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Symptomatic tibial vein aneurysm – a diagnostic challenge

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Key message

Primary venous aneurysms are very rare vascular malformations causing serious thrombo-embolic complications. Ultrasound is the clue to a correct diagnosis.

Introduction

Primary venous aneurysms are very rare vascular malformations that can result in life-threatening thrombo-embolic complications. By definition, a venous aneurysm is a solitary area of venous dilatation that communicates with a main venous structure by a single channel and that has no association with an arteriovenous communication or a pseudoaneurysm [6]. Its prevalence is unknown, as there are no screening studies in unselected patients with otherwise healthy veins. Mostly, venous aneurysms are found incidentally on imaging. With the increasing use of duplex scanning, more cases of venous abnormalities are being diagnosed [16]. The most commonly reported aneurysm of the deep vein system is that of the popliteal vein and was first described by May and Nissel in 1968 [9]. More than 150 cases of popliteal venous aneurysms have subsequently been reported in the literature [1, 15], many of which presented with pulmonary embolism or other thrombo-embolic complications. In contrast, an infrapopliteal deep venous aneurysm has only been reported once, i.e. in a recent case report by Gabrielli et al [5] describing a thrombosed posterior tibial vein aneurysm.

We report a second case of a posterior tibial vein aneurysm in a patient presenting with pulmonary embolism, and in which we encountered an interesting diagnostic challenge.

Case report

A 54-year-old female presented with symptoms of dyspnea during exercise. She had no specific medical history and was not on any medication. Her clinical examination was unremarkable, except for a palpable mass in her left calf. This mass lesion was associated with intermittent nagging pain, which had started 3 months before presentation. Blood tests showed a highly increased D-dimer level. A pulmonary CT-angiography showed multiple bilateral segmental and subsegmental pulmonary emboli. Anticoagulation therapy was initiated with low-molecular-weight heparin (LMWH) followed by a vitamin K antagonist (VKA) adjusted according to the International Normalized Ratio (INR). A thrombophilia screening was negative. The mass in the left calf was subsequently investigated with duplex-US (not shown), which revealed a hypo-echogenic soft-tissue mass in the left soleus muscle with a diameter of 29 × 23 mm.

Triphasic contrast-enhanced CT (not shown) showed the slightly hypodense intramuscular soft-tissue mass with discrete peripheral contrast enhancement. Because these findings were still non-specific, an MRI was performed that confirmed the ovoid soft-tissue mass in the soleus muscle, which was located in close contact with the neurovascular bundle. The structure had a low signal intensity in the periphery, a brighter signal intensity in the central region on T1-weighted images and a bright signal intensity in the central region on T2-weighted images. After the administration of gadolinium there was circumferential contrast enhancement on T1-weighted images (Figure 1).

Due to its localization, aspect and symptoms, the mass initially was diagnosed as a soft-tissue tumor, potentially an ancient (cystic) schwannoma. The patient was then referred to our institution for a second opinion as to the diagnosis and treatment. The duplex-US of the left calf was repeated 3 months after the initial diagnosis and it confirmed a 4 × 2,3 cm saccular mass in the left soleus muscle. The mass, however, presented with mixed echogenicity and was continuous with a branch of the posterior tibial vein. Centrally, venous flow could be demonstrated (Figure 2), suggesting that the mass was a true venous aneurysm which was partially recanalised during the interval of 3 months between the 2 duplex-US examinations (Figure 3).

Two weeks later the aneurysm was surgically resected by tangential aneurysmectomy with lateral venorrhaphy. The histopathological examination confirmed the diagnosis of the venous aneurysm with preserved, normal architecture of the tunica intima, media and adventitia. Anticoagulation (LMWH with switch to VKA) was restarted postoperatively, aiming at another 3 months of anticoagulation. However, a postoperative intramuscular bleeding occurred at the resection site 1 week after surgery, requiring urgent hematoma drainage, despite the fact that the patient was wearing compression stockings. Thereafter, the patient received a prophylactic dose of LMWH for another 2 months, after which all anticoagulation was stopped. Further follow-up, i.e. until 6 months after cessation of prophylactic LMWH, remained un-

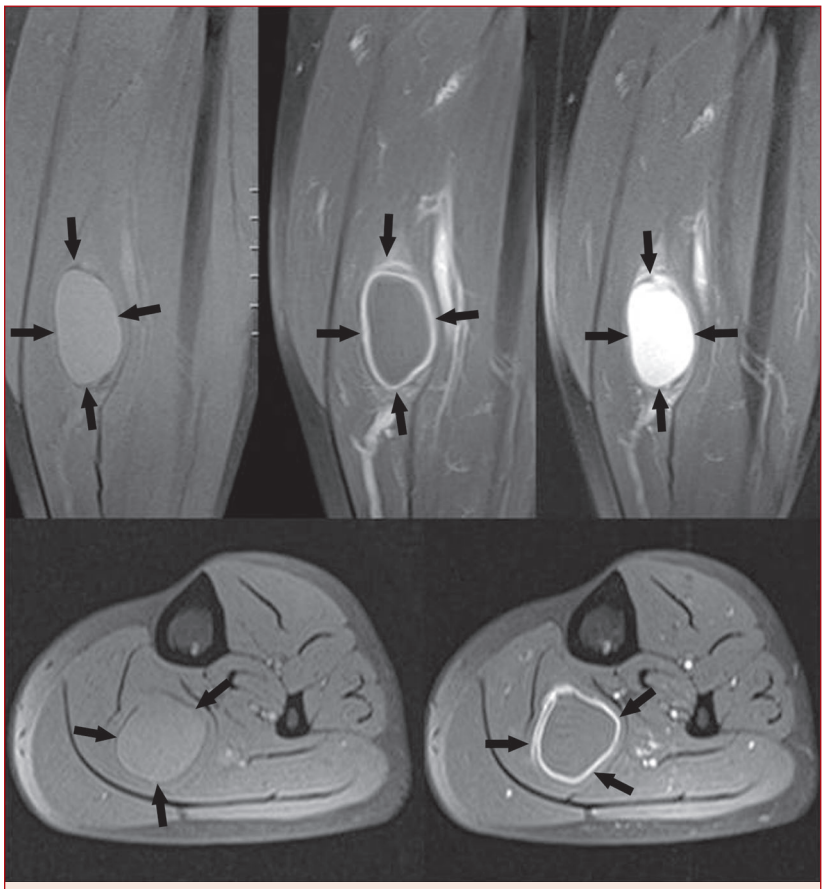

Figure 1: A giant thrombosed venous aneurysm in the calf as viewed by sagittal (a, b, c) and axial (d, e) MRI sections. (A, D) T1-weighted and fat-saturated images before contrast injection show an ovoid mass slightly hyperintense to muscle with a small surrounding hypointense rim (arrows) (B, E). T1-weighted and fat saturated images after contrast injection demonstrate marked circumferential contrast enhancement (arrows) (C) on T2-weighted and fat saturated images the lesion shows homogeneous high signal intensity.

complicated, including completely normal duplex-US of the operated vein.

Discussion

We present a case of a tibial vein aneurysm in a patient presenting with pulmonary embolism. To our knowledge, this is only the second case of a primary tibial vein aneurysm reported in the literature. The first case was recently described by Gabrielli et al [5] in a patient with recurrent pulmonary emboli despite therapeutic anticoagulation. Other reported localisations in the lower legs are the external iliac vein (1), the common femoral vein and the popliteal vein. In 1996, Otto et al already reported a posttraumatic venous aneurysm of the posterior tibial vein secondary to a stab injury that resulted in A-V fistula [14].

The etiology of venous aneurysms is not clear, but congenital factors (congenital vein weakness), inflammation, trauma, hemodynamic and localized degenerative changes have been suggested [1, 5, 6, 8, 10].

Most venous aneurysms are asymptomatic, but those located in the popliteal vein can give serious complications such as proximal deep vein thrombosis and pulmonary embolism[18, 19]. When located in the lower limb, local symptoms such as a palpable mass, distal deep vein thrombosis and signs of venous insufficiency may occur. The initial and first-line diagnostic modality for extremity venous aneurysms is duplex imaging since it is rapidly available, inexpensive and implies no radiation exposure. Duplex imaging can measure the size of the aneurysm and detect the venous flow. Also, the color flow function allows to differentiate between vascular and non-vascular pathology, such as a Baker's cyst. Further imaging with CT and MRI can give additional anatomical information and typical enhancement.

When there is incomplete thrombosis of the venous aneurysm, an accurate diagnosis can easily be made with the demonstration of venous flow in the lesion on US or of contrast enhancement in continuity with the popliteal vein in the venous phase. In our case, and in 3 previous cases of popliteal vein aneurysm, complete thrombosis of the aneurysm was misleading for diagnosis both on US and CT and MRI, and led to a suspected diagnosis of a soft-tissue mass or intramuscular hematoma [13, 15, 17]. The giant thrombosed venous aneurysm showed characteristics that represented potential pitfalls resulting in a false diagnosis of the lesion as a peripheral nerve sheath tumor, type ancient (cystic) schwannoma or an intramuscular spontaneous hematoma [11, 13]. Peripheral nerve sheath tumors present on T1-weighted images as a low signal intensity mass and are very hyperintense on T2-weighted images [11]. Sometimes, a neurofibroma and schwannoma can present a target sign on the T2-weight-

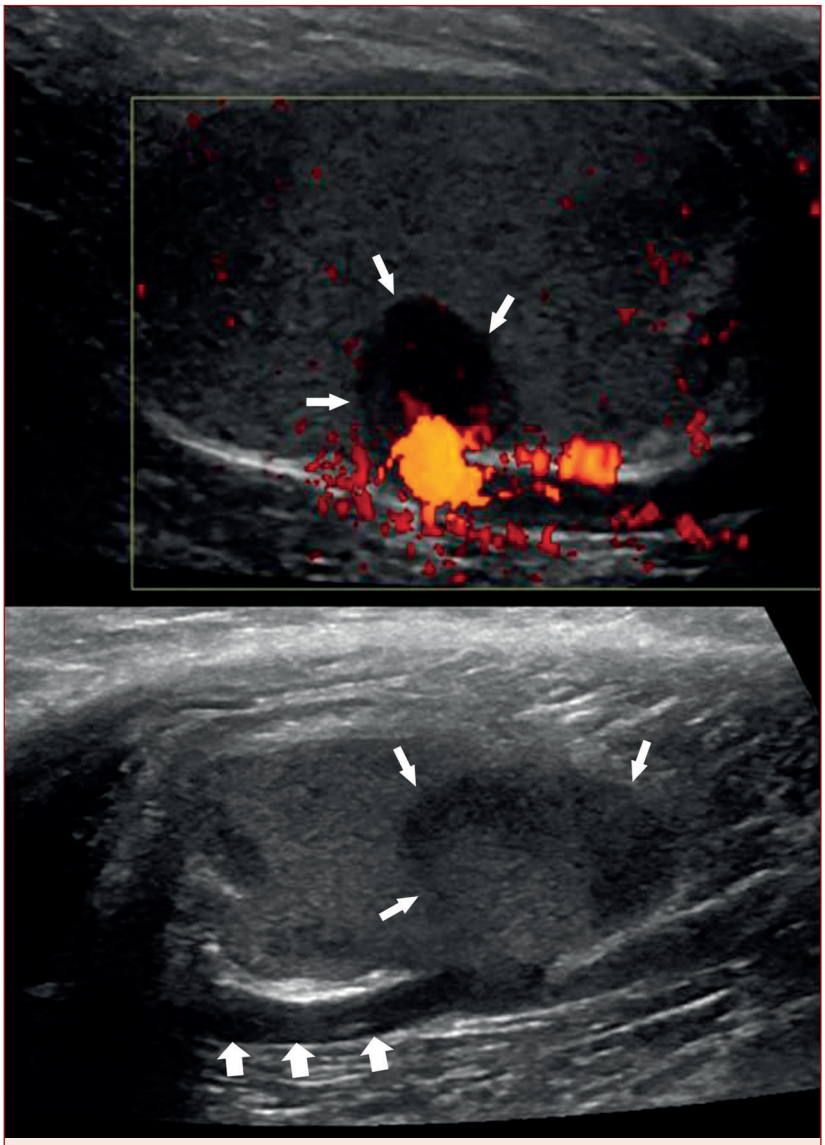

Figure 2: Repeat duplex scanning 3 months after the onset of symptoms. (a) color-coded duplex ultrasound; (b) gray-scale ultrasound. These images show the venous aneurysm with central venous flow (thin arrows), surrounded by thrombus, and the continuity with the left posterior tibial vein (thick arrows).

ed images using a high signal intensity (myxomatous tissue) peripherally and a low to intermediate signal centrally (fibrocollagenous tissue) [11, 13]. The contrast enhancement of neurogenic tumors can be quite variable.

An intramuscular hematoma most often presents on T2-weighted images with high signal intensity. Also on T1-weighted images, it can present with high signal intensity depending on the age of the bleeding. An intramuscular hematoma can have the same circumferential contrast enhancement on T1-weighted images due to the surrounding inflammation as in this case. The T2*-weighted gradient-echo images may also be important in demonstrating magnetic susceptibility effects related to blood product degeneration [13].

In this case, diagnostic certainty has been obtained 3 months later when the US was repeated, due to the partial recanalisation, potentially as a result of the anticoagulant therapy. Finally, the continuity of the mass with a branch of the posterior tibial vein made the diagnosis of a potentially recanalised tibial vein aneurysm very likely.

In managing venous aneurysms there is a difference between symptomatic aneurysms (i.e. presenting with thrombo-embolic complications) and asymptomatic aneurysms. The treatment of symptomatic venous aneurysms should primarily be surgical rather than medical. In 23 patients that were anticoagulated as first-line treatment, Nasr et al [12] and Falkowski et al [3] described a risk of recurrent pulmonary embolism of 43 % and 35 %, respectively, in popliteal vein aneurysms despite therapeutic levels of anticoagulation with medical therapy alone. Surgical options include tangential aneurysmectomy with lateral venorrhaphy, resection with end-to-end anastomosis, resection with interposition graft, or ligation of the proximal and distal vein [17]. In many cases of aneurysms of the popliteal vein/deep venous system, it is possible to resect the aneurysm and perform a lateral venorrhaphy. When the defect becomes too large, reconstruction is needed, either in the form of a venous patch or venous bypass. Ligation or resection is not an attractive option as this may give long-term problems of deep vein insufficiency and swelling in the popliteal vein but it can be sufficient in distal veins without serious risk for later venous insufficiency [1]. When there is a contraindication to surgery and recurrent pulmonary embolism, a vena cava filter might be considered,

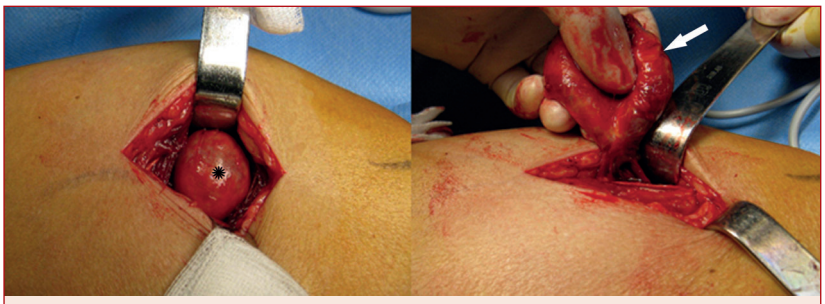

Figure 3: (a) Peroperative images show the saccular aneurysm (asterisk) that is (b) resected by tangential aneurysmectomy.

even though this is rather uncommon. Temporary placement of a vena cava filter to cover the perioperative time period seems to be redundant.

Pathological examination revealed normal aspect of all vein wall layers, which is in contradiction to other reports describing reduction of smooth muscle cells and increase in fibrous connective tissue (1), or destruction of the internal elastic lamina, loss of medial smooth muscle cells and fibrosis (5).

There is no consensus on postoperative management and follow-up, although, based on current limited information, a period of 3 months of anticoagulation after surgery was recommended [12]. Furthermore, regular follow-up with duplex scanning is advised due to the risk of aneurysm recurrence [4].

The optimal management in asymptomatic venous aneurysms is still debatable. Some authors suggest conservative management until symptoms occur, particularly in those aneurysms without thrombus within the sac [16]. On the other hand, a review of the literature showed that in 5 patients without intraluminal thrombus, there were 2 cases of pulmonary emboli despite therapeutic anticoagulation [12]. Therefore, in view of the potential complications, the high technical success rates and the low morbidity in case of surgical intervention, an early surgical treatment may be recommended in asymptomatic patients as well [15].

Conclusions

Primary venous aneurysms are very rare vascular malformations but can cause serious thrombo-embolic complications. A case of thrombosed tibial vein aneurysm is presented. Both CT and MRI were misleading for diagnosis. Repeat US, demonstrating partial recanalisation of the thrombosed aneurismal sac, was the clue to a correct diagnosis.

Conflicts of interest

There are no conflicts of interest existing.

Key words

Venous aneurysm, ultrasound, magnetic resonance

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