Popliteal Vein Aneurysm Associated with Varicose Veins, Hydrocele, and Multiple Congenital Osteomas: A Case Report and Review of the Literature

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INTRODUCTION

Popliteal vein aneurysms (PVAs) represent a rare form of venous aneurysms and necessitate prompt management because of their well-established risk of pulmonary embolism (PE) [1]. Once a massive PE occurs, the outcome can be fatal. Early suspicion and diagnosis of the disease are essential to prevent catastrophic complications. PVAs tend to be found in patients with severe thromboembolism without warning symptoms, such as leg pain or swelling [2]. Fortunately, in some cases, a PVA can be diagnosed in the absence of PE, on the basis of a high degree of suspicion by the vascular surgeon [3]. Herein, we report a rare case of PVA without PE, which was successfully treated with open surgical repair.

CASE

A 27-year-old man presented to our vascular clinic with chronic pain and multiple areas of swelling in his left lower limb. He had a long history of plastic surgeries of the face for the removal of congenital multiple osteomas. Physical examination revealed varicosities with an abnormal distribution. Duplex ultrasound showed a left PVA measuring 2.3 cm in diameter and 4 cm in length. Open surgical excision of the PVA with lateral venorrhaphy was performed through a medial approach. Prophylactic anticoagulation was performed postoperatively. In this case, the PVA was detected, with a high degree of suspicion by the clinician, before it caused fatal PE. The patient was successfully treated with aneurysm excision and lateral venorrhaphy.

Key Words: Popliteal vein aneurysm, Osteomas, Varicose veins, Lateral venorrhaphy
the PVA with lateral venorrhaphy was performed through a medial approach (Fig. 3). A 10-12-cm longitudinal skin incision was made on the medial aspect of the thigh along the anticipated anterior border of the sartorius muscle. The skin incision was advanced deeper through the subcutaneous tissue, exposing the adductor tendon anteriorly and the sartorius muscle posteriorly. The fascia between these two muscles was incised to enter the popliteal fossa. A self-retaining retractor was placed deep in the wound until the popliteal vein was exposed. Once the popliteal vein was exposed, the thin aneurysm sac could be easily differentiated from a grossly normal vein wall, and a vascular clamp was placed tangentially across the transition area. The clamp was then undersewn with running mattress stitches using 6-0 vascular sutures.

Rivaroxaban 10 mg (Xarelto, 10 mg once daily; Bayer, Leverkusen, Germany) was administered postoperatively for prophylactic anticoagulation. Three months later, DUS revealed deep vein thrombosis in the popliteal vein. Therapeutic anticoagulation was continued for 3 months thereafter.

DISCUSSION

PVAs are rare. In 2006, only 105 cases have been reported in the world literature [4]. By 2018, this number has increased to only 146 cases. Various etiologies associated with a popliteal venous pathology have been suggested, including congenital abnormality, trauma, localized degenerative changes, or inflammation in the form of varicose veins, venous aneurysm, or arteriovenous fistula.

Primary PVA represents a rare subset of venous aneu-

Fig. 1. (A, B) Multiple osteomas on both hands (blue arrows). (C) Radiographic image showing deformities in both hands with multiple osteomas (orange arrows).

Fig. 2. (A) Axial view of magnetic resonance venography showing a left popliteal vein aneurysm (PVA) with intraluminal thrombi (yellow arrow). (B) Coronal view of the left PVA (yellow arrow), showing superficial varicosities in the lateral thigh and calf (orange arrow) and a scrotal hydrocele (blue arrow). (C) Superficial varicosities (red arrows) and PVA with intramural thrombi (yellow arrows).
PVRs, with an estimated prevalence for asymptomatic PVAs of 0.1% to 0.2% among patients undergoing venous duplex imaging for various chronic venous symptoms [5]. PVAs can occur at any age, and they have been reported in patients aged 10-86 years. A female preponderance has been noted in three previous series, and the median ages at presentation were 51 years in men and 49 years in women [6].

The etiology of PVAs is unclear, although aneurysmal changes are known to result from increased hemodynamic pressure at the site of venous mural weakness, possibly caused by trauma, inflammation, congenital weakness, and degenerative changes [6]. In this case, the cause of the PVA was unclear, and the relationship between the PVA and the congenital multiple osteomas is unknown. To our knowledge, this is the first case of a combined presentation of PVA and osteomas ever reported in the English literature.

Aneurysmal dilatation may precede the formation of mural thrombi owing to turbulent venous flow. The presence of thrombi in PVAs may differ from the pathophysiology of deep vein thrombosis, which usually starts in the valve cusps or at injury sites.

PVAs tend to be found in patients with severe PE without warning symptoms such as leg pain and swelling [2]. Even small PVAs have been documented to be a source of emboli, precluding the stratification of thromboembolic risk according to aneurysmal size. Symptomatic PVA typically presents either acutely with PE or with a more chronic history of localized symptoms related to a popliteal mass or venous insufficiency. In some aneurysms, local pain originates from the PVA itself or from direct compression of neural structures due to aneurysmal dilatation.

In a case series, the potentially life-threatening complication of PE occurred in 43% of the patients [7]. The presence of an intra-aneurysmal thrombus is a clear risk factor for PE (69% patients had mural thrombi and 23% did not have thrombi) [8]. The characteristic microscopic features of PVA are fragmentation of elastic lamellae and fibrosis replacing the medial smooth muscle [9].

The size criterion for the treatment of PVA varied among different publications, with sizes two or three times larger than the normal vein size (5-7 mm) being recommended for treatment [10]. However, the best method for size measurement and the effect of the body position on the size measurement are not yet defined. The diagnosis can be made using ascending venography, computed tomography, magnetic resonance imaging, and DUS. We recommend magnetic resonance imaging as the best and most anatomic precise modality. However, the cost-effectiveness should also be evaluated. The treatment options for PVA are considered on a case-by-case basis. In patients with PE, surgery for PVA is the treatment of choice to prevent recurrence. Surgery is also recommended in cases of aneurysms with thrombus in the sac, as well as for saccular-type or large fusiform aneurysms because of their high risk for thromboembolism. However, if the aneurysm is fusiform in shape and smaller than 2 cm, close observation can be performed safely without complications [3]. Although no consensus has been reached about postoperative anticoagulation, prophylactic anticoagulation may be a reasonable option [3].

In conclusion, a PVA occurring with multiple osteomas was found, with a high degree of suspicion by the vascular surgeon, before it caused fatal PE. The patient was successfully treated with aneurysm excision and lateral venorrhaphy.

**CONFLICTS OF INTEREST**

The authors have nothing to disclose.

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