CASE REPORT

Arterioureteral fistula: an unusual clinical case

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SUMMARY

Arterioureteral fistulas (AUFs) are abnormal communications between a major artery and the mid to distal ureter. It is a rare but potentially life-threatening condition that is seldom recognised. We present a case of a 66-year-old man who was admitted to the surgical ward owing to infection of an aortic bifemoral bypass graft. During admission, the patient developed persistent haematuria with considerable loss of haemoglobin. He was submitted to urgent surgical exploration of the graft. An ascending pyelography performed at the beginning of the surgery clearly shows a communication between the left ureter and the vascular graft. Open surgical exploration was undertaken, the fistulised section of the ureter resected and an end-to-end ureteroplasty was performed. The vascular graft was removed and the patient later submitted to left supracondylar amputation. Urinary drainage remained intact.

BACKGROUND

Arterioureteral fistulas (AUFs) are abnormal communications between a major artery and the mid to distal ureter. It is a rare but potentially life-threatening condition that is seldom recognised. Until 2008, 139 cases were reported in the English literature; of these, 13% died of an AUF-related cause; in this group, only 22% of the fistulas were recognised before treatment.¹ As such, the diagnosis requires a high index of suspicion and awareness of this condition.

CASE PRESENTATION

A 66-year-old man who had undergone an aortic bifemoral bypass 1 year earlier presented to the outpatient clinic, with pain in the left groin and a foul discharge from the surgical incision. He was admitted to the surgical ward because of infection of the vascular graft, and started on broad spectrum antibiotics.

The patient had a history of acute myocardial infarction and peripheral vascular disease. He had been subjected to an aortic bifemoral bypass, and then to a sequential femoral-tibial bypass on the right lower limb. Later, two thrombectomies were executed and in the end a right supracondylar amputation was performed.

During admission, the patient developed persistent haematuria with considerable loss of haemoglobin.

INVESTIGATIONS

A CT angiography was performed, which revealed densification of the fat around the graft, with contrast leakage outside the graft and dehiscence of the

surgical anastomosis. Furthermore, there was hydronephrosis on the left collecting duct and reduction of the ureter lumen where it had contacted with the densification of the fat near the graft. There were, however, no signs of active AUF (figure 1) on the CT scan.

A cystoscopy was then performed. There were no signs of a vesical origin of the haematuria, and haematuria from the left ureteric orifice was noticed.

TREATMENT

The patient was taken to the operating theatre for urgent surgical exploration of the graft. Cystoscopy and ascending pyelography were performed initially. The ascending pyelography clearly showed a communication between the left ureter and the vascular graft, with contrast filling the graft and arterial system (figure 2). The left ureter was catheterised with a ureteric stent, under fluoroscopic guidance.

Open surgical exploration was then undertaken. There was extensive infection of the prosthetic vascular graft. The left ureter was identified. It presented clear inflammatory signs and was affected by the infection of the vascular graft.

The fistulised section of the ureter was resected and an end-to-end ureteroplasty performed. The ureteric stent was left in place.

Resection of the vascular graft was performed because of the extensive infection.

OUTCOME AND FOLLOW-UP

The patient developed signs of critical limb ischaemia and was later submitted to left supracondylar amputation.

Urinary drainage remained intact and the ureteric stent was removed 3 months later.

Figure 1 Axial CT image through the pelvis, in the arterial phase, showing fat densification surrounding the prosthetic vascular graft secondary to graft infection. The left ureter (*) is involved in the fat densification. There were no signs of active arterioureteral fistula.

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Figure 2 Retrograde pyelography performed before the open surgical exploration of the graft. Contrast flowed through the arterioureteral fistula and filled the vascular graft. Pseudoaneurysms of the left external iliac artery are also visible.

DISCUSSION

AUFs were first reported by Moschowitz, in 1908,² and have been classified as primary, secondary (iatrogenic) or pregnancy related.³ Primary AUFs account for <15% of fistulas⁴ and usually develop with erosion of an aneurysm into the ureter.⁵ Secondary AUFs form when predisposing factors such as prior irradiation, indwelling ureteric stent, prior pelvic surgery (including prior aortic or iliac vascular procedures) or a history of pelvic cancer result in erosion of an artery into the ureter.⁶ AUFs during pregnancy are even rarer; all three cases were reported before 1939.³

The incidence of prosthetic vascular graft infections (PVGIs) has been reported to vary from 1% to 6%.⁷ Reported risk factors for the development of PVGI include patient-related factors, such as older age, male sex, high body mass index, heart failure, immunodeficiency, diabetes mellitus, renal failure, chronic obstructive pulmonary disease, blood stream infection at the time of graft placement, skin ulcers in the lower extremity and prolonged pre-operative hospital stay.⁸ Staphylococci (*Staphylococcus aureus* and coagulase-negative staphylococci) account for 80% of PVGIs. Staphylococci and *Pseudomonas* spp are strong biofilm producers. PVGI is often polymicrobial, with involvement of anaerobes, and there are cases where cultures remain negative, often in the context of previous antimicrobial therapy.⁸

The most recent review by van Berg *et al* described 139 cases published up to 2008 (93% of these after 1970). The mortality rate was 13%, but when the AUF led to haemodynamic instability the mortality rate reached 23%.¹

The most common symptom of AUFs is gross haematuria, usually intermittent in nature. Patients may present with bleeding ranging from microscopic haematuria to life-threatening haemorrhage.⁴ Lumbar or back pain may be an accompanying symptom (17% of patients) and infection may be present (10% of cases). A common finding is the presence of a pseudo or true aneurysm of the common or external iliac artery (38% of patients),¹ which should raise suspicion for this diagnosis.

The preoperative diagnosis of these fistulas is difficult. Cystoscopy is not specific and ascending pyelography requires a favourable pressure gradient from ureter to artery, which may not be possible in the presence of significant bleeding. Ureteroscopy is not recommended because it can tear the fistula or dislodge a tamponading clot, leading to massive haemorrhage.⁹

Ascending pyelography and arteriography are the most sensitive tests, yet they were reported as only being diagnostic in 45% and 41% of cases, respectively.¹⁰ Angiographic techniques required to visualise the fistulas have included selective arterial catheterisation, use of multiple projections and provocative manoeuvres.¹¹ In fact, the diagnosis was considered in 36 patients only after open surgery had been performed. In another five patients, the diagnosis was performed only during autopsy.¹

At present, no consensus has been reached regarding the best treatment approach. Combined urological and vascular treatment has been performed in the majority of patients. An open approach with the resection of the fistulous connection between the artery and ureter has been the most commonly performed procedure.¹

Management of the arterial component of the AUF varies and is influenced by associated local infections, the presence of associated aneurysmal or occlusive disease, the available collateral circulation to the ipsilateral leg and the presence of an arterial graft. Vascular surgical procedures include local reconstruction (ie, arteriorrhaphy, patch closure, interposition graft and bypass), ligation with or without extra-anatomic bypass (if AUFs arise from either common or external iliac artery) and ligation of the internal iliac.⁴

Management of the ureter can involve ligation (with or without nephrectomy), primary repair, re-implantation to a site away from the vessels or diversion of the urine with a nephrostomy tube. In many, if not most, patients, an open operation is difficult and morbid due to the associated risk factors, namely, prior pelvic irradiation and/or pelvic surgery, and haemodynamic instability from the haemorrhage. For these reasons, an endovascular approach with a covered stent graft has become very appealing.⁶ A retrospective review in a single centre did not identify a clear advantage for endovascular or open vascular surgical management.¹²

Learning points

- ▶ Arterioureteral fistula (AUF) is a rare but potentially life-threatening condition that is seldom recognised.
- ▶ Primary AUFs account for <15% of fistulas and usually develop with erosion of an aneurysm into the ureter.
- ▶ Secondary AUFs form when predisposing factors such as prior irradiation, indwelling ureteric stent, prior pelvic surgery (including prior aortic or iliac vascular procedures), or a history of pelvic cancer result in erosion of an artery into the ureter.
- ▶ Sudden life-threatening haematuria in patients with known risk factors for AUF should raise the suspicion of AUF.
- ▶ The best treatment approach is unknown. Combined urological and vascular treatment has usually been performed.

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Competing interests None declared.

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