Surgical approach of a giant intra-parenchymal renal artery aneurysm associated with arteriovenous fistula in a woman with solitary kidney: A report of a case

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Abstract:

Background: Renal artery aneurysms (RAAs) and renal arteriovenous fistulas (RAVFs) are uncommon lesions and they rarely occur concomitantly. While less than 10% of RAAs are intra-parenchymal, they are usually multiple and they may be associated with AVFs. Treatment of these lesions should always be considered on the basis of the location of the RAA and AVF.

Case Report: The case reported is a RAA with a concomitant high-flow AVF of the right kidney. Due to the size and location of the aneurysm and AVF hemodynamics, endovascular therapy was rejected by the interventionalists, and open repair through right nephrotomy and partial aneurysmatectomy was considered as best surgical option.

Conclusions: Renal artery aneurysms with concomitant arteriovenous fistula are extremely rare and their surgical approach is very demanding.

INTRODUCTION

Renal artery aneurysms (RAAs) represent an uncommon clinical entity, with an estimated incidence of 0.01-0.09% in the general population. They account for 0.01-0.5% of all aneurysms[1,2]. Intra-parenchymal renal artery aneurysms are extremely rare. They are detected in less than 10% of all patients with RAAs[3]. Lesion's diagnosis is usually an incidental finding (increasing the incidence of RAAs), as patients are investigated for other pathologies. Management of these vascular lesions remains controversial. Renal arteriovenous fistulas (AVFs) are abnormal communications between the intrarenal arterial and venous systems[4]. AVFs are relatively rare, with an estimated incidence of less than 0.04%. First to report an intrarenal arteriovenous fistula was Varela in 1928. They can be congenital, idiopathic, or acquired. The case presented in this article is characterized by the simultaneous presence of an intra-parenchymal RAA of very large dimensions and an AVF, which is rarely reported in the literature[4-8]. In almost all published cases, the AVF formation has been associated to a RAA rupture into an adjacent intraparenchymal vein. This mechanism is due to the close proximity of the vein, which first becomes compressed and then stretched by the adjacent aneurysm. Finally, erosion of the aneurysmal sac and the vein wall (with concomitant rupture into the vein) occurs.

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CASE PRESENTATION

A 44-year-old woman was admitted electively to our institution for surgical management of a large (28.3 mm x 35.6mm) intra-parenchymal renal artery aneurysm (RAA) [Fig. 1] associated with arteriovenous fistula (AVF) of the right kidney. Her past medical history included left total nephrectomy, [Fig. 1] secondary to RAA rupture and total avulsion of the left kidney during the 27th week of pregnancy.

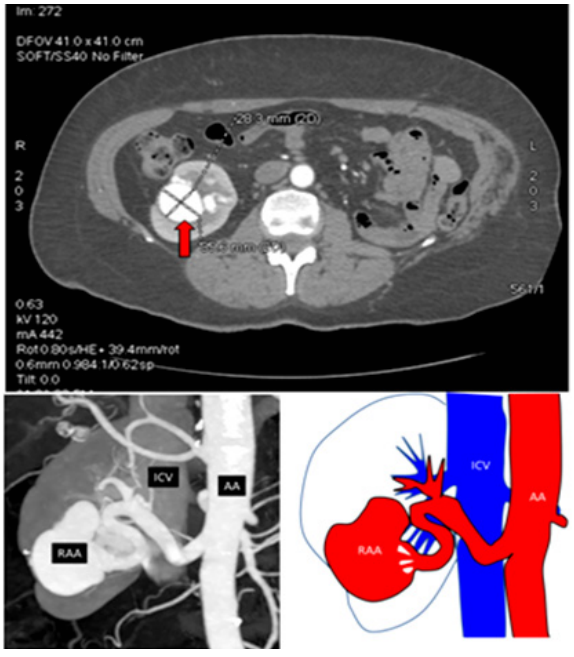

Figure 1. Computed Tomography (A: transversal CTA-aneurysm formation and its dimensions, B: 3D-CTA intra- parenchymal renal artery aneurysm in patient with solitary kidney and schematic representation)

Patient had no hypertension or other comorbidities. Aneurysm was detected during her last follow-up, in a routine abdominal examination due to her anamnesis. Operating procedure was performed with the patient in a left lateral decubitus position and lateral subcostal laparotomy between the iliac crest and the 12th rib. Retroperitoneal approach allowed identification and dissection of the hilus vessels. Secondary to vessel control with vessel loops, identification of the subcapsular aneurysm was feseable. The aneurysm was located between the middle part and the caudal pole of the kidney [Fig. 2].

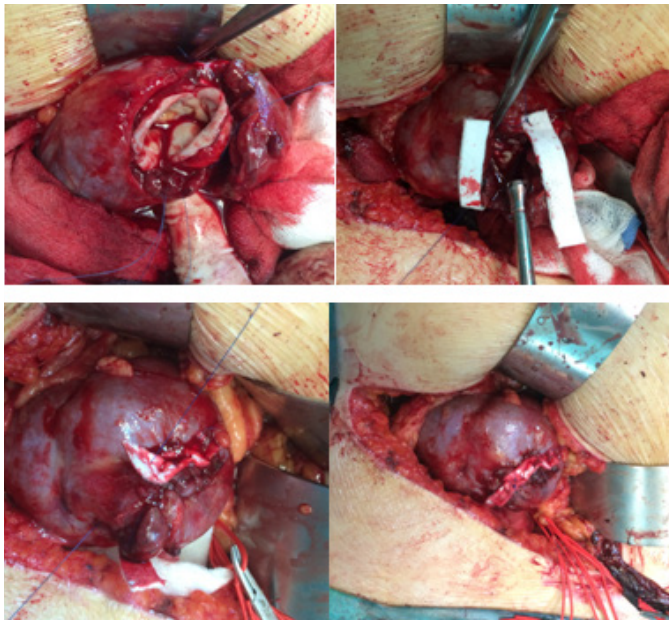

Figure 2. The intra-parenchymal aneurysm of the renal artery, peri-operative picture

Five thousands IU of unfractioned heparine (UFH) were administrated bolus, i.v., and peripheral nephrotomy with partial preparation of the aneurysmal sac and temporary vascular occlusion (unselected cross clamping of renal vessels) was performed [Fig. 3]. Incision of the aneurysmal sac (aneurysmatectomy) permitted an-at first sight- identification of the internal shunt, as well as identification and ligation of the origins of the inflow and outflow vessels with a 4-0 Polypropylene suture. Teflon pledgets were used to best seal the sutures. After partial aneurysmal sac resection [Fig. 2, 3] meticulous hemostasis of the parenchymal margins, and nephrotomy, the wound was closed in anatomic layers [Fig. 3]. The total duration of arterial and venous clamping was 23 min. Four units of red blood cells (RBC) were administered perioperatively, whereas the in-hospital post-operative period was uneventful. The patient was discharged on the 8th postoperative day in good clinical condition. Histological examination revealed degenerative aneurysmal disease. Thromboprophylaxis with Bemiparin 2500 IU/dl was administrated perioperatively and 100 mg of aspirin was prescribed for a 6 month period after surgery. In a two-years follow-up with Duplex scan to avoid contrast administration, patient's renal function showed no deterioration.

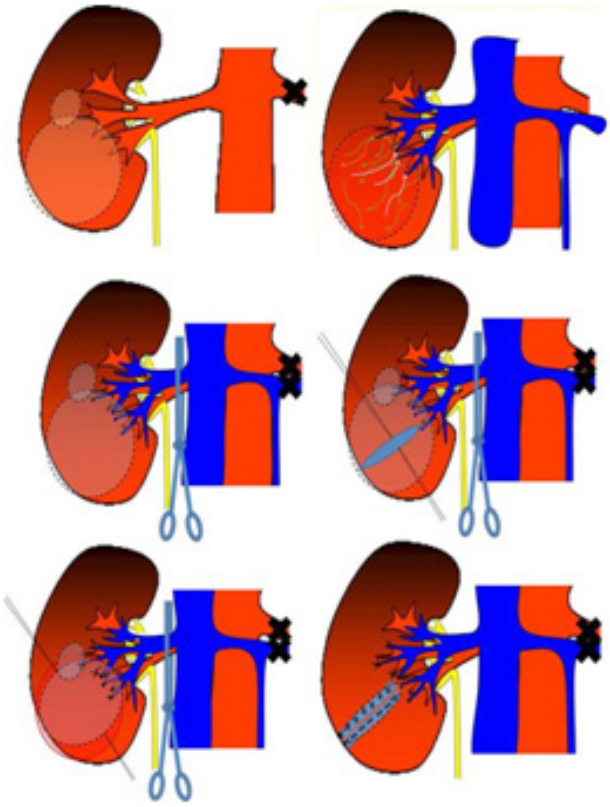

Figure 3: Schematic representation of the surgical procedure (A: localization of arterio-venous aneurysm formation, B: renal vessel cross-clamp and nephrotomy, C: partial aneurysmatectomy, suturing in anatomic layers step by step)

DISCUSSION

Coexistence of a RAA and an AVF is a rare condition[4-8]. RAA is defined as a 200% increase in the diameter of a normal renal artery[1,2]. Symptoms consist of flank pain, hematuria, renal infarction secondary to embolization and uncontrolled hypertension resistant to medical management. Indications for treatment of RAAs are rupture, rapid expansion, pregnancy, or aneurysm diameter > 2 cm[1,2]. Based on the treatment options for RAAs, Rundback et al proposed an angiographic classification system: Type 1 RAAs are saccular lesions arising from either the main renal artery trunk or from a proximal large segmental artery[9]. These aneurysms can be treated with stent grafts or coil embolization (endovascular repair)[10-11]. Open repair of Type 1 lesions includes excision and primary revascularization or patch angioplasty, with an autologous vein, or a PTFE graft[9]. Type 2 RAAs are fusiform and usually located at the main renal trunk, or at the proximal segmental arteries[9]. These are best treated surgically with segmental excision of the aneurysm and arterial reconstruction with an end-to-end small autologous tube graft or with aorto-renal by-pass grafting (in patients with no autologous veins available, a 6mm PTFE graft can be an option). Type 3 lesions are intra-parenchymal aneurysms involving the small, segmental or accessory arteries[9]. Type 3 aneurysms, due to their intra-parenchymal location are best treated with coil embolization (endovascular repair)[9].

In the era of endovascular repair, it is of high importance to evaluate every surgical option and deliver optimal individualized treatment in order to achieve the best medical outcome. This case-report refers to a Type 3 RAA combined with a high-flow AVF treated in our institution. These lesions, due to their intra-parenchymal location, are best treated with coil embolization. Nevertheless, due to the large dimensions of the RAA and the high-risk of embolization due to AVF high hemodynamics, segmental nephrectomy was performed on the basis of a safer outcome.

CONCLUSION

Renal artery aneurysms with concomitant arteriovenous fistula are extremely rare. Open repair approach is a safe and effective option even in cases with challenging anatomy.

No conflict of interest.

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