

Giant external-iliac-vein aneurysm secondary to traumatic arteriovenous fistula: a case report

Aneurisma venoso gigante de veia ilíaca externa secundário a fistula arteriovenosa traumática: relato de caso

Patrick Bastos Metzger¹, Heraldo Antonio Barbato²; Fernanda Maria Resegue Angelieri¹; Camila Baumann Beteli¹; Ana Claudia Gomes Petisco³; Jose Eduardo Martins Barbosa³; Mohamed Hassan Saleh³, Fábio Henrique Rossi², Nilo Mitsuru Izukawa⁴

Abstract

Venous abdominal aneurysms are rare entities, especially at the external iliac vein. We report the case of a young male patient who presented with a giant external-iliac-vein aneurysm secondary to an arteriovenous fistula acquired 20 years earlier, and treated successfully by conventional and endovascular methods in our service.

Keywords: aneurysm; iliac vein; arteriovenous fistula.

Resumo

Aneurismas venosos abdominais são raros. Os localizados nas veias ilíacas externas estão entre os mais infrequentes aneurismas venosos publicados na literatura. Relatamos o caso de um paciente jovem com aneurisma venoso gigante de veia ilíaca externa secundário a uma fistula arteriovenosa adquirida há 20 anos, tratado pelos métodos convencional e endovascular com sucesso.

Palavras-chaves: aneurisma; veia ilíaca; fistula arteriovenosa.

Introduction

Abdominal venous aneurysms (VA) are rare. Over the past 90 years, sporadic cases of aneurysms of the superficial and deep venous systems have been reported in the world literature¹. The external iliac vein is an infrequent site of those aneurysms². The first case, reported by Hurwitz and Gelabert¹ in 1989, was a thrombosed common and external-iliac-vein aneurysm.

The leading causes of iliac VA are arteriovenous fistulas, which are usually secondary to trauma (43%), venous out-flow obstruction (17%) and primary aneurysms (35%)^{3,4}. Symptoms of aneurysms result from the compression of pelvic structures, local thrombosis, thromboembolism and aneurysm rupture².

Unlike arterial aneurysms, few review articles on iliac VA can be found in the literature, so very little is known about their natural history and treatment^{1,5}.

The aim of this paper was to describe the management and repair of a large external-iliac-vein aneurysm, secondary to an acquired arteriovenous fistula (AVF).

Case report

A 48-year-old male patient, who had a history of gunshot wound on the middle third of the right thigh 20 years earlier, came to the emergency room with a two-month history of progressive right lower limb edema associated with pain and presenting a pulsatile mass at the right iliac fossa. Physical examination at admission showed marked

Study carried out at Instituto Dante Pazzanese de Cardiologia – São Paulo (SP), Brazil.

¹Residents of Endovascular surgery at Instituto Dante Pazzanese de Cardiologia – São Paulo (SP), Brazil.

²Vascular surgeons at Instituto Dante Pazzanese de Cardiologia – São Paulo (SP), Brazil.

³Cardiologists at the service of Vascular Ultrasound at Instituto Dante Pazzanese de Cardiologia – São Paulo (SP), Brazil.

⁴Head of the Service of Vascular Surgery at Instituto Dante Pazzanese de Cardiologia – São Paulo (SP), Brazil.

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swelling and erythema of the right lower limb, associated with a pulsatile mass in the lower abdomen. The femoral pulse was normal; no other pulses were palpable in the right lower limb.

Digital subtraction angiography (DSA) of the affected limb showed a high-flow AVF between the superficial femoral artery and the femoral vein within the Hunter's canal, and a spherical pelvic formation with early venous phase filling suggesting right external-iliac-vein aneurysm (Figure 1).

The AVF was treated by deploying two Hemobahn® 13 x 100 mm endografts (W.L. Gore & Assoc, Flstaff, Ariz) in the distal superficial femoral artery. Postoperative control angiogram showed an important decrease in venous filling (Figure 2), so the patient was discharged on the tenth postoperative day, with improvement of the swelling and of the pulsatile abdominal mass.

Two years later, the patient was readmitted to the hospital with marked swelling and inflammatory signs in the right lower limb associated with a painful large

abdominal pulsatile mass that had developed three days earlier. On physical examination, the right lower limb was swollen and presented purulent discharge from the skin and subcutaneous tissue. The presumptive diagnosis was phlegmasia cerulea dolens associated with local cellulitis (Figure 3). The patient was started on empirical antibiotic therapy (imipenem and vancomycin) after the cultures of the skin discharge were sent to analysis. Color-Doppler ultrasonography of the right lower limb showed right iliac vein aneurysm (17 x 11 cm) without intraluminal thrombi, associated with an AVF of the superficial femoral vessels. Abdominal CT angiography confirmed the finding of a giant right-external-iliac vein aneurysm measuring 17 x 15 x 12 cm and with a volume of approximately 3,000 mL. CT angiography of the right lower limb also showed persistence of the AVF immediately distal to the end of the endograft implanted two years earlier. Early filling of the right femoral vein suggested a high-flow AVF (Figure 4). Cultures yielded *Staphylococcus aureus*, sensitive to the antibiotics. After ten days of therapy, upon clinical and laboratory improvement of the infection, a new endovascular repair was successfully performed using an APOLO® 16 x 25 x 85 mm endograft (NANO² Endoluminal). Control angiography showed absence of early venous filling. The patient presented good recovery with decrease in the aneurysm size and improvement of limb swelling. He was discharged after 14 days, with discontinuation of antibiotic therapy.

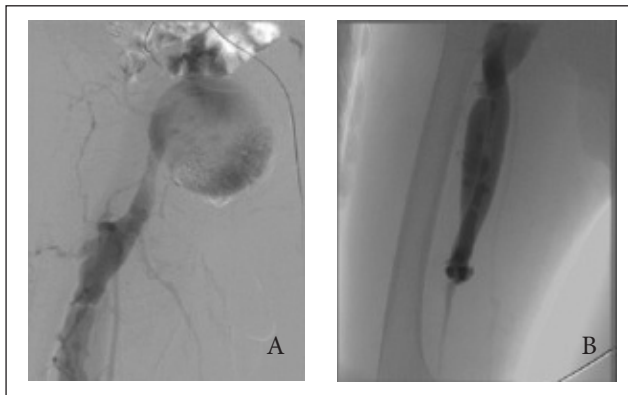


Figure 1. Digital subtraction angiography. (A) Giant vein aneurysm and (B) arteriovenous fistula in the superficial femoral artery with early venous filling.

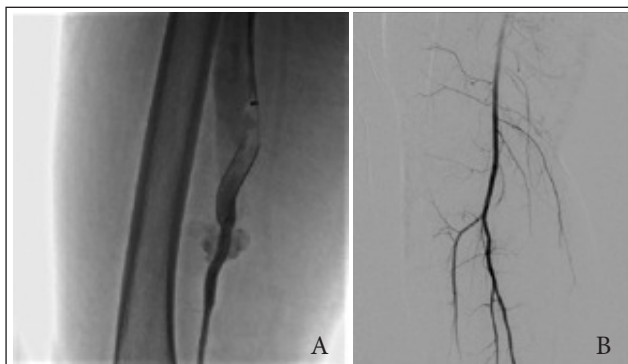


Figure 2. Digital subtraction angiography. (A) Follow-up image after placement of endograft showing distal arterial filling and late venous filling; (B) Endograft placed with filling of the leg arteries.



Figure 3. Asymmetric swelling of the lower limbs and signs of Phlegmasia Cerulea Dolens.

After a month of outpatient follow-up, Color-Doppler ultrasonography showed the giant venous aneurysm to be filled with thrombi (Figure 5) and persistence of the AVF at the adductor canal. He was readmitted for full anticoagulation therapy with unfractionated heparin. The patient developed renal failure due to bilateral compression of ureters by the aneurysm, with creatinine levels at 2.0 mg/dL. It was then decided to perform open surgical treatment of both the AVF and the aneurysm. An Optease® filter was placed in the inferior vena cava before the operation in order to avoid embolization during the procedure. On the following day, the AVF was treated by a femoro-femoral reverse saphenous vein interposition graft. The aneurysm was concomitantly repaired by a retroperitoneal approach. Simple

ligation of the external iliac vein was performed because of the absence of venous flow into the aneurysm after ligation of the thigh AVF (Figures 6 and 7).

The patient had a good recovery, presenting improvement of renal insufficiency (with creatinine levels back to normal levels) and of limb swelling, with normal distal pulses. The only postoperative complication was



Figure 4. CT angiography. (A) Pelvis – venous phase, showing giant vein aneurysm with 17 cm in diameter; (B) lower limbs – arterial phase showing new arteriovenous fistula after placement of endograft two years earlier.

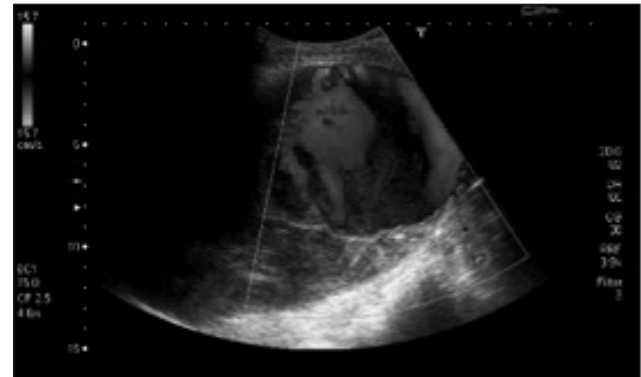


Figure 5. Doppler ultrasonography showing giant vein aneurysm containing a thrombus.



Figure 6. Femoro-femoral bypass using reversed saphenous vein graft after AVF repair.

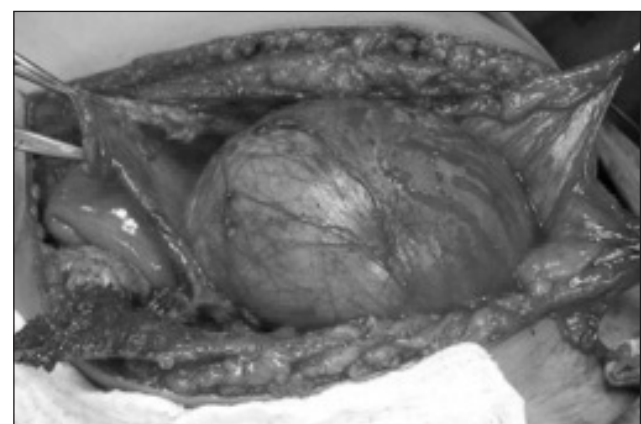


Figure 7. Giant external-iliac-vein aneurysm.

retroperitoneal bleeding, with a 2-mg/dL reduction of serum hemoglobin, which was resolved with discontinuation of the anticoagulation therapy. Follow-up Doppler ultrasonography showed patent graft and absence of AVF. The patient was discharged from the hospital after one month using acetylsalicylic acid 100 mg/day and statin, in good clinical condition and presenting improvement of limb swelling.

Discussion

Vein aneurysms are rare, difficult to diagnose and usually found incidentally at physical examination or by imaging methods⁶. They have been found in different locations but are rarely at the external iliac vein; most cases are secondary to a congenital or acquired AVF^{1,7,8}. The initial clinical presentation may be deep vein thrombosis (DVT), asymmetric edema of one extremity or pulmonary embolism^{7,9-11}.

The pathophysiology of iliac vein aneurysms remains uncertain. It has been proposed that the increased blood flow with consequent arterial dilatation proximal to the fistula causes significant venous outflow obstruction, which results in local venous hypertension. The increased flow and pressure on the iliac vein wall result in secondary aneurysmal dilatation².

Doppler ultrasound, phlebography, computed tomography (CT) and magnetic resonance imaging (MRI) are used for the differential diagnosis with intra- and extravascular retroperitoneal tumors, ovarian cysts, lymphoceles, "urinomas", arterial aneurysms and pseudoaneurysms. These imaging methods should be also used to plan surgical repair of venous aneurysms³.

Most authors agree that large abdominal vein aneurysms can potentially lead to thrombosis and pulmonary embolism. Despite being rare, when aneurysms are symptomatic and not fully thrombosed, resection or surgical repair may be indicated in order to avoid complications or clinical sequelae such as compression of adjacent structures, embolization or rupture, provided that the patient has an acceptable surgical risk^{2,3,12,13}.

Because there are few reports on this subject, little is known about the risk of thromboembolic events during aneurysm surgical repair or in the postoperative period. An important step in the assessment of thromboembolic risk is the performance of preoperative Doppler ultrasound or CT angiography to determine the presence and extension of thrombi within the aneurysm, and to decide the need for vena cava filter placement to reduce the risk of perioperative pulmonary embolism². In the case described, we decided to place a vena cava filter because fresh thrombi were

found inside the aneurysm immediately after endovascular repair of the AVF.

We decided to perform open surgical repair on account of the great volume of recent thrombi, which indicates high risk of pulmonary embolism. Besides that, compression on both ureters was causing acute obstructive renal failure.

Our initial plan was to repair the AVF and to perform tangential aneurysm resection with reconstruction by lateral venorrhaphy, thereby preserving the right lower limb axial venous drainage. However, after retroperitoneal exposure and venotomy, we observed low venous flow into the aneurysm with significant reduction of the venous system pressure after ligation of the AVF. We concluded that a bypass would not be necessary because long-term venous hypertension had provided an effective collateral circulation around the aneurysm. We then decided to perform only external iliac vein ligation because the low pressure and flow through an arterialized venous system would provoke thrombus formation within the graft.

Aneurysm resection with end-to-end anastomosis is indicated when it is possible to bring the two venous stumps together without tension. Otherwise, autologous vein grafts such as the saphenous vein or PTFE interposition are viable alternatives. The choice of the graft (autogenous or synthetic) depends on the availability of a venous conduit and on the diameter of the proximal stump. The best long-term patency results are obtained with tangential resection and lateral venorrhaphy, as success rates range from 40 to 93% in literature^{5,11,13-15}.

As we used inferior vena cava filter and placed the patient on full anticoagulation for 30 days (period of high risk for thrombosis and embolism), he had a good postoperative course, with cessation of the retroperitoneal bleeding, and was discharged from the hospital without oral anticoagulants.

In this case, we described two treatment approaches for lower limb AVF: endovascular and open surgery. Initial failure of the endovascular technique was probably due to the AVF high flow and to the differences in caliber between the upstream and downstream arterial segments that made endograft deployment more difficult. Despite the high morbidity rates related to conventional treatment, it is effective and should be the choice whenever the patient's anatomy is unfavorable or when closure of the fistula is not achieved with the endovascular treatment. Open surgical treatment for vein aneurysms should aim the reconstruction of the main venous axis either by autologous vein or synthetic graft interposition or by tangential venous resection with lateral

venorrhaphy whenever the anatomical conditions are favorable. The dearth of publications on iliac vein aneurysms, as well as the low frequency of cases, contributes to our limited knowledge about their natural history and adequate methods of treatment.

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Correspondence

Patrick Bastos Metzger
 Instituto Dante Pazzanese de Cardiologia
 Av. Dr. Dante Pazzanese, 500 – Vila Mariana
 CEP 04012-909 – São Paulo (SP), Brazil
 E-mail: patrickvascular@gmail.com

Author's contribution

Conception and design: PBM, NMI, HAB, FHR
 Analysis and interpretation: PBM, NMI, ACGP
 Data collection: MHS, JEMB, FMRA
 Writing the article: PBM, NMI, CBB
 Critical revision of the article: PBM, NMI, HAB, ACGP
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