



Case Report

Intraosseous anomalous drainage: a rare case of pretibial varicose vein[☆]



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ABSTRACT

Valve failure with reflux and post-thrombotic syndrome are the factors most commonly correlated with varicose disease. Other rare etiologies can be put forward when these two main causes are ruled out. We report a case in which a young man presented chronic pain in the left tibia, varicose veins in the lower limbs and frequent occurrences of erysipelas. During investigation of the etiology of the varicose veins, radiographs and magnetic resonance imaging of the left leg were requested. These showed images suggestive of an osteolytic lesion in the tibia, but led us to the diagnosis of an intraosseous vein with anomalous drainage. This was confirmed through vascular examinations comprising Doppler venous flow measurement and phlebography. Recognition of this rare intraosseous anomaly is fundamental for proper patient management, but an intraosseous surgical approach is unnecessary.

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Drenagem anômala intraóssea: um caso raro de varizes pré-tibiais

RESUMO

Incompetência valvar com refluxo e síndrome pós-trombótica são os fatores mais comumente relacionados à doença varicosa. Outras raras etiologias podem ser levantadas quando essas duas principais causas são descartadas. Relatamos o caso de um homem jovem com dor crônica na tíbia esquerda, varizes em membros inferiores e erisipela de repetição. Durante investigação da etiologia das varizes foram solicitadas radiografias e ressonância da perna esquerda, que evidenciaram imagens sugestivas de uma lesão osteolítica na tíbia, mas que nos levaram ao diagnóstico de veia intraóssea com drenagem anômala, confirmado pelos exames vasculares de dopplerfluxometria venosa e flebografia. O reconhecimento

Palavras-chave:

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[☆] Study conducted at the Universidade Federal de Goiás, Faculdade de Medicina, Hospital das Clínicas, Departamento de Ortopedia e Traumatologia, Goiânia, GO, Brazil.

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dessa rara anomalia intraóssea é fundamental para o manejo adequado do paciente, não necessita de abordagem cirúrgica intraóssea.

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Introduction

Varicose veins in the lower limbs are present in 10–40% of individuals aged 30–70 years. They are associated with conditions that impair venous return, such as pregnancy, physical inactivity, and clinical pictures of venous insufficiency. Their main pathophysiological substrate is the increased pressure in the peripheral venous system. Congenital abnormalities of the venous endothelial tissue and smooth muscle cells can result in the dilation of the venous wall, with secondary valvular insufficiency. However, there is evidence to support acquired valvular insufficiency.^{1,2} There are several causes of varicose veins in the lower limbs. These include venous insufficiency in the saphenofemoral or saphenopopliteal junction or in the usual perforating veins. Varicose veins are classified, according to their causes, as usual and unusual. Among the unusual causes, the following have been described: vulvoperineal varicosity, intraosseous perforating vein incompetence, round ligament varicosity, persistent sciatic vein incompetence, Klippel-Trenaunay syndrome, and portosystemic collateral pathways. Physicians should be familiar with the full range of primary causes of varicose veins in the lower limbs and their radiological manifestations; they should also recognize the complementary role of venography in their assessment.^{3,4}

The recognition of this rare anomaly in intraosseous venous drainage is critical to proper patient management. This study aimed to report a rare case of intraosseous venous drainage anomaly with pre-tibial varices in a patient who presented difficult-to-diagnose chronic pain and lower limb edema that coursed with a decrease in the patient's quality of life.

Case report

Male patient, 23 years old, obese, resident of a garbage dump in Goiânia, GO, Brazil, was attended to at the Orthopedics emergency room of the Hospital das Clínicas. He presented with chronic pain at the lower limbs, more severe on the left, along with pain on palpation of the middle-third of the tibia, associated with recurrent erysipelas, but with no signs of inflammation at that moment, with asymmetrical chronic edema at the left lower limb (LLL), suggestive of chronic venous insufficiency and truncular varicose veins in the LLL (Fig. 1). He had been attended at various services, without receiving a diagnosis or specific treatment, which limited his quality of life. Radiographs and computed tomography (CT) of the LLL showed a radiopaque lesion in the middle-third of the tibial shaft, with signs of adjacent bone sclerosis suggestive of an osteolytic lesion (Fig. 1).

Magnetic resonance imaging (MRI) disclosed an increased intraosseous vascularization at the middle-third of the tibia, with anomalous drainage and pre-tibial varices (Fig. 2). Venous Doppler ultrasound showed varicose veins throughout the affected limb. The patient was referred to the vascular surgery department; surgical treatment was indicated for correction of venous drainage, with resection of varicose veins and pain relief. An intraosseous surgical approach was not necessary.

Discussion

Boutin et al.¹ reported the first case of intraosseous varices in 1997; since then, 13 cases have been reported. All cases presented the same characteristics, with patients between 23 and 75 years and unilateral alterations with signs of venous insufficiency. The present patient falls into this age group, but was attended at several services without a definitive diagnosis. This is due to the low prevalence of this injury, which makes diagnosis difficult.

All reported patients were adults between 23 and 75 years, and most had pain in the lower leg. Only one patient described had intraosseous venous drainage anomaly at the fibula.³ All other reported patients had intraosseous venous drainage anomaly at the tibia.¹⁻⁷

From a radiological standpoint, the most important aspect to determine the primary cause of venous insufficiency is the presence of veins with valvular incompetence and the general characteristics of the varicose veins. Venous Doppler ultrasound has the ability to demonstrate venous dilation and valvular incompetence. In varicose veins with complex morphology and/or unusual manifestations, MRI, CT, or venography can be used for further evaluation. CT venography, as well as conventional venography, can be used to demonstrate the presence of an enlarged intraosseous vein, which is connected to the varicose veins.^{3,4}

However, MRI may be the preferred method to confirm intraosseous venous drainage anomaly, as it does not use ionizing radiation, is less invasive, and provides superior soft tissue contrast. Furthermore, MRI can also be used to rule out other vascular anomalies that could represent differential diagnoses, such as arteriovenous malformation (AVM), venous malformations, and hemangiomas.^{2,3} Notably, it is possible to differentiate varicose veins from intraosseous venous drainage anomaly through clinical aspects, because venous malformation typically occurs in childhood or early adulthood and increases proportionally with the child's age, without regression.²

The cause and clinical significance of varicose veins with intraosseous drainage anomaly are not yet fully understood. One hypothesis is that an auxiliary drainage, via intraosseous, may be favored in a picture of venous insufficiency (e.g., in



Fig. 1 – (A) clinical aspect of the left lower limb showing large and tortuous varicose veins; (B) radiographic appearance; (C) sagittal CT cut showing a radiotransparent and osteolytic image, with signs of adjacent sclerosis.

cases of deep venous thrombosis).⁵ Kwee et al.⁴ reported the case of a patient with a picture of unilateral lower limb varices originated from a venous communication between the peripheral venous system member and an intraosseous vein, causing chronic pain of the affected limb associated with varicose veins and signs of chronic venous insufficiency.

The diagnosis of this injury can be achieved through the combination of clinical assessment and imaging exams. Radiography may show signs of spinal sclerosis and cortical defects throughout the communicating vessel pathway. A CT can evidence this communication; however, MRI is the most sensitive and preferred method, allowing for a more precise

diagnosis of the communication site and its impact. Venography is an auxiliary method that allows for a detailed study of the venous system. Ultrasound would be the standard method for vascular evaluation, but the waves are reflected by high bone density, restricting its use to Doppler velocimetry of the limb, which discloses the signs of venous insufficiency that this condition can cause.^{3,4}

Intraosseous venous drainage leads to an increase in pressure in the peripheral venous system, which consequently culminates in venous valve impairment and signs of insufficiency. As a result, patients present massive varices and an increased risk for thromboembolic events. Therefore,



Fig. 2 – Magnetic resonance: (A) frontal cut in T1 showing prominent vessel, hypointense on the topography of the middle-third of the tibia; (B) perforating vein that enters the tibia, T2-hyperintense, sagittal cut; (C) numerous varicose veins in the adjacent soft tissues, T2-hyperintense.

surgical treatment of varicose veins should be established by the angiology team; an intraosseous approach is not necessary.^{6,7}

Conflict of interest

The authors declare no conflicts of interest.

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