



Endovascular Treatment of a Giant Renal Artery Aneurysm with High-Flow Renal Arteriovenous Malformation

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Renal artery aneurysms (RAAs) are rare lesions with a prevalence of less than 1% in the general population. Renal arteriovenous malformations (AVMs) are rare lesions with an estimated incidence of less than 0.04%. The coexistence of these two clinical entities is extremely rare and narrows the available treatment options by endovascular or open surgery. We describe a case of a giant symptomatic RAA type III, which was combined with a high-flow renal AVM in the right kidney. Using two vascular plugs, the RAA was excluded successfully. The perfusion of the right kidney's lower pole was preserved by implantation of two covered stents in the inferior segmental renal artery.

Key Words: Aneurysm, Endovascular procedures, Arteriovenous malformations, Arteriovenous fistula, Renal circulation

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INTRODUCTION

Renal artery aneurysms (RAAs) are rare vascular lesions. Although their true incidence and natural history remain elusive, the prevalence of RAAs in the general population is approximately 0.09% [1]. Renal artery arteriovenous malformations (AVMs) with the high flow can be idiopathic, congenital, or acquired and are also rare, with an estimated incidence of less than 0.04% [2,3]. The co-existence of RAA with AVM is an extremely rare condition that complicates the natural history of the disease, the clinical presentation, and certainly the operational treatment options [4].

We present a rare case of a symptomatic RAA (50 mm in

diameter), combined with a high-flow AVM, which originated at the right renal hilum and in the upper two-thirds of the kidney, occupying its parenchyma. Endovascular treatment was successfully performed; the RAA was excluded with vascular plugs. The perfusion of the lower pole was preserved with the implantation of covered stents in the inferior segmental branch of the renal artery.

The study was approved by the Institutional Review Board of the “G. Gennimatas” Hospital, Aristotle University of Thessaloniki (IRB no. 43/21) and the informed consent was obtained from the patient.

CASE

A 55-year-old female presented to the emergency department with complaints of acute right flank pain radiating to the anterior abdomen and pelvis. Past medical history included two childbirths and was negative for any abdominal or systemic disease, trauma, surgery, biopsy, vascular disease, or intervention. On presentation, her blood pressure was 155/82 mmHg, and her heart rate was 95 bpm. Physical examination revealed a right abdominal bruit and tenderness at the right costovertebral angle (Giordano sign), which indicated a potential involvement of the right kidney. Laboratory studies, including a basic metabolic panel, complete blood count, urinalysis, and cardiac enzymes, resulted within the normal range. The estimated glomerular filtration rate (eGFR) was 90 mL/min/1.73 m². Imaging with computed tomography angiography (CTA) showed the presence of a giant, fusiform aneurysm (50 mm in diameter) of the right renal artery located at the hilum, which occupied almost entirely the upper two-thirds of the renal parenchyma (Fig. 1). Additionally, a very high-flow AVM coexisted, which showed rapid clearance of the iodinated contrast medium to the inferior vena cava (IVC). This suggested the presence of direct communication between the renal arterial flow and IVC through the AVM. The right renal artery was slightly elongated and severely dilated at 13 mm in diameter, while the diameter of IVC at that point was 33 mm. The functioning lower pole was perfused by a 4-mm inferior segmental artery, which originated from the renal artery main trunk before the renal hilum and the RAA (Fig. 2). In addition, two small AVMs in the liver were found. The patient remained hemodynamically stable, the

flank pain relieved in 24 hours without painkillers, and the patient was discharged from the hospital with the explicit indication of the impending operative treatment. Due to the rarity of the disease and the lack of previous experience in this condition, the patient initially denied the proposed treatment options. However, ten days later, after two episodes of recurrent milder flank pain, the patient was readmitted to our department.

The patient was treated entirely by endovascular means, under local infiltration anesthesia of the access sites with Xylocaine 1% and Ropivacaine hydrochloride 7.5% solution. We used percutaneous access from both common femoral arteries and other access from the left brachial artery with cutdown. Three sheaths, Super Arrow-Flex (Teleflex, Morrisville, NC, USA), were inserted; two 8-Fr, 45-cm long from the femoral approaches and an 8-Fr, 80-cm long from the left brachial approach. Selective catheterization of the right renal artery was achieved from the right common femoral access and the brachial approach using 0.035 inch hydrophilic Radifocus guidewires (Terumo Europe NV, Leuven, Belgium) and 5-Fr headhunter angiographic catheters. Hyper-selective catheterization of the right renal inferior segmental artery (RISA) from the brachial approach was achieved with the same equipment (Fig. 3). With both sheaths in place, we excluded the RAA using two Cera vascular plugs (Lifetech Scientific Corp., Shenzhen, China), 20 and 16 mm in diameter. Both vascular plugs were deployed at the distal end of the renal artery main trunk, just before the aneurysm formation. Based on preoperative CTA measurements, the perfusion of the RISA was preserved with implantation of two 6-mm Viabahn VBX covered stents (W. L. Gore & Associates, Flagstaff, AZ, USA) with lengths of 79

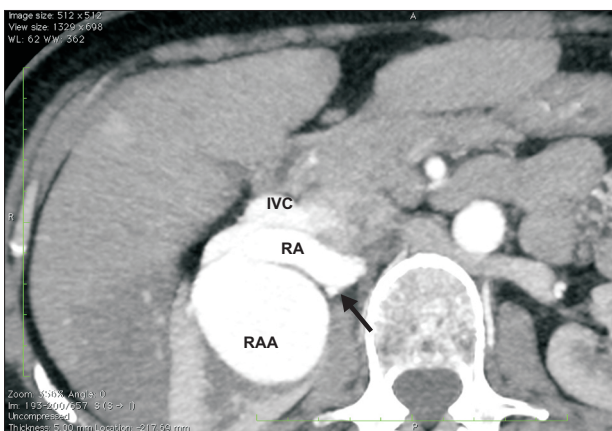


Fig. 1. The axial view of computed tomography angiography showed the huge renal artery aneurysm (RAA) and the relation with the renal artery (RA) and inferior vena cava (IVC). The black arrow showed the orifice of the inferior segmental renal artery.



Fig. 2. Preoperative computed tomography angiography with 3-dimensional reconstruction showed the right kidney and the RAA with high flow renal artery arteriovenous malformation. The arrow showed the inferior segmental renal artery.

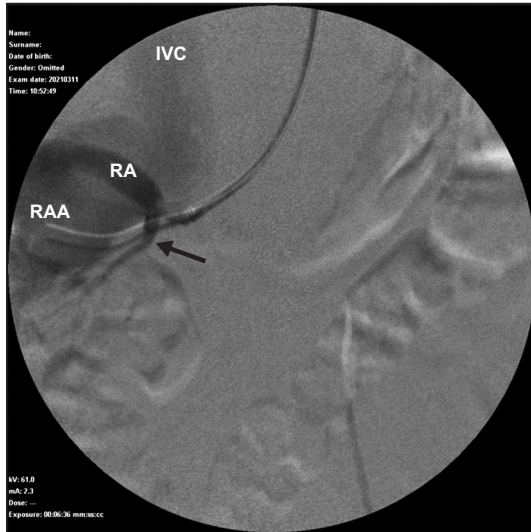


Fig. 3. Intraoperative selective angiography showed the renal artery (RA), the renal artery aneurysm (RAA), and the inferior vena cava (IVC). The black arrow showed the orifice of the inferior segmental renal artery.

mm and 59 mm. Intraoperatively, the implantation of the second vascular plug and a covered stent was considered necessary to ensure the complete exclusion of the flow in the RAA and the adequate perfusion in RISA. Stabilization of sheaths during the advancement of vascular plugs and covered stents was achieved using an inflated aortic occlusion balloon catheter at the juxtarenal aortic segment. Perioperative medication included an intravenous bolus administration of 2,500 units of heparin and cefoxitin (1 g). The postoperative clinical course included mild flank pain and fever for three days, with the highest at 38.3°C and a declining course. Additionally, we observed a drop of hemoglobin by 1 g/dL and an elevation of white blood cells at 23,000/ μ L. During the postoperative hospital stay, the patient received Cefoxitin (1 g) bidaily and thromboprophylaxis with tinzaparin (50 IU anti-Xa/Kg) and long-life single antiplatelet medication with aspirin (100 mg) every day. The patient was discharged on the third postoperative day. Although the final intraoperative angiogram showed a cloudy and potentially porosity related enrichment of RAA and IVC with a contrast medium, the follow-up imaging with CTA at six months confirmed the successful exclusion of the RAA and AVM, the preservation of flow in covered stents, and the adequate perfusion of the lower pole of the right kidney (Fig. 4). At six months, clinical and laboratory evaluations were normal, and eGFR was 97 mL/min/1.73 m².

DISCUSSION

The incidence of RAA in angiographical studies has been

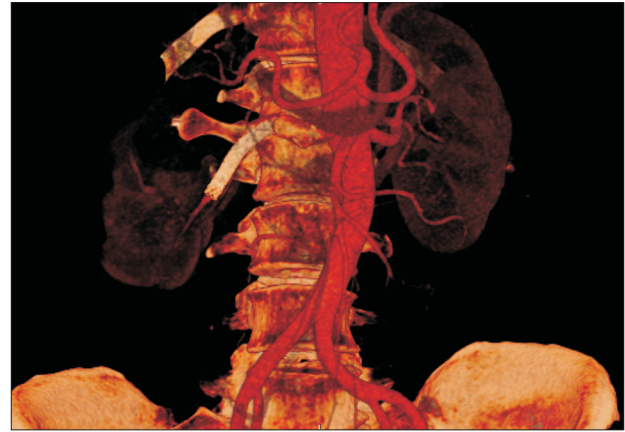


Fig. 4. Postoperative computed tomography angiography with 3-dimensional reconstruction showed the successful exclusion of the renal artery aneurysm with renal artery arteriovenous malformations and the preservation of flow in the inferior segmental renal artery.

estimated to be between 0.3 to 1%. The definition of an RAA is based on the artery's dilatation twice the diameter of a normal renal artery, and symptomatology includes one or more of the following: flank pain, hematuria, renal emboli, infarction, and renovascular hypertension [5,6]. According to the literature, the rupture of asymptomatic RAAs outside the pregnancy period is extremely rare, and for this reason, the suggested treatment is conservative [7]. However, these data were based mainly on mid-term follow-up of small RAAs, with a mean diameter of 20 mm, and considering the age of these patients, the operative treatment is indicated in RAAs larger than 25 mm. In giant or symptomatic RAAs, especially if a high flow AVM exists, the risk of an imminent rupture is very high, and urgent surgical repair is fully advocated [8-10]. Two types of renal AVMs have been recognized, the cirroid with multiple small and dilated arteriovenous communications and the aneurysmal type, which consists of a single arterial communication with a solitary vein. Cirroid type is a congenital AVM and represents a focal spontaneous vascular development failure during the 1st trimester of gestation [9]. Renal AVMs develop mainly in females in the third or fourth decade of life, with a 3-fold greater prevalence compared to males. The right kidney is more often affected than the left, and renal AVM may be associated with other genetic vascular developmental disorders [11,12]. Aneurysmal high flow AVMs are either congenital idiopathic in etiology or secondary to a previous intervention or trauma. In congenital idiopathic RAVMs, the pathogenetic mechanism for shunt formation between the adjacent segments of the renal artery and vein could be the erosion of a pre-existing RAA into the adjacent vein [9,11,13]. However, an inverted hypothesis could be that the solitary

congenital RAVM produces tremendous local hemodynamic shear stress, which results in the RAA formation and significant enlargement of both the RAA and RAVM through time.

Both these two rare conditions, the RAA with RAVM, coexisted in our case. In the absence of any history of trauma or previous interventions, it is rational to assume that the congenital RAVM developed first. It affected the renal arterial wall and gave rise to the RAA and, consequently, its growth to the remarkable diameter of 50 mm. This is further supported by the concomitant presence of vascular malformations in the liver.

The traditional classification of RAA was introduced by Poutasse in 1965 [14]. However, a simplified classification system was recently proposed by Rundback et al. [15], which described three types of RAAs and facilitated the decision-making regarding the operational treatment options [15]. Type I is a saccular aneurysm arising from the main renal artery or a proximal large segmental artery and represents the ideal place for endovascular treatment with covered stenting with or without adjacent embolization [8,10,16]. Type II is fusiform aneurysm involving the renal artery bifurcation. In selected cases, the endovascular treatment may be feasible with the unavoidable sacrifice of smaller renal branches or flow-diversion multilayer stenting. However, in most type II cases, open repair may be indicated. Open repair techniques include in situ excision and bypass (REB), ex vivo repair and auto-transplantation (RAT), and nephrectomy. Finally, type III RAAs are located at the hilum or intraparenchymal, and the open repair techniques have the absolute indication [9,10,17,18]. However, considering the advancement in both endovascular experience and equipment in preserving even small splanchnic branches, we believe that the endovascular approach should always be in our operational treatment algorithm [19,20]. In our case, the RAA was type III and occupied entirely the upper two-thirds of the kidney's parenchyma. REB and RAT were not feasible with these conditions, and nephrectomy remained the single suggested alternative for open repair [9]. However, an inferior segmental renal artery branch was apparent just before the RAA formation, which allowed excluding the aneurysm using vascular plugs with simultaneous salvage of the functioning kidney's lower pole with covered stents. A potential complication of our approach could be the distal migration of the implanted materials to the IVC, resulting in severe pulmonary embolism. For this reason, we chose the implantation of large diameter vascular plugs. Due to the giant size of the RAA, the second potential complication could be the immediate renal infarction requiring nephrectomy. Fortunately, the patient developed only a mild post-implantation syndrome, lasting for three days. Additionally,

follow-up CTA at six months confirmed the patency of RISA and the salvage of lower pole parenchyma. To the best of our knowledge, this is the first reported case of successful exclusion of an RAA combined with high flow RAVM and preservation of the remaining functional renal parenchyma by minimally invasive endovascular techniques.

In conclusion, the complex RAA combined with high flow RAVM represents a challenging open and endovascular repair case. The location of a complex aneurysm at the renal hilum or the intraparenchymal space increases the technical difficulty and narrows the feasible treatment options. This case demonstrates that even in complex cases, the endovascular approach offers the possibility of obviating the nephrectomy and could be considered the first-line treatment.

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CONFLICTS OF INTEREST

The authors have nothing to disclose.

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AUTHOR CONTRIBUTIONS

Concept and design: GAP, AGP. Analysis and interpretation: all authors. Data collection: DAC, TZ, TEK, CL, AP, MDT. Writing the article: AGP. Critical revision of the article: GAP. Final approval of the article: all authors. Statistical analysis: none. Obtained funding: none. Overall responsibility: GAP.

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