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Case Report

Percutaneous intervention for giant renal extrarenal aneurysm and arteriovenous fistula ^{☆,☆☆}

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ABSTRACT

Arteriovenous fistula is a high-flow vascular malformation characterized by a direct connection between an artery and a vein without the presence of a nidus (a network of arterial and venous channels). Renal artery aneurysm concomitant with a renal arteriovenous fistula (RAVF) is extremely rare. We report the case of a 45-year-old woman with a giant renal artery aneurysm combined with RAVF who presented with gross hematuria. The patient was successfully treated by embolization of a giant extrarenal aneurysm and RAVF using coils and a plug device. RAVF can lead to harmful complications, and treatment is necessary upon diagnosis. Endovascular embolization is a feasible, efficient, and safe method able to retain maximal normal renal function

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Introduction

Renal arteriovenous fistula (RAVF) is a rare lesion with a reported prevalence of below 0.04% [1]. Renal artery aneurysm concomitant with an RAVF is extremely rare and has considerable clinical impacts on hypertension, hematuria, abdominal pain, and high-output cardiac failure. Arteriovenous malformations can be classified as acquired, idiopathic, or congenital. Recently, percutaneous intervention embolization has be-

come the first-line treatment option for arteriovenous malformations [2]. Endovascular treatment is a less invasive alternative to traditional open surgery [3,4]. In this study, we aimed to describe a patient who presented with extensive hematuria and was diagnosed with a massive renal artery aneurysm and high-flow RAVF. The massive extrarenal aneurysm and RAVF were effectively embolized with coils and a plug device. We present the following case in accordance with the CARE reporting checklist.

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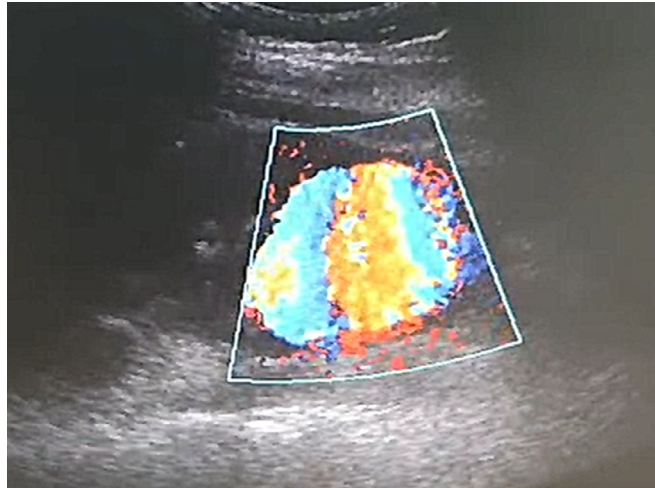


Fig. 1 – Color Doppler longitudinal plane showed a high-velocity, turbulent aneurysm in the left inferior renal pole.

Case presentation

A 45-year-old woman was admitted to our hospital with gross hematuria. The patient's medical history revealed hypertension for the prior 5 years (with blood pressure readings as high as 180/100 mm Hg, indicating poor control). A left RAVF was identified by color Doppler ultrasonography 7 years prior to admission. The patient reported occasional lower back pain and a small amount of blood in her urine. The patient's past medical history was unremarkable, but a family history of hypertension was described. The patient had no history of abdominal trauma, renal biopsy, or interventions involving the renal vascular system.

On the morning the patient presented to our hospital, the patient described experiencing severe pain in the left lumbar region and presented with massive hematuria and the passage of blood clots in urine. At the time of presentation, blood pressure was 105/60 mm Hg, and heart rate was 100 beats/minute. She was pale, with a soft abdomen, and a palpable mass was identified in the left lumbar region. Blood chemistry analysis showed hemoglobin at 7.4 g/dl (normal range: 12.0 to 16.0 g/dl), hematocrit at 22.4 % (normal range: 36% to 46%), and red blood cells at 2.7 T/l (normal range: 4.0 to 5.2 T/l). Other biochemical and coagulation parameters were within normal limits. Urine analysis revealed a large number of red blood cells. Ultrasound of the abdomen and abdominal computed tomography (CT) imaging were advised.

Abdominal ultrasonography revealed the right and left kidney diameters as 101 and 98 mm, respectively. Color Doppler sonography showed a vascular malformation in the left inferior renal pole. A giant extrarenal aneurysm measuring 55 × 65 mm was observed. Spectral Doppler tracings showed high-velocity, low resistance flow within the artery, and turbulent, pulsatile, arterialized flow was detected in the segmental draining vein (Fig. 1).

Abdominal CT images showed a giant left renal artery aneurysm (Fig. 2). The results of abdominal 3-dimensional CT imaging with contrast enhancement are presented in Fig. 3. A

huge, ball-shaped renal artery aneurysm developed outside of the anterior bottom region of the left kidney. The maximum diameter of the abnormal vascular mass was 65 mm. The interlobar renal artery was dilated and tortuous (measuring approximately 10 mm), and the draining vein was enlarged to approximately 19 mm.

Other diagnostic imaging tests were performed. Chest X-ray showed cardiomegaly with a cardiothoracic ratio of 55% and no pleural effusion. Electrocardiograms showed sinus rhythm without left ventricular hypertrophy (RV5 + SV1 amp = 3.0 mV). Transthoracic 2-dimensional echocardiography indicated mild mitral regurgitation. The left ventricle was dilated, with an end-diastolic diameter (LVDd) of 58 mm. The left ventricular mass was 192 g (calculated using the equation by Devereux). Body surface area was 1.47 m². The left ventricular mass index (normalized for body surface area) was 130 g/m². Ventricular wall motion was normal. Echocardiography showed a left ventricular ejection fraction of 68%. The pulmonary artery pressure was 15 mm Hg.

The patient was transfused with 2 additional blood units (blood type O⁺), which stabilized the patient's hemodynamic status. The patient was diagnosed with a giant renal artery aneurysm combined with high-flow RAVF and massive hematuria. After consulting a team of interventional doctors and surgeons, we decided to manage the RAVF with embolization.

A 6F diagnostic Judkin's right (JR) catheter (Terumo Inc., Tokyo, Japan) was placed using the Seldinger technique through the right brachial artery under local anesthesia. Selective angiogram performed on the left renal artery showed an enlarged left interlobar renal artery branch with increased blood flow velocity and volume. This artery drained directly into a giant renal aneurysm, forcing blood into the drainage vein, the left renal vein, and the inferior vena cava (Fig. 4). The left renal artery and vein displayed almost simultaneously, and the feeding artery leading to the fistula was uniformly dilated and tortuous. The left renal artery was engaged with a Fortress 6F 100 cm guide catheter (Biotronik Inc., Germany) accessed through the right brachial artery. We attempted to push the catheter into the interlobar renal artery; however,

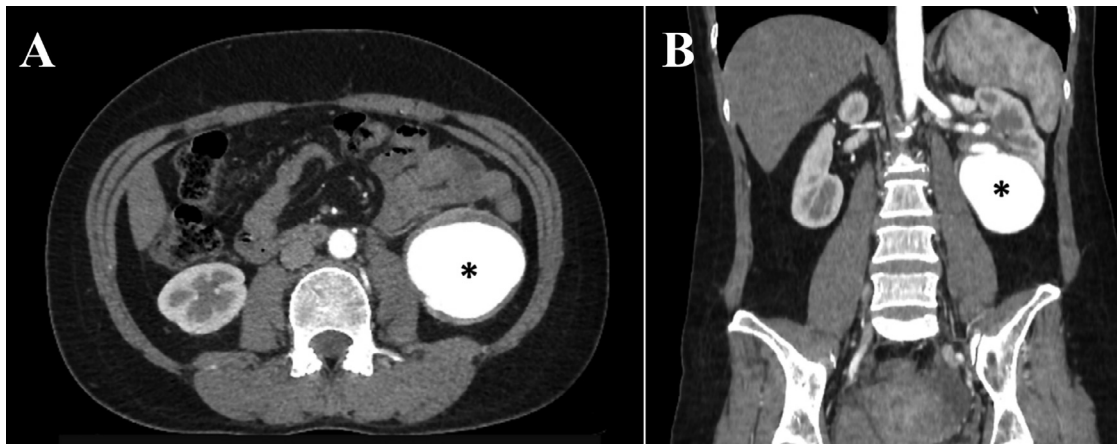


Fig. 2 – Abdominal CT images (A) axial plane and (B) coronal plane showed a giant left renal artery aneurysm (asterisk).

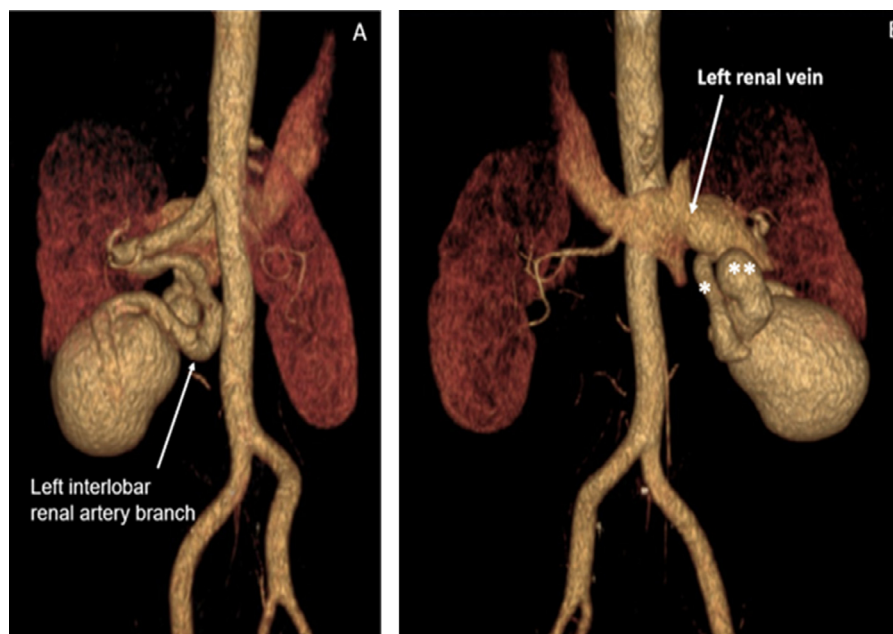


Fig. 3 – Abdominal 3-dimensional computed tomography scan. A: Posterior view showing the left interlobar renal artery branch (arrow). B: Anterior view, showing the left renal vein (arrow). (*: Left interlobar renal artery branch, **: Left interlobar renal vein branch).

the left interlobar renal artery was very tortuous, preventing catheter insertion. We used both J-Tip 0.035-inch (Terumo Inc., Tokyo, Japan) and stiff J-Tip 0.035-inch (Terumo Inc., Tokyo, Japan) guidewires to support the catheter. Finally, a Catheter Fortress was able to approach the aneurysm. A 20 mm Amplatzer vascular plug II (Abbott Inc., USA) was introduced using a delivery cable passed through the delivery sheath to the mouth of the giant aneurysm. We confirmed the proper positioning and landing zone using non-selective renal contrast injection through the 6F JR catheter. The vascular plug was deployed in the left segmental renal artery. Post-deployment contrast injection showed mild flow in the aneurysm; therefore, we opted to supplement the plug with endovascular coils. A separate microcatheter was placed at

the front of the vascular plug, and 2 Ruby Coils (16 mm × 60 cm and 20 mm × 60 cm; Penumbra Inc., California, USA) were placed. After 10 minutes, selective left renal contrast injection showed total occlusion of the left interlobar renal artery, with no flow in the aneurysm (Fig. 5). Angiography demonstrated a near-complete nephrogram with excellent peripheral flow.

After the procedure, the patient was monitored postoperatively for 2 days. She presented with no postoperative symptoms, including the absence of chills and fever. After 2 days, no signs of hematuria were detected, and no obvious abnormalities were observed on urinalysis or renal function tests (creatinine 56 $\mu\text{mol/L}$; normal range 45 to 84 $\mu\text{mol/L}$). Color Doppler ultrasonography showed no blood flow signals in the



Fig. 4 – Selective angiogram of the left renal artery.
Arrow: Left interlobar renal artery branch; dashed line: renal artery aneurysm.



Fig. 5 – Selective angiogram of the left renal artery after the procedure.
Arrow: Two Ruby Coils and one vascular plug II were inserted.

vasculature of the RAVF. The patient's symptoms of lumbago gradually subsided after 2 weeks.

Discussion

Arteriovenous fistulas are high-flow vascular malformations characterized by the presence of a direct connection between an artery and a vein without the presence of a nidus (a network of arterial and venous channels) [5]. RAVFs are classified as congenital (14%–27% of cases), idiopathic (3% of cases), or acquired (70%–80%) [6]. Some frequent causes of acquired

RAVFs include needle biopsy, injury, surgery, inflammation, infection, and malignant tumors. The increasing use of interventional procedures, such as percutaneous needle biopsy and percutaneous nephrostomy, has been accompanied by the increasing incidence of iatrogenic fistulas. In our patient, no clinical evidence indicated the development of iatrogenic or traumatic arteriovenous fistula, and the lesion was classified as congenital or idiopathic [3].

The clinical manifestations of arteriovenous fistulas depend on their localization and size. The most frequent clinical signs are hematuria, mild tachycardia, hypertension, and symptoms of congestive heart failure. A bruit is less frequently detected for cirroid arteriovenous malformations, whereas

hematuria is more common in congenital malformations [7]. Increased cardiac output and hypertension lead to the development of cardiac failure in approximately 40% of patients with an arteriovenous fistula, and the severity of symptoms and hemodynamic changes depend on the blood flow through the fistula. In our patient, congestive heart failure was not present; however, the left ventricle was dilated, and the left ventricular muscle mass index increased, indicating volume overloading, suggesting that future congestive heart failure should be considered [4].

Gross hematuria is the primary complaint reported in 72% of RAVF cases. Hematuria results from minute ruptures of the thin-walled veins caused by increased pressure, allowing blood to flow into the collecting system. Therefore, even small, peripherally located arteriovenous malformations can lead to massive hematuria [8].

Many studies have indicated the effectiveness of color Doppler sonography for diagnosing and following patients with RAVF. However, multi-slice CT or magnetic resonance imaging is essential for determining an appropriate treatment strategy, and a definite diagnosis of arteriovenous malformations requires confirmation by angiography [9].

The current treatment options for arteriovenous malformations include surgical approaches and interventional embolization. However, surgical approaches, such as nephrectomy or renal artery ligation, are traumatic and fail to retain functional nephrons. In recent years, the interventional embolization of arteriovenous fistulas using elastic coils, gelatin sponge particles, and polyvinyl alcohol particles has become a feasible, efficient, and safe method for successfully occluding fistulas while retaining maximal renal function. However, for large, high-output RAVFs, emboli can shift to unexpected areas, and these cases are generally indicated for surgical treatment. The choice of embolization material depends on the anatomical characteristics of the involved blood vessels and the experience of the interventional physician. The risks of embolotherapy include reflux of obliterating agents, the loss of normal renal parenchyma, and pulmonary embolism. Mori et al. reported that a postembolization syndrome might occur, characterized by pain in the embolized area, nausea, vomiting, and fever lasting up to 5 days [10].

Conclusion

RAVF can lead to harmful complications. Treatment of this pathology should be recommended upon diagnosis. Endovascular embolization is a feasible, efficient, and safe method that can maximize the retention of normal renal function.

Availability of data and materials

Data sharing is not applicable to this article as no datasets were generated or analyzed during the current study.

Authors' contributions

Nguyen The Huy and Phan Thao Nguyen contributed equally to this article as co-first authors. All authors read and approved final version of this manuscript.

Ethics approval

Not applicable.

Patient consent

Written informed consent was obtained from the patient for the publication of patient information in this article.

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