A rare case of concurrent proximal and hilar renal artery aneurysm with renal arteriovenous fistula

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ABSTRACT

Renal artery aneurysm (RAA) and renal arteriovenous fistula are rare vascular pathologies with reported incidences of 0.3% to 1.0% and 0.04% in the general population, respectively. We describe a 61-year-old Caucasian man who presented to the hospital with symptoms of right flank pain. Imaging demonstrated a right RAA with concurrent hilar RAA and renal arteriovenous fistula. He ultimately underwent an open right nephrectomy, ligation of the fistula, and bovine patch repair of the aortic defect. (J Vasc Surg Cases Innov Tech 2022;8:142-5.)

Renal artery aneurysms (RAA) and renal arteriovenous fistulae (RAVF) are uncommon pathologies. Here we present a rare case of a proximal main RAA with concurrent hilar aneurysm with fistula to the renal vein in a patient with no other vascular history and review the literature.

CASE REPORT

A 61-year-old Caucasian man with history of hypertension, alcohol dependence, and previous blunt trauma in 1979 requiring open splenectomy presented with chief complaint of right flank pain for 5 days. He was bacteremic presumably from a urinary tract infection. Computed tomography angiography (CTA) showed a dilated, tortuous right renal artery (RRA) with a proximal aneurysm measuring 3.5 cm and a concurrent 4.5 cm hilar aneurysm with early contrast opacification of the renal vein and inferior vena cava (IVC) concerning for RAVF (Fig 1). A diagnostic arteriogram depicted an aneurysmal RRA with rapid filling of the suprarenal IVC (Fig 2). The most proximal RAA had a wide neck takeoff from the aorta; it was unclear where the RAVF originated (Fig 3). A multidisciplinary approach was taken owing to the complex anatomy, and he was deemed to not be a candidate for endovascular management. After completing a 14-day course of antibiotics for *Enterococcus* bacteremia, he was offered nephrectomy with primary repair of the aorta.

The patient subsequently underwent a midline laparotomy, right nephrectomy, ligation of the RAVF, and bovine patch repair of the aortic defect. The aorta was exposed via the transperitoneal approach and the right kidney and IVC via the Cattell-Braasch maneuver. The RRA was expectedly tortuous and aneurysmal; a thrill was palpated in the right upper quadrant. The fistula was found between the hilar aneurysm and the adjacent renal vein that drained directly to the IVC (Fig 4). Together with urology, we deemed the kidney nonsalvageable. The aorta was clamped above and below the RRA and below the left renal artery for proximal and distal control. The aneurysmal orifice of the RRA was identified and resected with a 2-cm defect, which was repaired with bovine patch. A right nephrectomy was performed. There were no intraoperative complications and the patient was discharged 7days later. His postoperative peak creatinine was 1.47 mg/dL and his new baseline was between 0.8 to 0.9 mg/dL. A follow-up CTA demonstrated no issues with the aortic repair and a normal appearing IVC.

DISCUSSION

Recently published clinical practice guidelines on the management of visceral aneurysm by the Society for Vascular Surgery listed the following indications for intervention in RAA: size of more than 3 cm, female gender within childbearing age regardless of size, symptomatic (pain, hematuria), medically refractory hypertension, and functionally important renal artery stenosis.[1] The previous repair threshold of 2 cm was increased to 3 cm based on more contemporary data describing the lower associated risk of rupture, slow growth rate, and improved survival after rupture.[1] Currently, there is no prospective or randomized control trial directly comparing surgical repair of RAAs that are more than 2 cm with surveillance. Intervention for RAA repair is driven by the risk of rupture, whereas the indications for treatment of an RAVF are symptom based, which include progressive increase in fistula size, hematuria, and medical complications associated with RAVF such as hypertension, high-output heart failure, and circulatory overload.[2,3]

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Author conflict of interest: none.

The patient was contacted before submission of the article and has given consent for the publication of this study.

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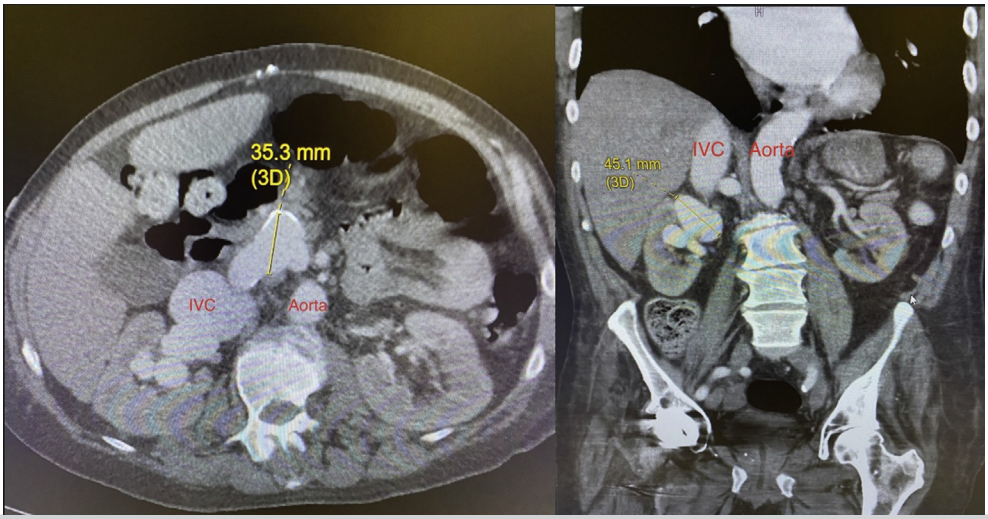

Fig 1. Axial view of computed tomography angiography (CTA) of the abdomen and pelvis depicting a 35.3-mm renal artery aneurysm (RAA) coming off of the aorta (left). Coronal view of CTA of abdomen and pelvis depicting a distal 45.1-mm RAA nearby the renal hilum (right). *IVC*, Inferior vena cava.

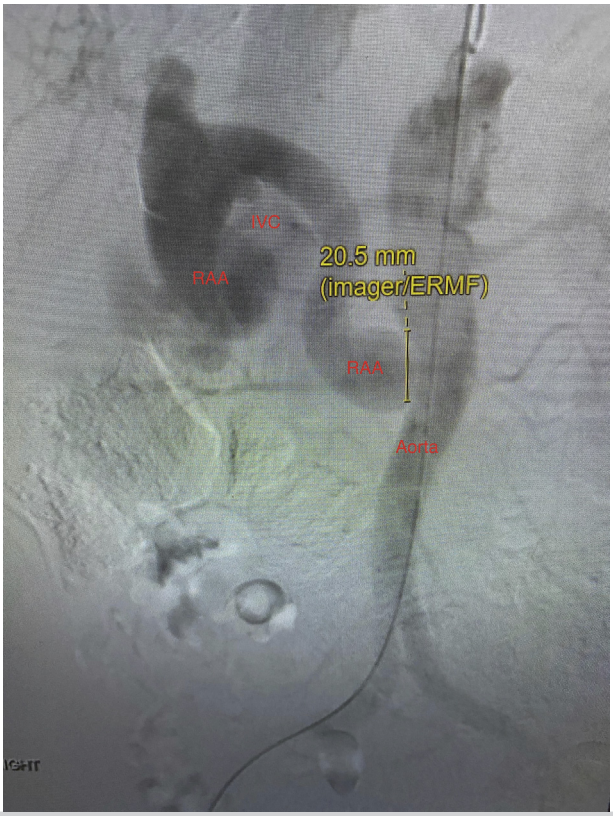

Fig 2. Aortogram showing the wide neck of the proximal renal artery aneurysm (*RAA*) coming off of the aorta measuring about 20.5 mm. *IVC*, Inferior vena cava.

RAAs can be repaired by an open or endovascular approach. Open aneurysmorrhaphy with arterial reconstruction is well-established, with the goal of resecting the aneurysm and restoring renal perfusion. After resection, vascular continuity can be achieved by primary closure with or without branch reimplantation, patch angioplasty, primary reanastomosis of the renal artery, interposition bypass, aortorenal bypass, or splanchnorenal bypass. An autogenous vein, typically the greater saphenous, or prosthetic grafts can be used with excellent patency rates for both. Saccular aneurysms can be repaired with suture plication or unroofing followed by patch angioplasty, with latter being the favored approach.[1,4,5] Complex distal branch lesions and multibranch RAAs can be treated with ex vivo repair; cold perfusion should be considered if the expected warm ischemic time exceeds 30 minutes.[4] Finally, nephrectomy is indicated in patients who have nonreconstructable anatomy, failed reconstruction, or contraindications for kidney salvage, such as renal cell carcinoma or end-stage ischemic nephropathy.[4-6] Reports of robotic repairs have been recently published without short-term or long-term outcome assessments.[1,4,5] The principle of endovascular repair of RAA is similar to the open approach: exclusion of the RAA without compromising renal blood flow and anatomy dictates whether a patient is an endovascular candidate. The largest series published comparing endovascular and open surgical approaches to RAA repair spanned between 1988 and 2011 and reported that endovascular repair was not independently associated with an improvement in mortality. A similar article echoed these findings; the authors found that endovascular RAA repair contributed to the increasing of overall RAA repair without decreasing open repair using statewide data.[1]

Similar to RAA repair, open and endovascular repairs are available for RAVF. The decision for either approach is determined by patient characteristics and anatomy. Traditional surgical approach includes ligation of feeding

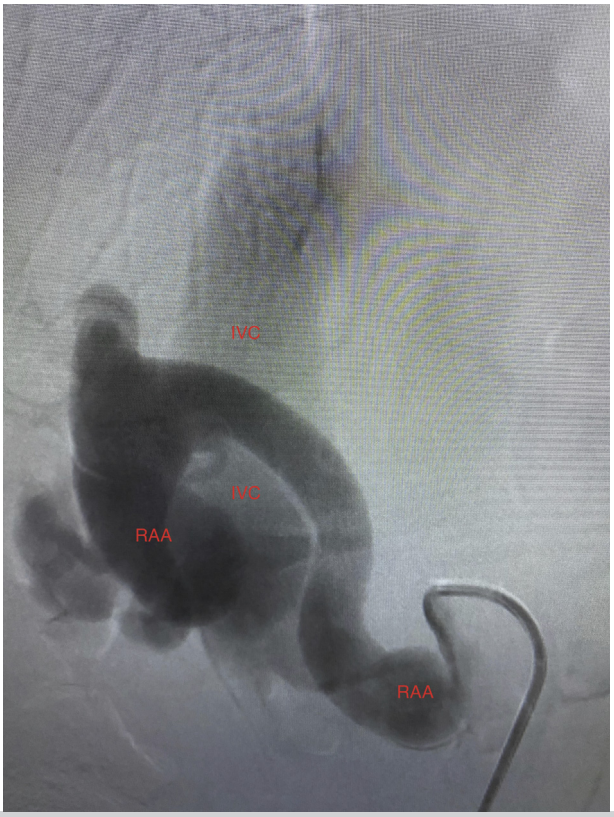

Fig 3. Right renal arteriogram showing a proximal renal artery aneurysm (*RAA*) and distal RAA with quick contrast filling of the inferior vena cava (*IVC*) indicating presence of a renal arteriovenous fistula (RAVF).

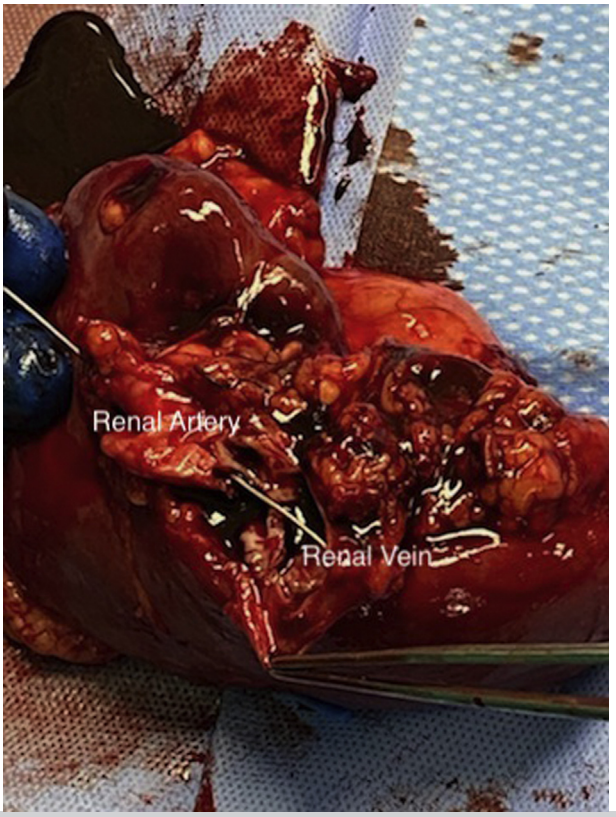

Fig 4. Intraoperative photo of the right kidney showing the fistulation between the renal artery and the renal vein.

artery to the RAVF followed by partial or total nephrectomy. Endovascular approach has gained popularity over time due to the morbidity of open surgery and preservation of renal parenchyma with endovascular techniques. Coil embolization is often used in small RAVFs and covered stents or plugs are described in larger AVFs.[3]

To our knowledge, concurrent ipsilateral RAA and RAVF are rare in literature with its earliest debut in 1975.[7] The first reported endovascular management of combined RAA and RAVF was published by Trocciola et al[8] in 2005; the authors reported successful embolization of feeding vessels of the 13-cm aneurysm without complications. In the past decade, numerous cases reported on endovascular approaches for these combined lesions involving stent graft (aneurysm <3 cm with associated RAVF) and implantation of patent ductus arteriosus occluder (aneurysm 6.3 cm in length with associated RAVF).[9,10] Despite these reported successes, open surgical repair continues to be the treatment of choice in patients with complex anatomy. The Table below summarizes open surgical repairs of combined RAA and RAVF reports from the past decade.[11-14]

Our case is unique in that our patient was asymptomatic until his development of a urinary tract infection; whether his symptoms were caused by his RAA/RAVF or urosepsis was unclear. However, he did not have pyelonephritis on his initial CTA. The etiology of his pathology was thought to be due to blunt trauma from the past; he did not have any invasive procedures of his kidneys, nor did he have any other arterial aneurysmal disease on his CTA aside from the RAA/RAVF. Although endovascular approach has shown similar outcomes for RAA repair as open approach, open surgical repair was indicated in our patient for multiple reasons. Neither CTA nor angiography provided a clear anatomical connection of his RAVF and, therefore, made endovascular embolization risky. Furthermore, the proximal RAA takeoff was at the aorta, and it was wide based, measuring about 2 cm, requiring a patch repair after ligation. Finally transplant surgery and urology were consulted regarding renal preservation; neither team believed the kidney was salvageable due to complex anatomy, especially at the hilum, involving two RAAs and RAVF.

CONCLUSIONS

We present a case of an RAA with a concurrent additional hilar RAA/RAVF. An open right nephrectomy, ligation of RAVF, and bovine patch repair of the aortic defect was performed. Currently, there is no consensus regarding the treatment protocol for a combined RAA

Table. Case reports of open surgical repair of a renal artery aneurysm (*RAA*) and renal arteriovenous fistula (*RAVF*) in adults in the last decade

Year	Authors	Symptoms	Anatomy	Procedure	Complications
2014	Okamoto et al.[11]	Heart failure	Two giant left RAAs (4.5 cm and 7.5 cm maximal diameter) with associated RAVF with renal vein. Tortuous and dilated renal artery.	Left nephrectomy	None
2015	Manogran et al.[12]	Rapidly enlarging RAA during pregnancy causing abdominal discomfort	A 10-cm right RAA with erosion into the right ovarian vein with formation of an RAVF.	C-section followed by right nephrectomy	None
2017	Franz et al.[13]	Hematuria, hypertension, palpitation, acute kidney injury	Enlarged RRA measuring 1.4 cm, RAA measuring 4.6 cm with direct fistulation to the renal vein.	Right nephrectomy	None
2018	Ivandaev et al.[14]	Initial presentation with abdominal discomfort. Postendovascular intervention with complications of thrombus in IVC and aneurysm rupture	Bilateral giant renal aneurysms of the lower renal artery with associated RAVF.	Right RAA treated with Amplatzer followed by thrombectomy of IVC, ligation of right lower renal vein and feeding arterial branch of fistula. Left RAVF and RAA treated with coil embolization followed by ex vivo RAVF ligation and RAA resection with autotransplantation	Occlusive thrombus and floating thrombus following right RAA and RAVF endovascular treatment. RAA rupture after left RAA and RAVF embolization. No complication after open procedures.

IVC, Inferior vena cava; *RRA*, right renal artery.

and RAVF. The approach is individualized and largely dependent on patient anatomy.

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