Radiology Case Reports 16 (2021) 2289–2294

Available online at www.sciencedirect.com

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journal homepage: www.elsevier.com/locate/radcr

Case report

Large renal arteriovenous fistula treated by embolization: a case report☆,☆☆

VT Duc, PhD, MD[a,b], NQT Duong, MD[a,b], NT Phong, MD[a], NH Nam, MD[a], DA Quoc, MD[c], TTQ Cuong, MD[b], NH Huy, MD[b], TL Duy, MD[b], PC Chien, MD[b,]*

[a] *Department of Radiology, University of Medicine and Pharmacy, Ho Chi Minh City, Vietnam*
[b] *Department of Diagnostic Imaging, University Medical Centre, Ho Chi Minh City, Vietnam*
[c] *Department of Pediatric Heart Surgery, University Medical Centre, Ho Chi Minh City, Vietnam*

ARTICLE INFO

Article history:
Received 21 May 2021
Accepted 23 May 2021

Keywords:
Renal arteriovenous fistula
Computed tomography
Digital subtraction angiography
Endovascular embolization
KONAR-MF occluder

ABSTRACT

Renal arteriovenous fistula (RAVF) is an uncommon vascular malformation of the kidney, which can be congenital, acquired or idiopathic. Although most patients are asymptomatic, RAVF can lead to hypertension, heart failure, renal insufficiency, hematuria, and progressive increase in size of renal vessels. Diagnosis is aided by radiological studies, with digital subtraction angiography as a gold standard. Besides, ultrasound with color Doppler and computed tomography angiography are noninvasive imaging techniques and can be useful for planning the treatment. A large fistula are generally treated by nephrectomy. Intervention can ameliorate the hemodynamic effects of high flow and to preserve the renal parenchymal function. Although endovascular therapy may be challenging due to the large size and high flow of fistula, this report describes a case of huge RAVF was successfully treated by embolization instead of surgery.

Renal arteriovenous fistula is a rare vascular anomaly, in which there is a direct connection between the artery and the vein of the kidney. RAVF can be classified into acquired (70%), congenital (25%) and idiopathic (3%–5%) [1]. Small RAVF usually has no symptoms, however a large RAVF with significant left-to-right shunt can cause hypertension (50%), high-output heart failure (32%), renal dysfunction and gross hematuria (21%) [2]. Previously, RAVF was treated by surgically ligating the renal artery with or without partial or total nephrectomy. Thanks to the advancement of endovascular therapy, transcatheter embolization is now considered an alternative treatment which is less invasive and helps preserve the kidney function and improve hemodynamics. However, it is controversial whether a large high flow fistula is best managed by surgery or by embolization. We report a case of a middle-aged woman diagnosed with a right spontaneous

☆ Acknowledgments: We would like to thank the patient and her family for the permission to publish this report.
☆☆ Competing of interest: None.
* Corresponding author.
E-mail address: chien.pc@umc.edu.vn (P. Chien).
https://doi.org/10.1016/j.radcr.2021.05.058

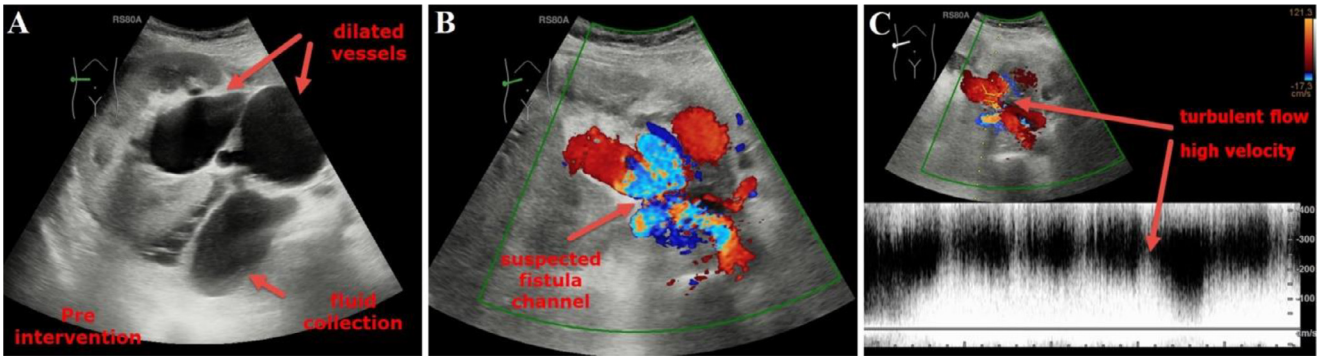

Fig. 1 – A-C On gray-scale ultrasound (A, long arrow), there are some cystic lesions in renal pelvis, which have mosaic patterns on color Doppler (B, arrow). Spectral analysis shows turbulent flow with high velocity and low resistance (C, arrows)

RAVF having large size and high flow. This case demonstrates that good results may be achieved with endovascular intervention using occluder.

Case report

A 45-year-old female patient was admitted to University Medical Center due to her incidental findings of renal arteriovenous fistula at the previous hospital. She has been diagnosed with hypertension three months ago. She had no history of trauma, biopsy or abdominal surgery. She was thin and pale. Vital signs were normal. Abdominal examination demonstrated a pansystolic murmur and a thrill in the right upper quadrant. The kidneys were not palpated. Results of her complete blood count were red blood cells 4.39 T/L, hemoglobin concentration 88 g/L, hematocrit 29% and platelets 197 G/L. Serum creatinine 0.75 mg/dl, serum urea 29.96 mg/dl and estimated glomerular filtration rate was 83 mL/min. Coagulation tests were within the normal range. There were erythrocytes in her urine, approximately 200 cells/μL. Subsequently, she underwent chest X-ray, echocardiography, abdominal ultrasound and abdominal CT scan.

Chest X-ray showed normal heart and lung parenchyma. Echocardiography showed normal left ventricular systolic function with ejection fraction 62%, and moderate mitral regurgitation. On gray-scale ultrasound, her right kidney had some anechoic cystic lesions with heterogeneous fill-in on color Doppler, which was confirmed to be aneurysmal dilatations. There was a turbulent flow with maximum velocity of 250 cm/s on spectral analysis, which was suggestive of a fistula (Fig. 1).

Contrast-enhanced abdominopelvic CT in the arterial phase demonstrated several dilated branches of right renal vein, which enhanced almost as bright as renal artery. These findings are indicative of a RAVF. There was a fistula between an interlobar branch of the right renal artery and a branch of the right renal vein, which later formed three intermittent fusiform aneurysms. The largest diameter of these aneurysms was 4.6 cm. They compressed the inferior vena cava and pushed the pancreas head forward. Additionally, there were some low-density fluid collections in the perirenal and posterior pararenal space (Fig. 2).

The patient underwent endovascular intervention to occlude the fistula. The sheath was inserted into right common femoral artery by using Seldinger technique. The Pigtail catheter was then placed into the abdominal aorta for angiography showing arteriovenous fistula at the 1/3 lower portion of the right kidney, which was compatible with abdominal CT findings (Fig. 3). The ConcierGE Guiding 5F catheter was placed selectively via the right renal artery to the narrowest site of the fistula which was just before the junction between the feeding artery and the first aneurysmal vein. Then a KONAR-MF 8-6 occluder (discs' diameter of 12 mm, two waists' diameter of 8–6 mm, waist 's length of 4 mm) was released there. Angiography obtaining at the origin of right renal artery via ConcierGE Guiding showed no contrast filled the dilated renal veins (Fig. 4). Follow-up ultrasound after one month revealed completely thrombosed aneurysms and normal flow of renal artery and vein (Fig. 5). CT scan after one month and six months showed that embolizing material was in the correct position and dilated renal veins shrunk and had no contrast in the delay phase (Fig. 6). There was no evidence of renal parenchymal infarction.

Discussion

Renal arteriovenous fistula was first reported by Vorela in 1928 [2]. The incidence was approximately less than 0.04%. The disease is comprised of acquired type 70%, congenital type 25% and idiopathic type 3-5% [1],[3]. The recent development of percutaneous intervention leads to the increased incidence of acquired RAVF [4]. Acquired RAVF can be secondary to renal intervention (biopsy, surgery or transplant), injury, inflammation or tumor [5]. The exact cause of congenital RAVF is still unknown, yet this condition is believed to have been present since birth [6]. Congenital fistula often leads to hematuria. Its typical manifestations are multiple varicose vessels, while the acquired and idiopathic types usually present with a simple aneurysm with one feeding artery [7]. Idiopathic RAVF occurs in the absence of triggers and is most common in middle-aged

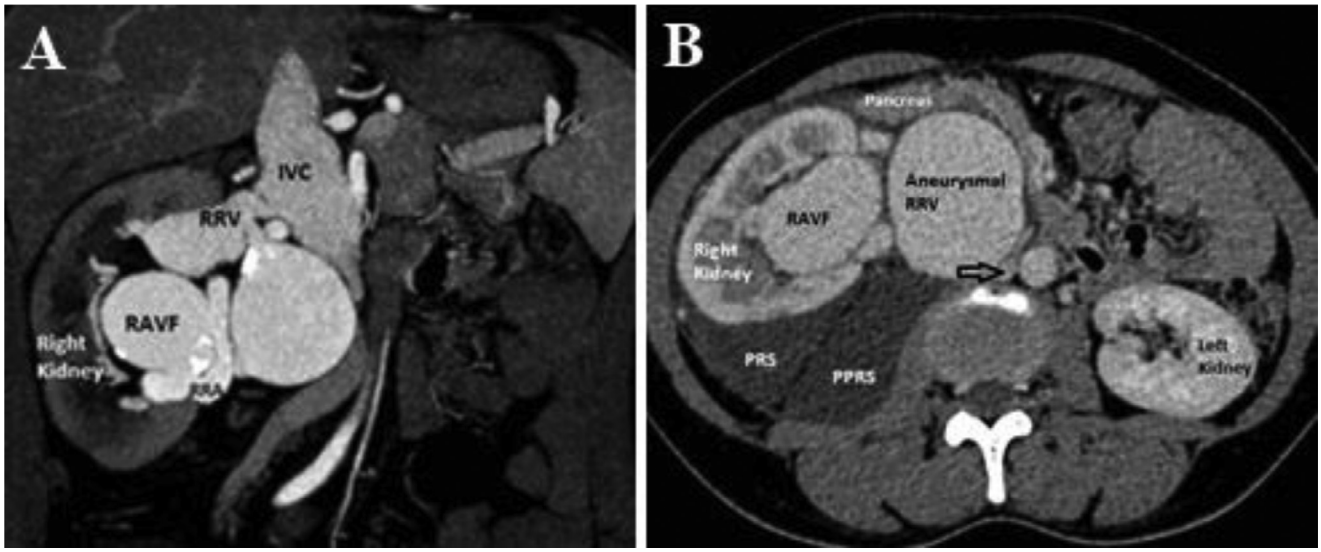

Fig. 2 – A-B Coronal (A) and Axial (B) contrast-enhanced CT show the early opacification of aneurysmal right renal vein (RRV) in the arterial phase, indicating a RAVF with dilated right renal artery (RRA). RAVF pushes the pancreas head forward and compresses the inferior vena cava (arrow). Note the low-density fluid collections in the right perirenal space (PRS) and posterior pararenal space (PPRS)

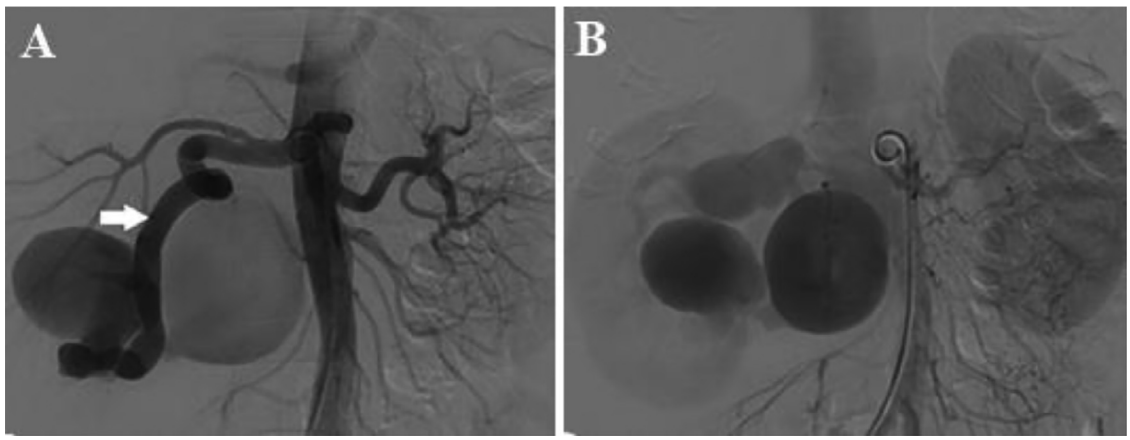

Fig. 3 – A-B. Angiogram shows the arteriovenous fistula at the 1/3 lower portion of the right kidney

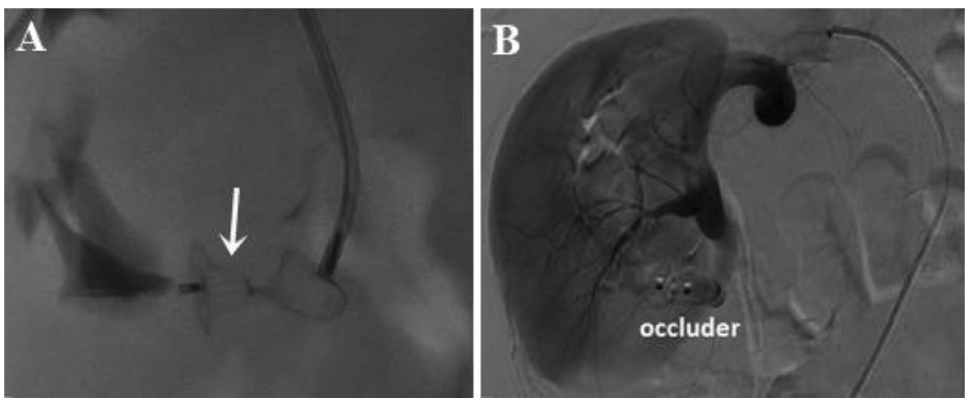

Fig. 4 – A-B A KONAR-MF plug (arrow) was released into the dilated branch feeding the fistula (A). Angiogram shows no contrast filled the dilated renal veins after releasing the plug (B)

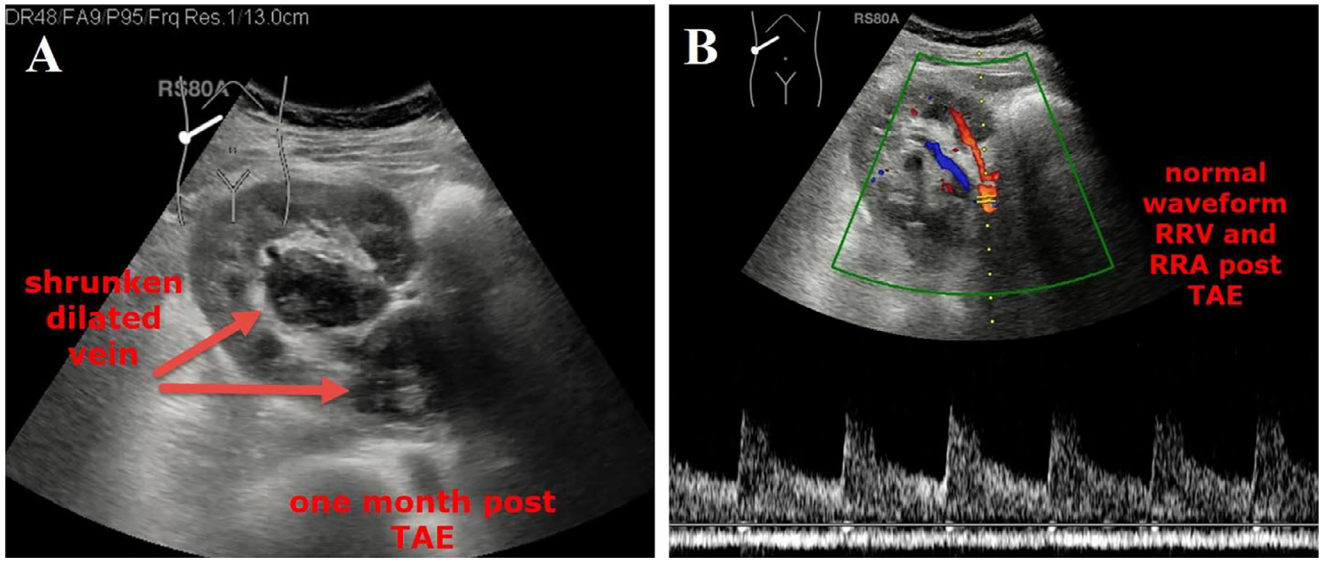

Fig. 5 – A-B After one month of trans-arterial embolization (TAE), abdominal ultrasound demonstrates shrunken dilated veins containing thrombus (A) and normal waveform of right renal artery (RRA) (B) and vein (RRV) (not shown)

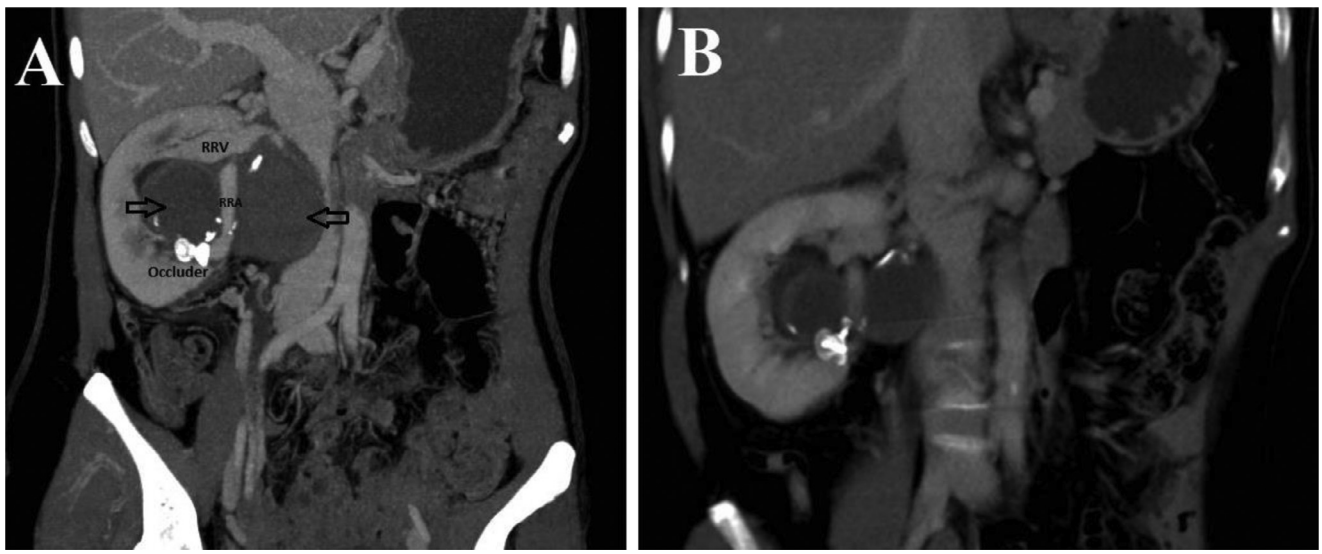

Fig. 6 – A-B Coronal CT after one month (A) and six months (B) of follow-up shows complete exclusion and decreased size of fistula in the delay phase of contrast injection (arrow)

patients (peak between 30 and 40 years [6]). It often presents with cardiovascular symptoms such as hypertension or heart failure. It is hypothesized that idiopathic RAVF is caused by the erosion of an aneurysm of an intraparenchymal renal artery into an adjacent vein [8]. In idiopathic RAVF, the right kidney is more likely to be involved, and females are affected twice as compared to males, similar to the presented case [9]. Idiopathic and congenital fistulas are often grouped together as spontaneous RAVF [10]. There were no predisposing factors recognized in this case, therefore the diagnosis of idiopathic RAVF could be made.

The common symptoms and signs associated with RAVF are abdominal bruit (100%), hypertension and cardiomegaly (50%), congestive heart failure (32%), and gross hematuria (21%) [2]. Laboratory and imaging studies should focus on renal, cardiac and hemodynamic complications. In our case, the patient presented with well-controlled hypertension, physical examination showed abdominal thrill and murmur. She had moderate anemia and normal kidney function tests. No clinical or sonographic signs of heart failure was noted.

Imaging studies play a pivotal role in the diagnosis and treatment of RAVF [4]. Doppler ultrasound should be the one of the first imaging studies. On gray scale ultrasound, the aneurysms can be seen as anechoic cyst-like lesions. Color Doppler ultrasound reflects the mosaic pattern filling in these cystic lesions. Spectral analysis shows increased flow velocity,

decreased arterial resistance, and arterial wave forms in the draining vein [5]. Doppler ultrasound is useful in evaluating renal cystic lesions, for the differentiation between a simple or complicated cyst and vascular pathologies, and for showing the vascularization of the septae or the solid component of the cystic lesion [5]. DSA is the gold standard for diagnosing and evaluating RAVF, however it is invasive. Multi-slice CT allows the evaluation of lesions on several different reconstructed planes. Thanks to the short acquisition time, the dynamics of AVF can be easily evaluated which leads to an accurate diagnosis. CT findings of RAVF are dilated arteries, early opacified draining veins, dilated veins due to high return flow. In addition, CT helps in the assessment of location and size of the fistula, which is helpful for treatment planning [4]. In the aforementioned case, physical examination with signs of abdominal murmur and thrill was consistent with previous diagnosis in the local hospital. She underwent CT scan pelvic abdominal scan without performing a prior abdominal ultrasound. The CT images show a large RAVF in the right kidney with typical features such as a dilated feeding artery, dilated and early opacified draining veins. In addition, there is low-attenuation fluid in the perirenal space, suggestive of stagnation and increased pressure on renal vein lumen returns due to high supply.

Indications for treatment include progressive increase in fistula size, recurrent or persistent hematuria, hemodynamic effects associated with the fistula, particularly circulatory overload, hypertension and high-output heart failure [5]. The possibility of cardiac failure, kidney failure and rupture of dilated veins warranted a multidisciplinary consultation meeting to decide which treatment was the most appropriate. Surgery and endovascular intervention were both considered. The purpose of RAVF treatment is to preserve renal function and eliminate fistula-related symptoms and hemodynamic [5]. Previously, surgically constricting renal arteries with or without partial or total nephrectomy was the main treatment. Embolization is now the preferred option due to the presence of fewer complications and the ability to preserve kidney function. However, large size and high flow complicate endovascular therapy due to the possibility of incomplete occlusion and migration of embolizing material [4]. Therefore, the surgical approach should be considered if endovascular intervention seems unfavorable.

To avoid a major surgery in this woman, after discussion with her, we decided to performed embolization as first-line treatment. Transcatheter embolization of RAVF was first described by Bookstein and Goldstein in 1973. This could be a definite procedure or a palliative one to reduce the size of the fistula for further more limited surgery [11]. The materials for embolization include gel foam sponge, steel coils, cyanoacrylate, detachable silicone balloons, and covered stents [12]. However, these are too small for huge fistulas, being at risk of dislodgment and passing through the draining vein into the pulmonary or other vessels. In addition, normal perfusing vessels might be injured in the intravascular interventional procedure [11]. We embolized the fistula by using a occluder with its diameter larger than the feeding branch, with no risk of distal embolism. KONAR-MF is usually used for transcatheter treatment of ventricular septal defects. It is a self-expandable device made from double nitinol wire mesh layers. It is comprised of two discs linked together by a cone-shaped waist [12]. These two discs have the same diameter which can be up to 18 mm. It is still retrievable before detachment from the delivery catheter if the plug is put incorrectly in the fistula.

Conclusion

RAVF is an uncommon vascular anomaly. Hematuria, hypertension, heart failure, abdominal murmur and thrill are common symptoms and signs in patients with large fistula size, with or without a history of abdominal injury or renal intervention. Color Doppler ultrasound and computed tomography are non-invasive tools which are useful for diagnosis and treatment planning. Cardiac and renal complications should be carefully evaluated. Endovascular intervention should be the preferred treatment due to its effectiveness, non-invasiveness, and conservation of kidney function. Surgery is considered when the fistula is too large or when endovascular intervention fails.

Consent statement

The protocol was reviewed and approved by the Human Research Ethics Committee of the University of Medicine and Pharmacy at Ho Chi Minh City. The study was performed in accordance with the Declaration of Helsinki.

Patient consent

The patient wrote informed consent.

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