Aorto-left renal vein fistula is a rare complication of abdominal aortic aneurysm with unique clinical presentation

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Spontaneous aorto-left renal vein fistulas (ALRVF) are extremely rare, with only 30 cases presented in the literature. In the majority of the reported cases, the fistula involved an anomalous retroaortic left renal vein. In some aspects, the clinical findings differ from those of aortocaval fistulas, often making the correct diagnosis difficult and contributing to the delay in treatment. In this article, we present 2 such cases, review previously reported data, and discuss clinical features and treatment options of this rare condition. (J Vasc Surg 2010;52:1658-61.)

Since De Bakey et al first described the successful treatment of spontaneous rupture of abdominal aortic aneurysm (AAA) into the inferior vein cava (IVC), most vascular surgeons have become familiar with the syndrome consisting of a tender pulsating abdominal mass, machinery-like bruit, signs of pelvic and lower extremity venous hypertension, hematuria, and hyperdynamic cardiac failure. However, if the aneurysm ruptures into an aberrant retroaortic left renal vein (RLRV), creating an aorto-left renal vein fistula (ALRVF), clinical findings, although consistent, somewhat differ from those of aortocaval fistulas, often misleading the physician to search for a urologic disorder. In 1964, Lord et al described the first case of ALRVF, and since then only 30 other cases of ALRVF have been reported, usually involving an anomalous RLRV. We present 2 new cases of ALRVF recently treated in our institution and review previously published cases, pointing out the most common clinical and diagnostic features of this rare condition.

CASE REPORTS

Patient 1. A 71-year-old man with a history of hypertension and chronic pulmonary obstructive disease was admitted to the urology department of our hospital for the sudden onset of scrotal edema, groin pain, and gross hematuria. For the past 7 days, the patient was unsuccessfully treated in the regional hospital for suspected acute epididymitis. Physical examination revealed a pulsatile, moderately tender abdominal mass with an audible “machinery-like” bruit, and painful scrotal edema. The patient had gross hematuria, proteinuria, and elevated serum creatinine (345 µmol/L; 3.9 mg/dL). An abdominal ultrasound scan revealed a 9.5-cm infrarenal abdominal aortic aneurysm without signs of retroperitoneal hematoma. Multislice computed tomography, however, showed a large communication between the aneurysm and the RLRV (Fig 1). The aneurysm was infrarenal, with a short neck, unsuitable for endovascular repair. Upon isolation and infrarenal aortic clamping, the aneurysm was opened to discover a brisk venous bleeding from the posterior aneurysm wall. Bleeding control was achieved with digital compression, and the 3-cm defect in the aberrant retroaortic renal vein was oversewn with 3-0 polypropylene continuous suture. Aortic reconstruction with tubular 18-mm Dacron graft (InterVascular, Montvale, NJ) followed without complications. The postoperative course was complicated with bilateral bronchopneumonia requiring prolonged mechanical ventilation and antibiotic therapy. Hematuria, scrotal edema, and renal impairment resolved within 72 hours. The patient was discharged on the 22nd postoperative day in good condition and was doing well at his 1-month follow-up, with normal serum creatinine (80 µmol/L; 0.9 mg/dL).

Patient 2. A 68-year-old man was referred to our institution from the regional urology department where he was observed during the past 2 weeks for the acute onset of left-sided varicocele and intermittent hematuria. Physical examination revealed a non-tender pulsatile abdominal mass and a left-sided varicocele. Laboratory results included microscopic hematuria, proteinuria, elevated serum creatinine (175 µmol/L; 2.0 mg/dL) and erythrocyte sedimentation rate (90 mm/h). Abdominal ultrasound showed a large (8.5 cm), juxtarenal AAA without signs of rupture or arteriovenous fistula, although an experienced angiologist was warned of the high clinical suspicion for ALRVF (a previously reported case was treated only 5 weeks earlier). Transfemoral aortography demonstrated early opacification of retroaortic left renal and gonadal veins with absence of contrast in the left kidney (Fig 2). Since the aneurysm was juxtarenal, unsuitable for endovascular repair, the patient underwent open surgery. Intraoperatively, we discovered an inflammatory juxtarenal AAA, closely adherent to the duodenum and IVC. After minimal dissection in order to avoid injury to the retroperitoneal structures, the aorta was clamped below the diaphragm. After the opening of the aneurysm sac, venous bleeding was encountered from a 1.5-cm defect in the RLRV, which...
obliquely coursed the posterior aneurysm wall to join the IVC. Control of the fistula was accomplished by digital compression, and the venous defect was sutured from within the aneurysm with continuous 3-0 polypropylene. Aortic continuity was restored with a 20-mm Dacron interposition graft (InterVascular, Montvale, NJ). The postoperative course was uneventful with serum creatinine returning to baseline values within 48 hours. The patient was discharged on the eighth postoperative day in good condition. At his 6-month follow-up, the patient was doing well, with serum creatinine of 87 μmol/L (1.0 mg/dL). Multislice computed tomography, however, revealed left renal vein occlusion, with venous drainage from the left kidney through the left gonadal vein, whereas renal scintigraphy demonstrated moderately decreased left kidney function (Fig 3).

**DISCUSSION**

RLRV is a rare anatomic variation, with an estimated incidence of 1.8% to 3.4%. This anomaly occurs during the complex embryological development of caval and renal venous systems through subsupracardinal anastomosis and circumaortic venous collar, when the anterior limb abbrantly disintegrates leaving only the posterior limb as the only left renal vein. As noted by most clinicians, RLRV usually has an oblique downward course between the aorta and lumbar vertebrae, joining the IVC significantly lower than its right counterpart. The presence of RLRV has several surgical implications, such as the risk of venous injury and bleeding during abdominal aortic surgery, potential for the development of posterior “nutcracker” phenomenon, and finally the possibility of ALRVF formation.

As noted by Mansour et al, ALRVFs have some unique clinical features that have to be appreciated on the way to the correct diagnosis, which can often be elusive for a critical period of time. In fact, similar to our 2 cases, most reported patients with ALRVF had a delay in treatment from 2 to 21 days (average 6.5 days), primarily due to the initial misdiagnosis of a urologic condition.

The majority of previously reported patients were men (93.5%) with an average age of 65 years, presenting with left flank, abdominal, or scrotal pain, often associated with scrotal edema or varicocele, such as in our 2 cases. Hematuria was present in 90%, proteinuria in 60%, while various degrees of renal impairment were noted in 83% of the cases, all probably as a direct result of acute venous hypertension affecting the left kidney. In contrast to patients with fistulas directly involving the IVC, signs of cardiac failure or hemodynamic instability were present in less than 20% of the cases.
possibly due to shunt reduction by direct RLRV compression against the vertebral bodies by the aneurysm. All but two aneurysms were larger than 7 cm (average 8.5 cm), while the position of the RLRV was retroaortic in 90% of the cases. Four patients were treated with endovascular techniques, while the rest received open surgical repair with excellent results (6.5% mortality).

In most cases, surgical treatment was straightforward: bleeding from the fistula was controlled by digital compression and the defect in the RLRV was sutured from within the aneurysm, followed by routine aortic reconstruction. Only 1 patient required chronic hemodialysis, while others demonstrated quick postoperative recovery of renal function. In 1 of our cases, this more or less blind fashion of RLRV suture probably caused left renal vein obstruction that led to deterioration in left kidney function, although serum creatinine remained within normal ranges.

Anatomic considerations (short aneurysm neck) precluded endovascular treatment in both of our cases. Successful endovascular aneurysm repair does not necessarily close the communication between the left renal vein and the aneurysm sac, causing persistent type II endoleak, which was the cause for reintervention in 2 reported cases. However, bearing in mind the possible complications of open repair (respiratory complications, excessive bleeding, LRV occlusion), high-risk patients who meet the anatomic criteria could be best served with endovascular repair with close follow-up.

CONCLUSION

All patients presenting with hematuria, abdominal or flank pain, scrotal edema, or varicocele who have a finding of AAA on abdominal ultrasound scan should be immediately expedited to a vascular surgeon for further evaluation. Open surgery for ALRVF gives excellent results, but endovascular aneurysm repair should be considered for hemodynamically stable high-risk patients with favorable anatomy.

REFERENCES


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