesion. Noncontrast CT obtained a month after delivery showed no calcification. Dynamic CT revealed gradual enhancement suggestive of a slow-flow vascular malformation (Fig. 2). As her symptoms improved after delivery, she was observed without treatment. US performed 3 months after delivery showed a heterogenic mass. Color Doppler images showed the absence of flow in about half of the mass. Venous waveforms were observed in some vascular cavities. There was no arterial flow (Fig. 3). A second MRI obtained 15 months after delivery showed a remarkable reduction of the lesion size (16 × 20 × 15 mm) (Fig. 4). Her symptoms disappeared completely.

Discussion

VMs are slow-flow vascular malformations classified by the ISSVA. The ISSVA classification system divides vascular anomalies into 2 categories: vascular tumors and vascular malformations. The ISSVA classification is useful for understanding vascular anomalies, diagnosing them accurately, and effectively treating patients with vascular anomalies [3]. VMs are the most common vascular malformations, with an incidence of 1 to 2 per 10,000 births [6]. They usually appear in late childhood or early adulthood and sometimes show acute enlargement with pain due to hemorrhage, thrombosis, or hormonal changes [2,3]. VMs are categorized into 2 major

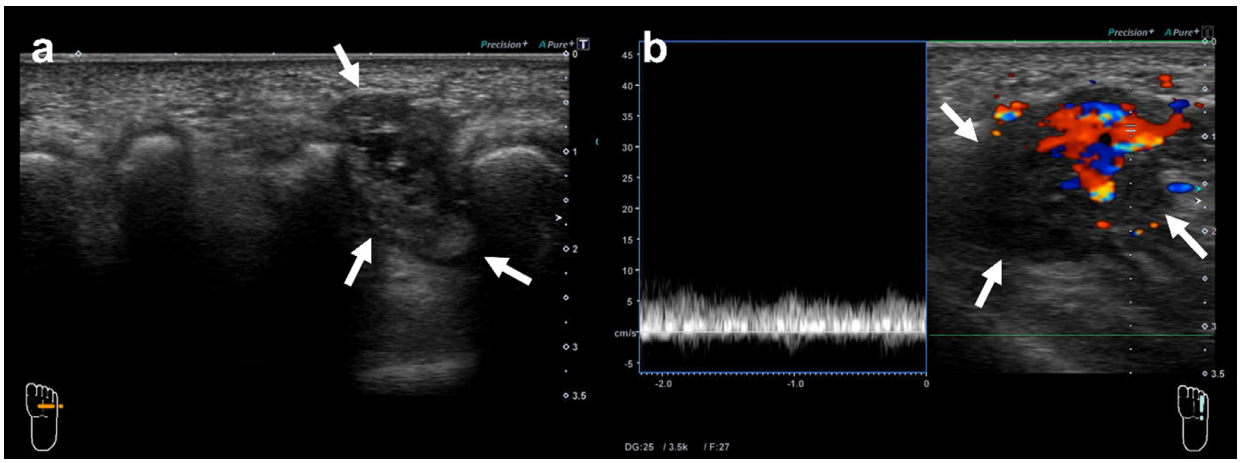

Fig. 3 – Gray-scale US performed 3 months after delivery shows a heterogeneous mass (a, arrows) between the fourth and fifth metatarsal bones extending to the subcutaneous tissue. Color Doppler US shows venous waveforms (b, left) and an absence of flow in about half of the mass (b, right, arrows).

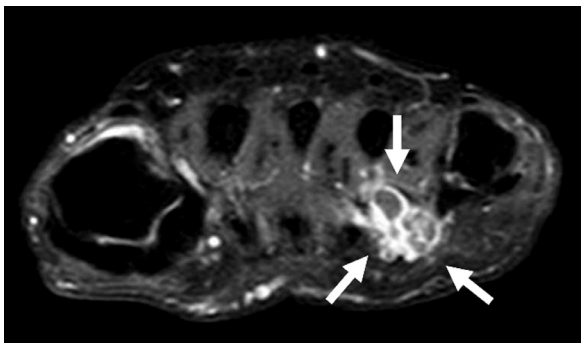

Fig. 4 – Coronal STIR image obtained 15 months after delivery at the same level as Fig. 1c shows a significant diminishing in the size of the VM, measuring 16 × 20 × 15 mm in size (arrows).

morphologic types: lobulated and varicose [7]. Most VMs are of the lobulated type.

MRI and ultrasonography (US) are the 2 major imaging modalities used to make a differential diagnosis of vascular anomalies as they provide information about blood flow and morphologic characteristics. On MRI, lobulated type VM usually present as septated and lobulated masses without mass effect, and have intermediate to low signal intensity on T1-weighted images and high signal intensity on T2-weighted and STIR images. Occasionally, T1-weighted images show high signal intensity areas in the fat-containing lesion [8]. Dynamic perfusion images demonstrate no enhancement in the arterial phase and gradual enhancement in the delayed phase. Fluid-fluid levels may be seen, indicating a hemorrhage or high protein content [9]. In the case of thrombosis or hemorrhage, heterogeneous signal intensity can be observed on T1-weighted images [8] and hypointense on T2-weighted images [2]. In our case, the lesion contained amorphous hypointense areas on T2-weighted images suggesting thrombi.

On gray-scale US, VMs appear to have hypoechoic or heterogeneous echotexture with well-circumscribed, sponge-like vascular spaces, or poorly marginated collections of veins [10,11]. Color Doppler US reveals venous waveforms or absence of flow because of sluggish vascular channels or thrombosis [3,10]. Although arterial flow can be detected in the septa or surrounding tissue, these represent small capillaries or supplying arterioles and do not suggest a fast-flow lesion [3]. In our case, there was no flow in about half of the mass, suggesting thrombi.

CT is more sensitive than conventional radiography for the detection of calcifications, which means phleboliths. CT angiography can be used for demonstrating detailed vascular anatomy in fast-flow lesions. However, it is not generally performed to diagnose slow-flow vascular anomalies. Repeat CT examinations are not suitable for young patients due to its high cost and the inherent risks of ionizing radiation [3]. In our case, dynamic CT was performed to detect vascularity after delivery because the lesion was not initially recognized as a VM.

There have been few reports of the spontaneous regression of VMs confirmed by MRI [4,5]. One report is of a 35-year-old woman who complained of pain behind her left eye, 1 month after delivery, and the other is of a 27-year-old pregnant woman with proptosis in her right eye. MRI demonstrated a mass in their orbita, which regressed completely without treatment. The authors suspected their masses as VMs. To our knowledge, there are no reports of a reduction of VM in an extremity associated with pregnancy demonstrated with MRI. In our case, MRI obtained 15 months after delivery showed a remarkable reduction in the size of the lesion. Although we did not know the imaging appearance of the lesion before pregnancy, we assume that the VM enlarged during pregnancy and spontaneously reduced to its original size postpartum.

VMs are characterized by stagnant blood flow, which can spontaneously thrombose [8]. Compression of the vena cava and iliac veins from the gravid uterus causes venous stasis of the lower extremities [12]. Hypercoagulability and vasodila-

tion of the veins also increase the likelihood of venous thrombosis during pregnancy [13]. The increase of coagulation factors leads to approximately double the coagulation activity. Furthermore, the elevated levels of estrogen induce vasodilation by activating endothelial nitric oxide synthase in the vascular endothelium [14]. As MRI and US revealed massive thrombosis within the lesion in our case, thrombosis induced by these mechanisms was considered to be the main cause of the enlargement.

The course of our patient may be useful for the future clinical management of VMs in pregnancy. Invasive intervention may not be needed for symptomatic VMs unless it worsens postpartum. VMs can be treated with conservative therapy, such as compression garments and anti-inflammatory medications. If the symptoms persist, sclerotherapy, embolization, and surgical resection can be considered. Multiple re-interventions are often needed [3]. In our case, the patient complained of progressive pain and swelling of the affected site. Accordingly, sclerotherapy might be indicated. However, sclerotherapy should be avoided during the first trimester, and after week 36, as there are limited data on its safety and efficacy [15]. Although the pathophysiology is different, varicose veins developed during pregnancy have a high probability of spontaneously improving postpartum. Therefore, it is recommended that clinicians wait for 6 to 12 months after delivery prior to pursuing sclerotherapy [16]. In some cases of VMs in the central nervous system, generally called cavernous hemangioma, postpartum spontaneous regression has been reported [17,18]. The authors suggest that urgent and invasive interventions are not always needed for VMs in pregnancy.

In conclusion, MRI demonstrated a case of VM of the foot that spontaneously regressed postpartum. Invasive intervention may not be needed for symptomatic VMs found during pregnancy unless they worsen postpartum.

Patient Consent Statement

Informed consent was obtained from the participant included in this case report.

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