Successful surgical treatment with ex vivo technique in a patient with renal artery aneurysm rupture and bilateral arteriovenous fistula

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ABSTRACT

Renal arteriovenous fistulas are pathologic communications between the renal arteries and veins without interconnecting capillaries. Reports on the combination of fibromuscular dysplasia, aneurysms, and renal arteriovenous fistula are extremely rare in the literature. In the case of renal arteriovenous aneurysm rupture, urgent nephrectomy was the only life-saving procedure reported. The ex vivo approach seems to be an appropriate alternative to nephrectomy. This article presents a case of successful treatment of bilateral fibromuscular dysplasia with aneurysm and arteriovenous fistulas complicated by renal artery aneurysm rupture. (J Vasc Surg Cases and Innovative Techniques 2018;4:232-6.)

Keywords: Arteriovenous fistula; Fibromuscular dysplasia; Renal artery aneurysm; Ex vivo; Embolization

Renal arteriovenous fistulas (AVFs) are rare pathologic communications between the renal arteries and veins without interconnecting capillaries. They are divided into two categories: congenital and acquired, with the latter type accounting for up to 70% cases of all renal fistulas.[1] According to the literature, the association of fibromuscular dysplasia (FMD) and renal artery aneurysm with shunting into the renal vein has been reported in only few patients.[2-4] To the best of these authors' knowledge, the present case is the only reported case of treated renal artery aneurysm rupture with the ex vivo technique in a patient with bilateral FMD and AVF.

Consent for publication was obtained from the patient.

CASE REPORT

A 36-year-old woman presented with a bruit in both flanks and discomfort in the right lower back. These symptoms appeared after childbirth in 2008. A systolic thrill of the anterior abdominal wall was observed during self-examination. The patient had no history of trauma, surgery, or any kind of invasive procedures.

All laboratory test values were within the normal limits, including the baseline creatinine level of 83 mmol/L and blood urea nitrogen of 2.79 mmol/L. During the examination, duplex ultrasound examination revealed bilateral giant renal aneurysms with AVF. The peak systolic velocity in both renal AVF was 250 cm/s. Subsequently, the patient underwent contrast-enhanced computed tomography (CT) angiography (Philips Brilliance iCT-256, Philips Medical System, Cleveland, Ohio) of the abdomen. CT images showed enhancement of renal arteries and bilateral giant renal artery aneurysms with early enhancement of the dilated renal veins and the inferior vena cava (Fig 1). The right kidney had two renal veins, and the left kidney was supplied by two arteries.

The string of beads sign that is specific for FMD was revealed on the selective right renal arteriography. An Amplatzer Vascular Plug IV 9-AVP038-008 (St. Jude Medical, St. Paul, Minn) was implanted in the right renal artery aneurysm. At completion angiography, no AVF was detected, and opacification of the veins was seen 7 to 9 seconds after injection, which is indicative of fistula closure (Fig 2).

The next day, the patient began to experience weakness and pain in her right lower back. These symptoms progressed, and renal infarction or renal aneurysm rupture was suspected. A contrast-enhanced multidetector CT scan showed partial thrombosis of the right renal artery aneurysm and occluding thrombosis of the lower right renal vein with a floating thrombus in the inferior vena cava. The patient underwent urgent surgery. Thrombectomy of the inferior vena cava with ligation of the right lower renal vein was performed. Additionally, the feeding arterial branch of the fistula was exposed and ligated (Fig 2, *c*, and *d*).

On the second postoperative day, the patient was transferred from the intensive care unit and discharged from the hospital on postoperative day 16 in good general condition.

The patient was readmitted to hospital a few weeks later for closure of a left renal AVF. Laboratory test results were within the reference range. Despite the previous experience, an endovascular approach was chosen as a less invasive procedure.

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Author conflict of interest: none.

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The editors and reviewers of this article have no relevant financial relationships to disclose per the Journal policy that requires reviewers to decline review of any manuscript for which they may have a conflict of interest.

2468-4287

https://doi.org/10.1016/j.jvscit.2018.03.007

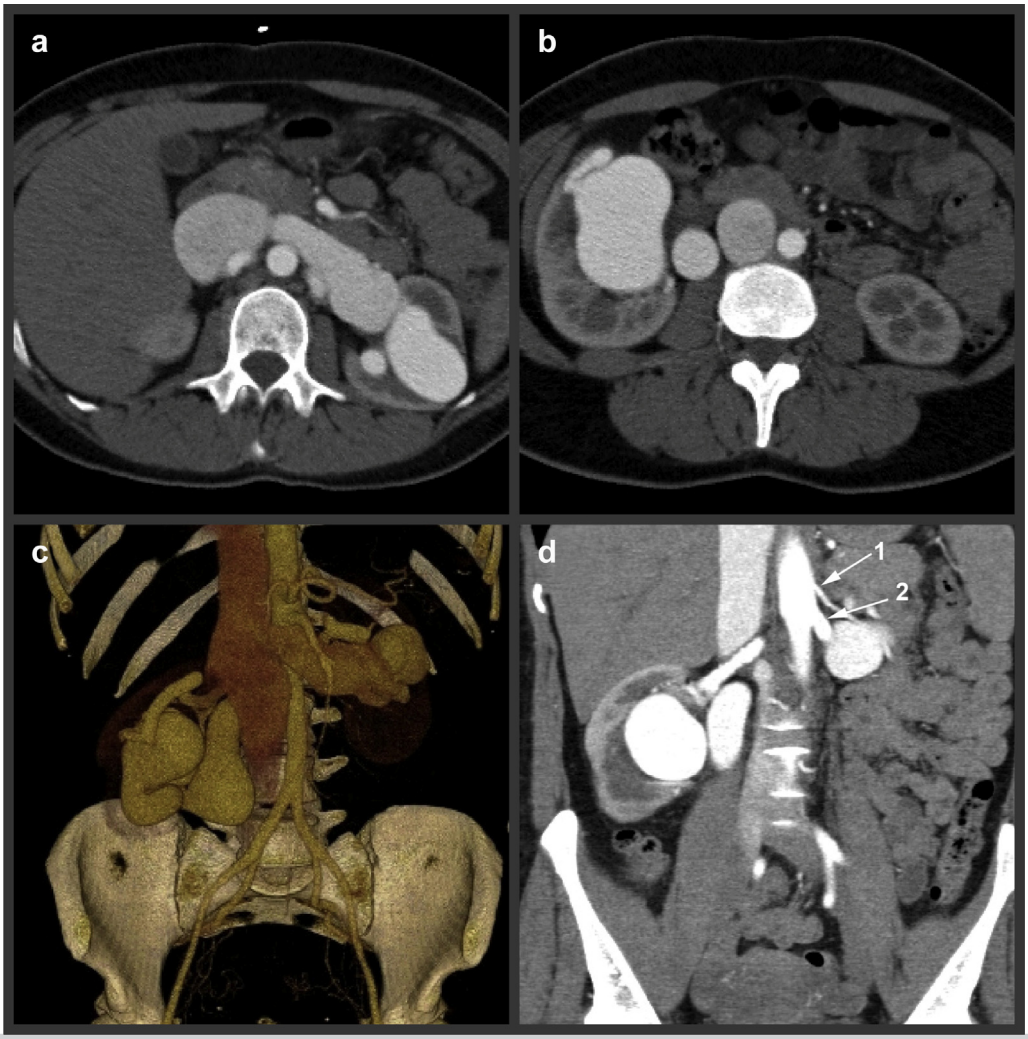

Fig 1. Computed tomography (CT) angiography of bilateral arteriovenous fistula (AVF). **a** and **b,** Axial images and **(c)** the volume-rendered image show aneurysmal dilatation of both renal arteries and veins with early contrast enhancement of the renal veins and inferior vena cava. **d,** The coronal reformation reveals the left lower pole renal artery (*1*) and the left upper pole renal artery (*2*).

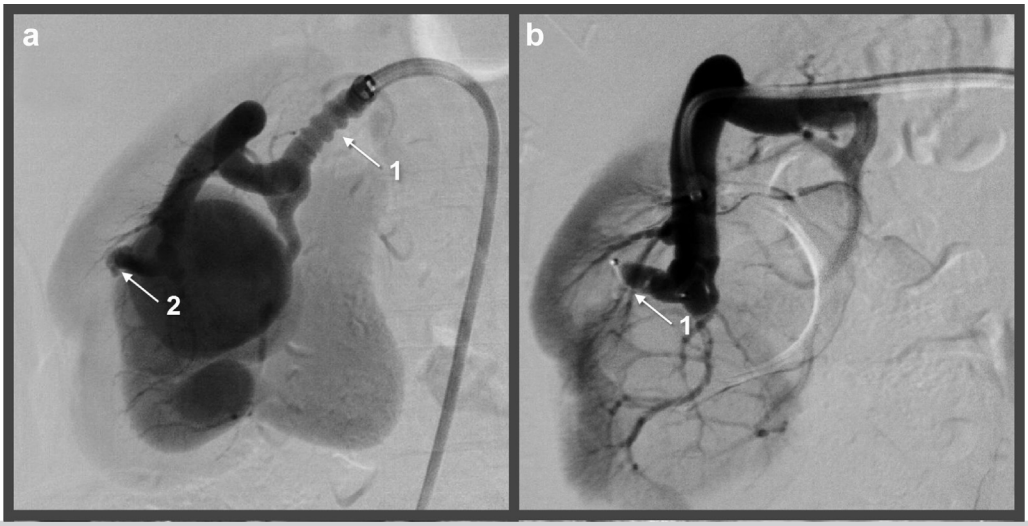

Fig 2. Angiographic and intraoperative images of the right renal arteriovenous fistula (AVF). **a,** Selective right renal angiogram shows the "string of beads" sign (*1*) of fibromuscular dysplasia (FMD) and the arteriovenous shunt (*2*). **b,** Right renal digital subtraction angiography image after transarterial embolization.

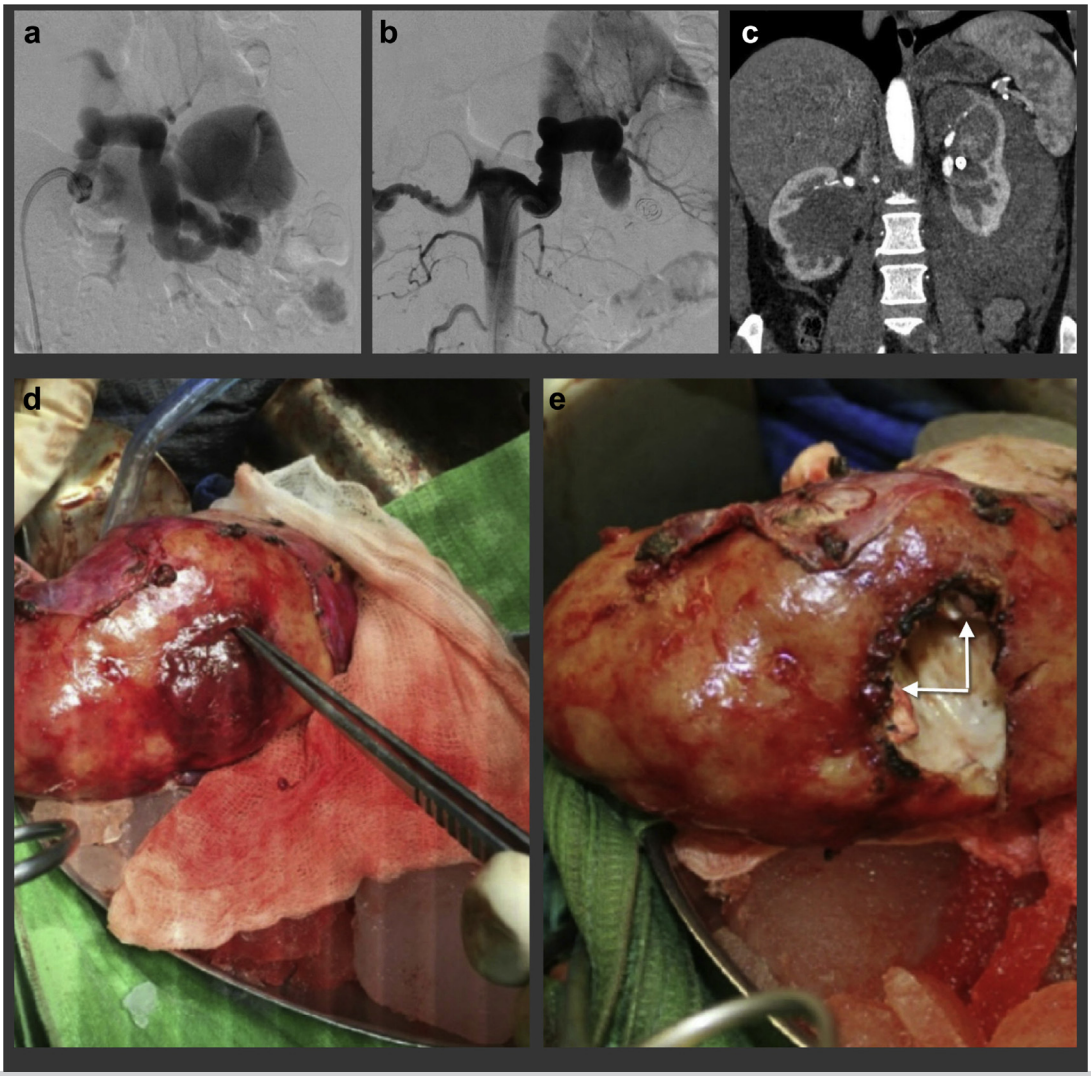

Fig 3. Digital subtraction angiography, computed tomography (CT), and intraoperative images of the left renal arteriovenous fistula (AVF). Left renal angiogram before **(a)** and after **(b)** coil embolization shows occlusion of fistula. **c,** CT angiography (coronal reformation) shows a perinephric hematoma. **d,** Intraoperative picture. The left kidney is placed in the tray with ice cubes. The tip of forceps inserted in the ruptured renal aneurysm. **(e)** Intraoperative image. The oversewn orifices of feeding and draining arteries with patches (*arrows*).

Selective renal arteriography showed the string of beads sign in the lower left renal artery, and the upper pole artery was intact. A renal artery aneurysm was observed in the upper pole with an AVF, which was fed by the lower renal artery. IMWCE 35-8-8 coils (Cook Medical, Bloomington, Ind) were implanted. The completion arteriography showed complete left renal aneurysm occlusion and blood flow cessation in the AVF (Fig 3, *a*, *b*). Vein opacification was seen at 6 to 8 seconds after injection.

The immediate postoperative period was unremarkable except for progressive weakness. On postoperative day 2, planned duplex ultrasound and CT imaging revealed a thrombosed left renal aneurysm and perinephric hematoma with contrast extravasation (Fig 3, *c*).

The patient underwent an urgent operation through an L-shaped laparotomy. A massive retroperitoneal hematoma was found and removed, which revealed a rupture of the capsule. Considering that potential ischemia time could be too long, an ex vivo technique was used. Both renal arteries and both veins were ligated, with the kidney placed in crushed ice without ureter ligation.[5] The kidney was flushed as a bolus through the renal arteries with 700 mL of cold (4°C) Custodiol solution (HTK-Bretschneider, Dr Franz Köhler Chemie, Alsbach-Hähnlein, Germany). The ruptured renal aneurysm was incised. After thrombus, the aneurysm draining veins were visualized and oversewn, and the kidney capsule was sutured. The upper left renal artery was anastomosed to the main renal artery side. The lower left renal artery was reanastomosed to the aorta. The left renal vein was reattached to the native renal vein (Fig 3). Warm and cold ischemia times were 10 minutes and 2 hours 25 minutes, respectively. The patient recovered uneventfully.

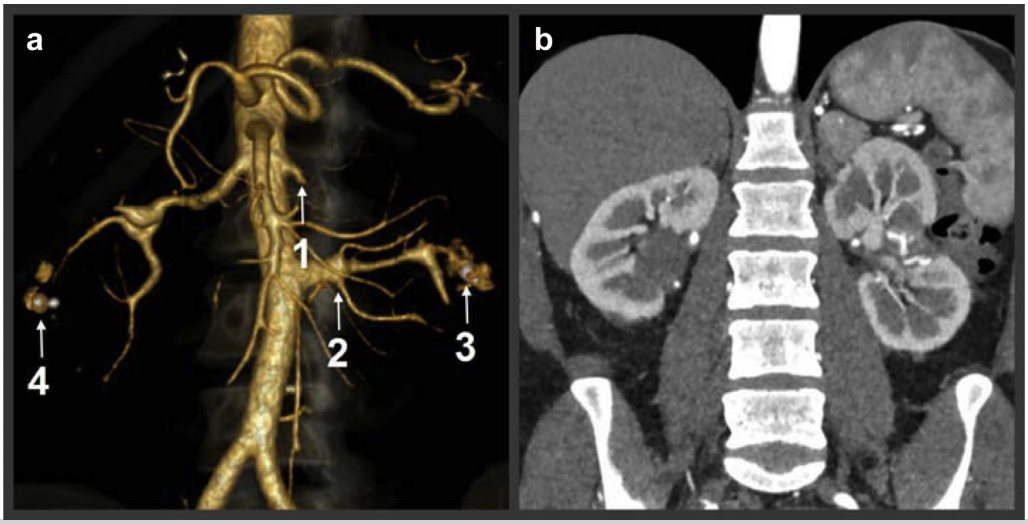

Fig 4. Computed tomography (CT) at 9-months follow-up. **a,** Volume rendering image demonstrates the optimal surgical result after repair: the stump of the left upper-pole renal artery (*1*), reimplanted left upper pole renal artery (*2*); coil (*3*); and occluder (*4*). **b,** Renal parenchyma of both kidneys shows normal enhancement, there is only a small scar in the middle third of the left kidney.

At a follow-up multidetector CT examination 9 months after the last surgery, persistent occlusion of both renal artery aneurysms and AVF was diagnosed (Fig 4). On radionuclide scintigraphy, both kidneys' excretory function was normal, with no signs of renal function impairment.

DISCUSSION

FMD of the renal arteries may be associated with hypertension and, less commonly, with spontaneous dissection, aneurysm, and renal infarcts or hemorrhage.[6,7] Association of the FMD with renal aneurysm and AVF is extremely rare and is presented in the literature by a limited series of case reports. In 2002, Torres et al[2] performed successful ex vivo resection of a renal artery aneurysm with AVF in a patient with FMD and hematuria. Arteriovenous aneurysm embolization was described by Altit et al[3] in 2009, who performed two elective coil embolization procedures to interrupt blood flow in the aneurysm and fistula. Garg et al[4] presented the Mayo Clinic experience in the treatment of AVF. In their series, one patient with FMD underwent successful coil embolization.

From our point of view, the modality of choice in patients with high blood flow velocity and/or wide diameter of AVF is open surgery.[8,9] Previously, in the case of renal arteriovenous aneurysm rupture (the complication we faced in the left-sided aneurysm), nephrectomy was a life-saving procedure.[10] An ex vivo approach seems an appropriate alternative to nephrectomy.[2,11]

The present report is the second case of extracorporeal treatment of AVF and the first one to be performed in an urgent situation. A severe complication such as aneurysm rupture can also be successfully treated with kidney function salvage. It seems that aneurysm rupture occurred during aneurysm embolization. For most AVF, endovascular management is possible, but in patients with large AVF and complex anatomy, endovascular and surgical management may be required for a successful outcome.

The authors thank A. Teplov, I. Timina, A. Gritskevich, and A. Adyrkhaev for assistance.

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Submitted Aug 19, 2017; accepted Mar 15, 2018.