Arteriovenous Malformation: An Unusual Reason for Foot Pain in Children

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The incidence of vascular anomalies is rare, and they are mainly localized in the head or upper extremity. We report the case of an 8 year-old boy with arteriovenous high-flow malformation of the foot. Presentations of diagnostic and therapeutic opportunities as well as post surgical clinical follow-up are included.

Key words: Arteriovenous malformation, angiography, foot, children.

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Vascular anomalies are classified into two main types: vascular malformations and benign vascular endothelium tumors. Vascular malformations (e.g. venous, arteriovenous and capillary malformations) are congenital abnormalities with manifestation during childhood or adolescence due to various stimuli (e.g. trauma, hormonal changes). They arise from defects in vascular tissue during embryonic development. Spontaneous regression of these anomalies is rare. Vascular tumours (e.g. haemangioma, Gorham-Stout-disease) are characterized by cellular hyperplasia and the expression of growth factors but they can also spontaneously become involuted.¹⁻⁶ “High” and “low-flow” malformations can be differentiated by their haemodynamic characteristics and the vascular architecture.^{7,8}

Case report

We report the case of an 8 year-old boy who presented in 2008 with a one-year history of progressive left foot pain and local swelling without previous trauma. Weight-bearing activities exacerbated the pain.

On clinical examination, a plantar soft tissue swelling with local paraesthesia and hypersensitivity was seen. The left foot was held in a relieving pes equinus posture.

Magnetic resonance imaging (MRI) of the foot was performed. The swelling showed up as a vascular malformation with high uptake of contrast medium which stretched out intrafascially, intramuscularly and subcutaneously from the metacarpophalangeal joints to the tarsal bone. (Figs. 1 and 2)

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Figure 1 Sagittal MRI of the left foot with hypervascularized tumor on T2 (arrow).

Figure 2 MR image of the left foot with hypervascularized tumor on T1.

An open biopsy was performed and histological examination confirmed the radiological diagnosis. To minimize the trauma of treatment for the boy, selective percutaneous as well as endovascular transarterial embolisation of the malformation were discussed. After percutaneous puncture and illustration of the malformation by injection of contrast medium, a venous lacuna with a varicose vein of plantar venous arc was seen. (Fig. 3)

Figure 3 Percutaneous puncture and illustration of the malformation by injection of contrast medium.

Additionally in projection of the third toe a high-flow arteriovenous malformation was depicted supplied in an en-passant fashion by an interdigital artery and arteries to the back of the foot. (Fig. 4) Due to these arterial collaterals a percutaneous embolisation was not performed because of the risk of necrosis of the toe.

During the course of further treatment planning MR angiography was performed and revealed a hypertrophy of the posterior tibial artery and of the lateral part of the plantar artery. The distal part of Ramus profundus of A. plantaris medialis was the focus of the malformation after its passage through the abductor hallucis.

Expected circulatory disturbance of the third toe and the probability of an incomplete occlusion of the malformation using transarterial embolisation limited the therapeutic options.

Figure 4 Percutaneous puncture and illustration of the malformation by injection of contrast medium.

Finally, marginal surgical resection of the malformation was performed. Histological examination confirmed the diagnosis of arteriovenous malformation with a central thrombus.

Only eight weeks after surgery, the boy could bear his full weight on his foot and the local pain as well the swelling were significantly reduced. (Fig. 5) At the one year follow-up the boy had returned to sporting activity. No signs of local recurrence were evident.

Discussion

Vascular anomalies occur with an incidence of 1-10/100000. They are apparent in 1 – 2.6% of neonates.⁹ Cephalic localizations are most common, followed by the trunk, lower and upper extremities. In rare cases vascular malformations may be part of a syndrome complex like Klippel-Trenaunay-Weber and Sturge-Weber syndromes or genetic disorders, so when evaluating these abnormalities a family history should be performed.^{10,11,12}

Figure 5 Clinical control six months after surgery.

The presented case describes a high-flow arteriovenous malformation of the foot. This localization appears to be rare with only few reports in literature.

Whereas a third of vascular tumors (e.g. haemangioma) are visible at birth, the remaining 70% appears in the first years of childhood. They are characterized by a rapid growth in the first years but can also spontaneously become involuted.

In contrast, congenital malformations increase in size as the child grows.¹³ According to their pathophysiology and vascular architecture low flow and high flow malformations are distinguished. Low flow lesions comprise capillary, lymphatic and venous malformations. High flow arteriovenous malformations are characterized by a high blood flow in a direct connection between arteries and veins without capillaries. The *Schobinger* staging system of arteriovenous malformations includes four grades of severity: dormancy, expansion, destruction and decompensation.¹⁴ Clinical manifestation may occur during adolescence and due to various stimuli (e.g. trauma, hormonal changes).

In the presented case, the cause of expansion and clinical manifestation could not be determined.

Depending on the localization, arteriovenous malformations present with different symptoms: local pain, swelling, skin ulceration, length discrepancy of extremities or severe bleeding with neurological complications.^{13,15,16} After clinical examination further diagnostic investigations are recommended. Useful non-invasive methods to define the extent of the lesion are ultrasonography and duplexsonography. Additional non-invasive imaging modalities are magnetic resonance imaging and MR angiography including dynamic sequences with high temporal resolution. Transarterial catheter angiography is required in order to visualize the architecture of the vascular malformation and the arterial supply and venous drainage of the surrounding structures prior to treatment.^{7,17,18,19,20} Depending to the extension of the malformation and the associated involvement of surrounding tissue, minimally invasive interventions or surgical resection may be necessary.

This report describes an unusual localization of an arteriovenous malformation. Nevertheless, the possibility of its existence should be considered in cases with the symptoms presented here. To facilitate rapidly diagnosis and to avoid unnecessary treatment, interdisciplinary therapy is recommended.

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