

Case
Report

Unilateral Lower Extremity Edema and Lymphorrhea as Manifestations of a Ruptured Iliac Artery Aneurysm and Arteriovenous Fistula

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An arteriovenous fistula is a rare complication of ruptured abdominal aortic or iliac artery aneurysms (IAAs). Its clinical symptoms depend on its size, with signs of heart failure dominating if the fistula is large. Herein, we present a case of arteriovenous fistula with an unusual presentation. An 86-year-old male patient presented with extreme lower extremity edema, skin erosion, and continuous lymphorrhea (starting 3 months prior). Despite the large fistula between the right common IAA and the left common iliac vein after the rupture of the aneurysm, he did not develop heart failure symptoms, as the large shunt was predominantly directed toward the left lower extremity due to iliac vein compression. Careful physical examination and a high index of suspicion could have contributed to an earlier diagnosis and management.

Keywords: abdominal aortic aneurysm, fistula, shunts, edema

Introduction

An arteriovenous fistula is a rare complication of a ruptured abdominal aortic aneurysm (AAA) or iliac artery aneurysm (IAA).^{1,2)} Various signs and symptoms develop as a result of arteriovenous shunting, including dyspnea, hematuria, signs of renal or cardiac insufficiency, leg ischemia, and leg edema. The clinical symptoms depend on the size of the fistula, and signs of high-output heart failure may dominate if the fistula is as large as 15 mm in diameter.³⁾ We

encountered a case of a large arteriovenous fistula due to aneurysm rupture, with unique manifestations of extreme unilateral lower extremity edema, skin erosion, and continuous lymphorrhea, but with no signs of heart failure.

Case Report

An 86-year-old male patient was referred from a local hospital for evaluation and treatment of a swollen leg and continuous lymphorrhea from skin erosion (starting 3 months prior). His hemodynamic parameters were stable, with blood pressure of 98/54 mmHg and heart rate of 70 beats/minute. His chest radiograph and echocardiogram did not show any signs of heart failure. His left lower extremity had marked edema as well as discoloration, thickened and hardened skin, and skin erosion and infiltration, from which continuous lymphorrhea was observed (**Fig. 1a**). A computed tomography (CT) angiogram revealed an AAA (57 mm in diameter), right common IAA (92 mm in diameter), and small left internal IAA (**Fig. 2a**). The left external and internal iliac veins as well as the left femoral veins were opacified with contrast media in the early phase, indicating arteriovenous

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Fig. 1 Photographs of the patient's lower extremities. (a) Preoperative photograph showing swelling and discoloration of the left lower extremity as well as skin erosion and infiltration. (b) Photograph on postoperative day 14 showing decreased swelling, subsidence of the redness of the skin, and healed skin ulcer erosion.

shunting; however, the inferior vena cava was only vaguely enhanced. The left common iliac vein was occluded due to compression by the huge right common IAA (**Fig. 2b**). Therefore, arteriovenous shunting was mainly directed toward the left lower extremity, leading to the dominant manifestation of venous stasis in the left leg instead of heart failure.

Emergency surgery was performed through a midline abdominal incision. Strong adhesion between the right common iliac artery and the left common iliac vein precluded the dissection aimed to separate these vessels and to clamp the left iliac vein. A cross-clamp was applied on the infrarenal aorta, the right external and internal iliac arteries, and the left common iliac artery. Upon opening the aneurysm, a large fistula between the right common IAA and the left common iliac vein (diameter, 10×20 mm) was noted (**Fig. 3**). The bleeding from the fistula was controlled by compressing the distal left common iliac vein. The fistula was closed using a bovine pericardial patch. The AAA and IAA were then replaced with a bifurcated Dacron graft. The postoperative course was uneventful. His lymphorrhea

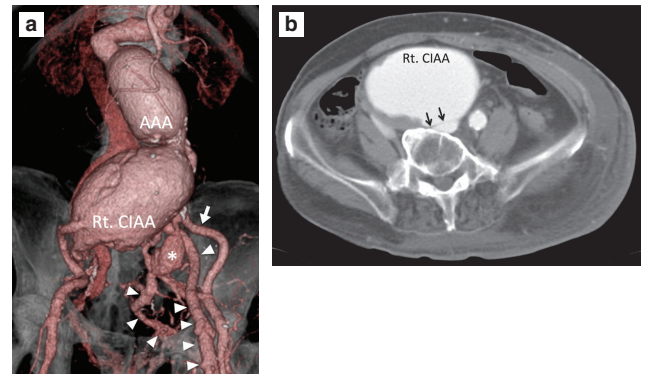


Fig. 2 Postoperative CT. (a) Preoperative CT angiogram revealing the right common IAA (Rt. CIAA), and left internal IAA (asterisk). The left internal iliac vein (arrowheads) is clearly enhanced in the early phase, whereas the inferior vena cava is barely opacified. The arrow indicates the left external iliac artery. (b) The proximal portion of the left iliac vein (black arrows) is compressed between the right common iliac artery and the vertebrae. The left iliac vein is equally enhanced as the iliac artery. AAA: abdominal aortic aneurysm; IAA: iliac artery aneurysm; CT: computed tomography

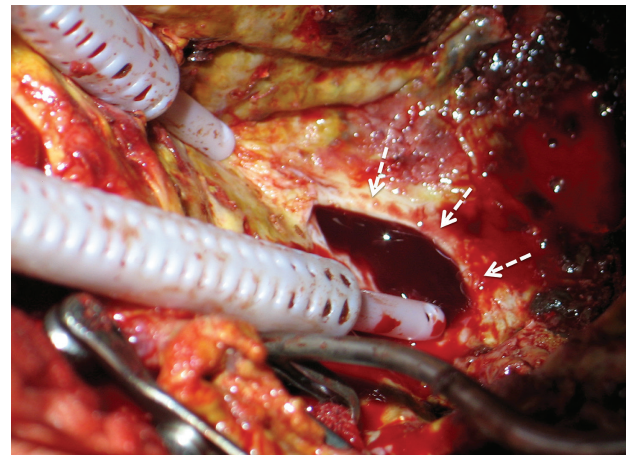


Fig. 3 Intraoperative photograph delineating the arteriovenous fistula, measuring 15×20 mm in diameter (dotted arrow).

ceased soon after the operation, and with compression therapy, only mild edema remained on postoperative day 14 (**Fig. 1b**). A postoperative CT scan revealed the disappearance of the shunt and persistent occlusion of the left common iliac vein, with thrombus formation only at the proximal portion of the left iliac vein (**Fig. 4**).

Discussion

An arteriovenous fistula is known to have different clinical manifestations. Clinical symptoms depend on the



Fig. 4 Serial axial images of postoperative CT shows thrombus formation at the most proximal portion of the left iliac vein (arrows), where persistent occlusion of the proximal left iliac vein even after decompression of the aneurysm (asterisks) is recognized. CT: computed tomography

size of the fistula, and signs of high-output heart failure may dominate if the fistula is large, leading to mortality.³⁻⁵⁾ The present case is unique, as the size of the communicating fistula was large enough to develop severe venous stasis of the left lower extremity with concomitant lymphorrhea, but the patient did not experience heart failure, as the shunting was directed predominantly toward the left lower extremity. Therefore, the diagnosis

was delayed, resulting in the patient experiencing chronic venous stasis of the leg.

Iliac vein compression syndrome, also known as May–Thurner syndrome, is an obstruction of the left iliac vein between the iliac artery and the lumbar vertebrae. Long-standing obliteration results in fibrotic adhesion of the intima, which is an irreversible process. The most common clinical presentation is deep vein thrombosis; however, symptoms can be either acute or chronic, including left lower extremity swelling, pain, venous claudication, ulceration, and varicose veins.^{6,7)}

The symptoms of venous obstruction might have been caused by the occlusion of the left iliac vein or the superimposed rupture of the aneurysm, leading to a large shunt. Considering the severity of the symptoms, the sudden onset 3 months prior to admission, and the prompt relief of the symptom after the operation, despite the persistent occlusion of the left iliac vein, the symptoms are presumably attributed to arteriovenous fistula formation, but the exact onset of the aneurysm rupture was not clearly recognized by the patient.

Postoperative CT showed persistent occlusion of the left common iliac vein, which had not been directly treated during the operation. Currently, the use of endovascular techniques is highly successful and carries less risk than invasive surgical treatments. In the present case, once the large arteriovenous fistula was closed surgically, clinical symptoms related to the venous stasis of the left lower extremity were well controlled with conservative treatment, including compression therapy and exercise, despite persistent venous occlusion after the operation. In such a situation, collateral development might be enough to preclude the indication for further intervention. Hence, no invasive treatment for iliac vein occlusion was planned in the present case.

Conclusion

Arteriovenous fistula caused by large AAAs and IAAs can present unique manifestations such as unilateral lower extremity edema and lymphorrhea. Careful physical examination and a high index of suspicion, which might lead to CT evaluation, can contribute to an early diagnosis.

Disclosure Statement

All the authors have no conflicts of interest to report.

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