# Hepatorenal syndrome from an ilio-iliac arteriovenous fistula: A rare complication from an endoleak

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## ABSTRACT

Arteriovenous fistula is a rare and often unrecognized complication of aneurysms, with a varied and frequently inconsistent presentation. We present the case of an ilio-iliac arteriovenous fistula formation in a 71-year-old man associated with a type III endoleak after endovascular iliac branch repair. Because of rapidly progressing congestive heart failure and hepatorenal syndrome, we performed urgent endovascular repair with successful endoleak exclusion. After the procedure, the patient demonstrated a remarkably rapid and complete recovery. (J Vasc Surg Cases and Innovative Techniques 2021;7:654-8.)

**Keywords:** Arteriovenous fistula; AVF; Endoleak; Endovascular repair

The complications of aneurysms such as rupture and embolism have been well described. However, arteriovenous fistula (AVF) remains a rare and frequently unrecognized complication of aneurysms. In such cases, aneurysm progression will result in erosion into an adjacent vein, creating an arteriovenous shunt.[1] The anatomic proximity of the iliac vessels makes iliac artery aneurysms susceptible to fistula formation. These ilio-iliac AVFs are most often caused by trauma, although spontaneous occurrences have been reported.[2,3] Patients can present with a lower abdominal mass accompanied by a thrill or bruit, unilateral lower limb arterial and/or venous insufficiency, and/or symptoms of congestive heart failure. The prompt diagnosis of ilio-iliac AVF is vital, and failure to treat ilio-iliac AVFs expeditiously can result in limb ischemia, shock, or death.[2,3] We have presented an uncommon case of an ilio-iliac AVF resulting from progression of a common iliac aneurysm in the setting of a type III endoleak after endovascular iliac branch repair that caused congestive heart failure and hepatorenal syndrome (HRS). The patient provided full written consent for the report of his case.

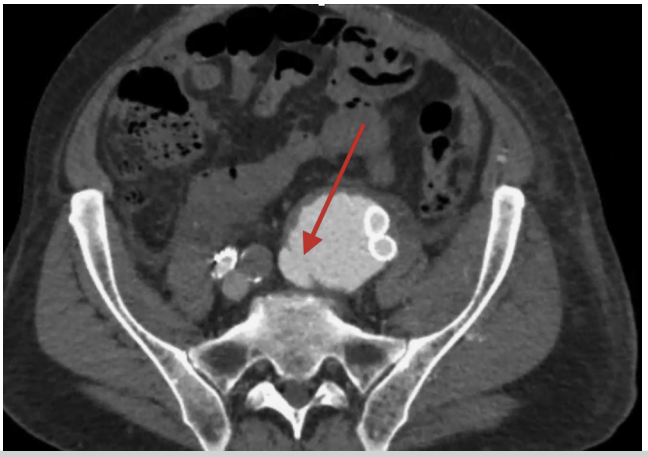

**Fig 1.** Computed tomography angiogram demonstrating a type III endoleak in the left internal iliac artery and compression of the left common iliac vein by the aneurysm sac.

## CASE REPORT

A 71-year-old man with a 6-cm infrarenal aortic aneurysm and a 5.5-cm left common iliac artery aneurysm had undergone endovascular aneurysm repair with a Gore Excluder iliac branch endograft (W. L. Gore and Associates, Flagstaff, Ariz) at an outside hospital. At the time of surgery, a compliant aortic balloon was used to seal the internal iliac branch gate with the internal iliac stent extension (Viabahn VBX stent-graft; W. L. Gore & Associates). The patient tolerated the procedure well but had developed a type III endoleak of the internal iliac branch found on postoperative imaging studies. Reintervention was planned but was delayed owing to the outbreak of the coronavirus 2019 pandemic.

The patient presented 5 months later to the same hospital with a 3-week history of left leg swelling and dyspnea on exertion. He had a history of hepatitis C virus infection with mild cirrhosis and had been receiving ongoing treatment. On

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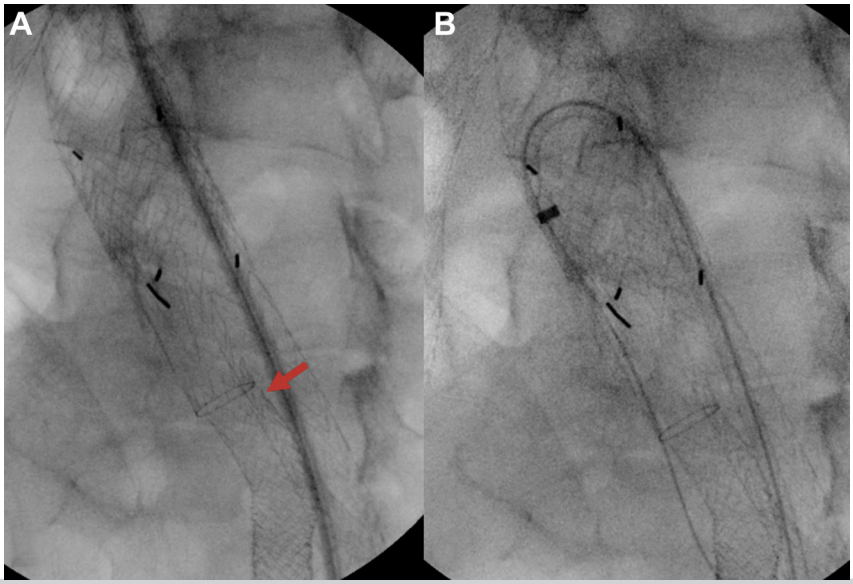


**Fig 2. A,** Fluoroscopic image of stent separation (*red arrow*) before repair. **B,** Fluoroscopic image of wire demonstrating nonunion of the iliac endograft and internal iliac stent.

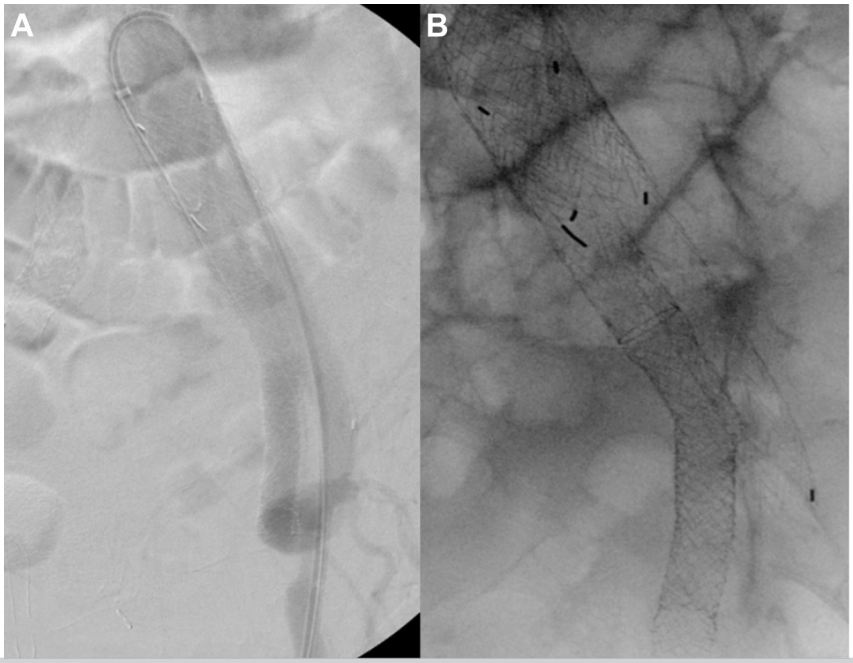


**Fig 3. A,** Subtraction imaging after repair of type III endoleak. **B,** Fluoroscopic image of the stent after type III endoleak repair.

admission, his laboratory test results were notable for total bilirubin of 1.5 mg/dL, creatinine of 1.6 mg/dL, an international normalized ratio (INR) of 1.7, and sodium of 138 mEq/L, for a MELD (model for end-stage liver disease) score of 18. On physical examination, he had significant left leg edema with a palpable left femoral pulse. A lower extremity duplex ultrasound scan to rule out deep vein thrombosis demonstrated a lack of compressibility of the left common femoral vein to left popliteal vein. However, arterial waveforms were present for all deep veins on spectral analysis. A computed tomography scanning protocol for pulmonary embolism failed to show pulmonary embolism. The findings were concerning for cardiomegaly, right atrium enlargement, ascites, and hepatic steatosis. Computed tomography angiography of the abdomen and pelvis

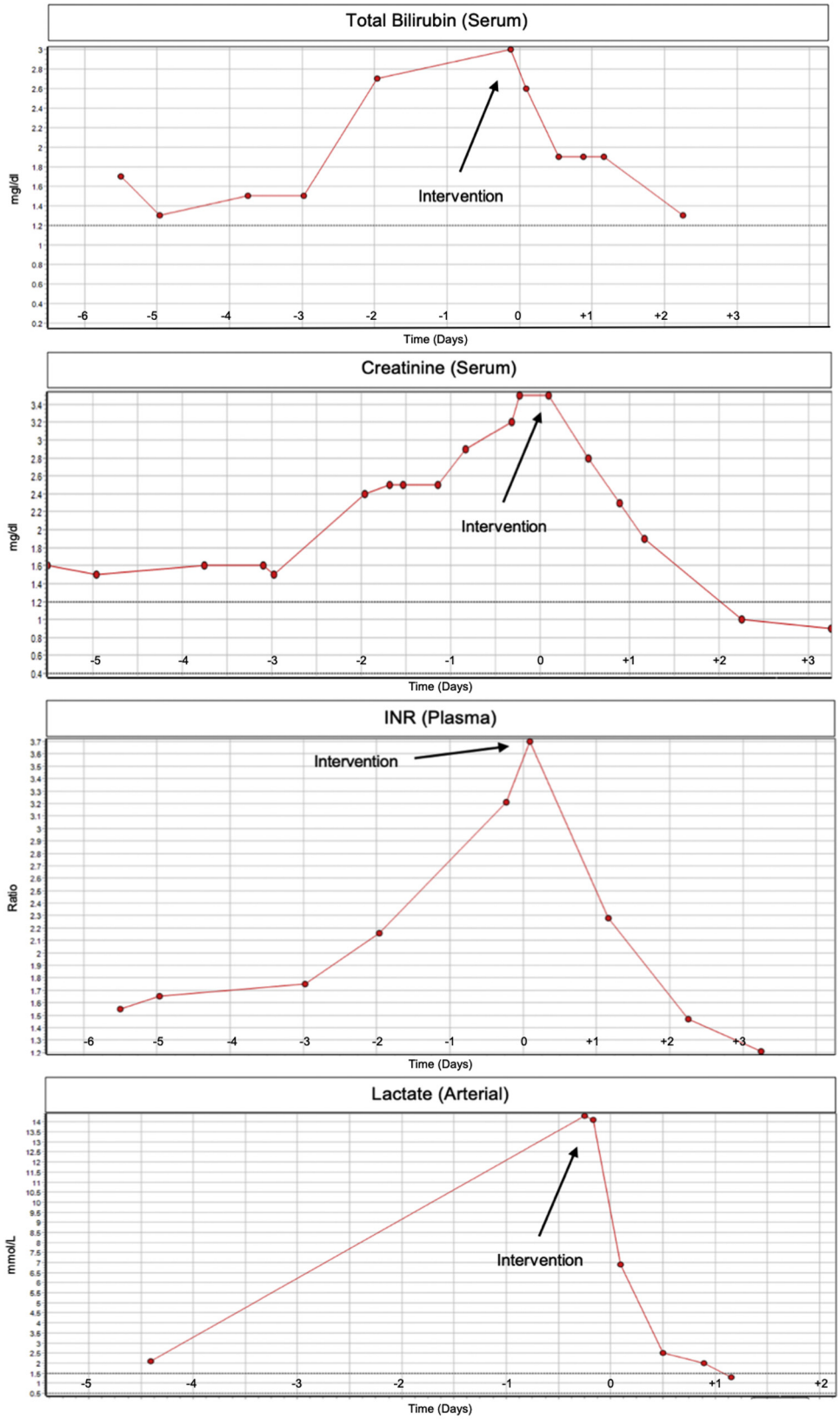


**Fig 4.** Graphs demonstrating the patient's rapid and significant improvement in international normalized ratio (INR), creatinine, total bilirubin, and lactate after fistula repair.

demonstrated a type III endoleak within the left internal iliac artery with separation of the stent-graft at the iliac gate and compression of the left common iliac vein by the common iliac aneurysm sac. Additionally, filling of the left iliofemoral veins and inferior vena cava during the arterial phase was present, secondary to erosion of the plane between the left common iliac artery and vein (Fig 1).

During interfacility transfer and preoperative optimization for 3 days, the patient deteriorated rapidly, developing progressive right heart failure and HRS (creatinine, 3.5 mg/dL; INR, 3.7; total bilirubin, 3.5 mg/dL; lactate, 14 mmol/L). Given this precipitous decline, he underwent urgent endovascular repair. After obtaining ipsilateral femoral access, a steerable sheath (Nagare; Terumo Medical Corp, Somerset, NJ) was used to obtain wire access across the internal iliac branch and stent extension. The type III endoleak was confirmed and the presence of a distal type Ic endoleak excluded before repair (Fig 2). A Viabahn VBX stent-graft (11 × 59 mm) was used to bridge the iliac branch and stent extension and was dilated at the iliac branch interface with a larger, noncompliant balloon (14 × 60 mm) to prevent a subsequent type III endoleak (Fig 3). Completion angiography showed direct filling of the internal iliac artery with exclusion of the type III endoleak. The patient's clinical course improved dramatically during the next 3 days, including his shortness of breath and leg swelling (creatinine, 1.0 mg/dL; INR, 1.2; total bilirubin, 1.3 mg/dL; lactate, 1.3 mmol/L), and he was discharged home (Fig 4).

## DISCUSSION

Ilio-iliac AVFs are exceedingly rare, occurring in <1% of patients with common iliac aneurysms.[4] Spontaneous formation is the rarest etiology of ilio-iliac AVFs, occurring in only 16% of cases, with the remainder typically resulting from trauma, malignancy, or iatrogenesis.[5,6] The clinical presentation of ilio-iliac AVFs can vary and can include rapid-onset high output cardiac failure, a pulsatile abdominal or pelvic mass, unilateral leg edema, and acute kidney injury.[2,6] These findings do not occur consistently or uniformly and could be present in fewer than one half of patients.[2,5,6] Less common symptoms include limb ischemia, acute liver failure, paradoxical emboli, and intestinal angina.[2,6]

Ilio-iliac AVFs lead to profound physiologic changes, with a reduction in afterload creating a steal-like phenomenon in which blood is shunted from the arterial system to the venous system. To overcome this shunt, the cardiac output must increase to maintain perfusion to the body, leading to sustained tachycardia. Over time, this can lead to high output cardiac failure and hepatic congestion. As in the present patient, progressive hepatic congestion can exacerbate existing liver failure and, in severe cases, can evolve into HRS. However, repair of the ilio-iliac AVF will reverse many, if not all, of its sequelae, as demonstrated in our patient (Fig 4). Recognition of these physiologic effects is paramount during the perioperative period, because they can affect a patient's risk of perioperative morbidity and mortality and influence the anesthetic strategies.[7,8]

Renal dysfunction has often been noted in patients with ilio-iliac AVF without liver disease, frequently attributed to hemodynamic compromise from decreased arterial pressure and increased venous pressure.[6] However, this is distinct from the occurrence of HRS in our patient. HRS, which typically stems from decompensated cirrhosis, is marked by splanchnic vasodilation, decreased arterial resistance, and functional hypovolemia, leading to severe renal vasoconstriction.[9,10] In addition to the hepatic congestion exacerbating our patient's existing cirrhosis, the fistula-related decrease in arterial resistance and arterial volume likely precipitated the rapid onset of HRS.

In recent years, the management of iliac aneurysms and related pathologies (ie, AVF) has shifted from open to endovascular repair owing to the lower morbidity and mortality.[1,5] Similar to the repair of common iliac aneurysms, the endovascular repair of ilio-iliac AVFs was initially approached with embolization of the internal iliac artery, followed by iliac stent exclusion. The advent of iliac branch devices has offered an alternative method for successful ilio-iliac AVF repair with preservation of pelvic perfusion.[1]

Our patient's AVF had either resulted from aneurysm enlargement and adjacent erosion or an iatrogenic injury that had occurred during wire manipulation during the initial repair. The failure to use a noncompliant balloon led to improper stent-graft apposition to the gate of the branch piece, resulting in the type III endoleak. As such, an endovascular approach was the most attractive to repair the endoleak by bridging the dislodged components and excluding the fistula. Although exclusion of the fistula without direct repair did leave the possibility of a future type II endoleak, it is unlikely that the low pressure venous system will cause aneurysm sac expansion.[5] However, close follow-up is necessary and, in the setting of a persistent leak, further intervention might be warranted.[11] The present case illustrates the importance of including AVFs in the differential diagnosis of patients presenting with symptoms of high output heart failure in the setting of aneurysmal disease.

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