

## Uretero-Arterial Fistula : Two Observations

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**Abstract.** Two cases of life-threatening haematuria, secondary to an uretero-arterial fistula, are reported. Both cases present predisposing causative factors.

One patient had a combination of previous aorto-bifemoral bypass grafting, an iliac artery aneurysm (retrogradely perfused), and an indwelling ureteral stent for ureteral compression. The other patient had previous aortoiliac surgery and obstructive uropathy with chronic urinary tract infection.

Preoperative diagnosis of uretero-arterial fistula was made in only one patient. He was successfully operated (exclusion of the iliac aneurysm). In the other patient, nephrectomy was attempted to control reno-ureteral bleeding of unknown origin. Fatal recidive of brisk haematuria occurred some days later.

Factors contributing to the development of uretero-arterial fistula, their diagnosis and optimal treatment are discussed.

### Introduction

Uretero-arterial fistula is a rare cause of gross haematuria, that can be life-threatening. The first case of uretero-arterial fistula associated with arterial disease has been described in details by Taylor [1] in 1939. Since then, 38 cases have been reported in the literature [2]. The authors report two other observations, illustrating some characteristic features and promoting factors, incriminated in the development of uretero-arterial fistulas, as well as diagnostic uncertainty of this condition.

Based on a review of recent literature, presumptive causes, clinical features, diagnostic investigations, and management of uretero-arterial fistulas are discussed.

#### Case 1 :

A 76-year-old man was admitted in July 1996 for massive haematuria, resulting in hypovolemic shock (blood pressure 60 mmHg). Haematocrit fell to 23%, and haemoglobin to 8.0g/l. There was moderate renal insufficiency (creat 18.0 mg/l, blood urea nitrogen (BUN) 0.96g/l). He presented repeated episodes of gross haematuria since one week.

In 1988, the patient underwent surgical correction of an infrarenal abdominal aortic aneurysm (aorto-bifemoral grafting). Seven years later, aneurysmal degeneration of the ligated, retrogradely perfused common iliac arteries was evidenced as a cause of moderate hydronephrosis and disturbed renal function

(creat 22 mg/l, BUN 0.9 g/l). A double J-polyurethane catheter (Angiomed® 8-F) was placed in both ureters. Hydronephrosis partially resolved and renal function improved (creat 14 mg/l, BUN 0.55 g/l). Seven months later, haematuria ensued (July 1996).

On admission, a tender pulsatile mass was palpable in the left iliac fossa. There was clinical evidence of bladder tamponade. Urethral catheterization evacuated fresh blood. Intravenous urography revealed a functionally excluded left kidney and the two ureteral stents in place (Fig. 1). Computer-assisted tomography confirmed a left iliac artery aneurysm of 5 cm diameter and an adherent mega-ureter (18 mm diameter) filled with blood clots (Fig. 2). Emergency operation consisted in ligation of the left external iliac artery, to exclude the iliac aneurysm. No ureterolysis was performed and the double J-catheter was left in place to avoid urinary leakage. No further episodes of haematuria occurred. Echographic control one week later confirmed thrombosis of the iliac aneurysm. Renal function improved (creat 13 mg/l, BUN 0.7 g/l). At four months follow-up, the patient is doing well.

#### Case 2 :

A 70-year-old man presented in November 1980 with a history of intermittent gross haematuria since one month. From patient's past history we withhold an aortobifemoral bypass graft five years before. Chronic urinary tract infection and left sided hydronephrosis developed in the postoperative course. He



Fig. 1

Preoperative intravenous urography (case 1). The two double J ureteral catheters are in place. The left kidney is not visualized.

On admission, a tender pulsatile mass was palpable in the left iliac fossa. There was clinical evidence of the intermittent massive haematuria. Cystoscopy revealed blood emanating from the left ureteral orifice. Selective arteriography of the left kidney did not disclose the origin of the haematuria. Intravenous pyelography showed hydronephrosis with multiple filling defects in the pyelocaliceal and ureteral lumen. Since no evident cause for the haematuria was found, the bleeding was misjudged to originate from the left kidney (suspicion of intrarenal microaneurysm ruptured into the urinary tract), nephrectomy was performed in an attempt to control the huge haematuria. Five days postoperatively, massive haematuria recurred, with haemodynamic instability.

Emergency laparotomy revealed a distended residual left ureteral stump, filled with blood clots. On opening of the ureter, an erosion by the adherent bifurcation graft was evidenced. Operation consisted in ureterectomy and excision of the left limb of the bifurcation graft. A femorofemoral cross-over graft was done to revascularize the left lower extremity. The hypovolemic shock was not well tolerated and the patient suffered fatal myocardial infarction the next day.

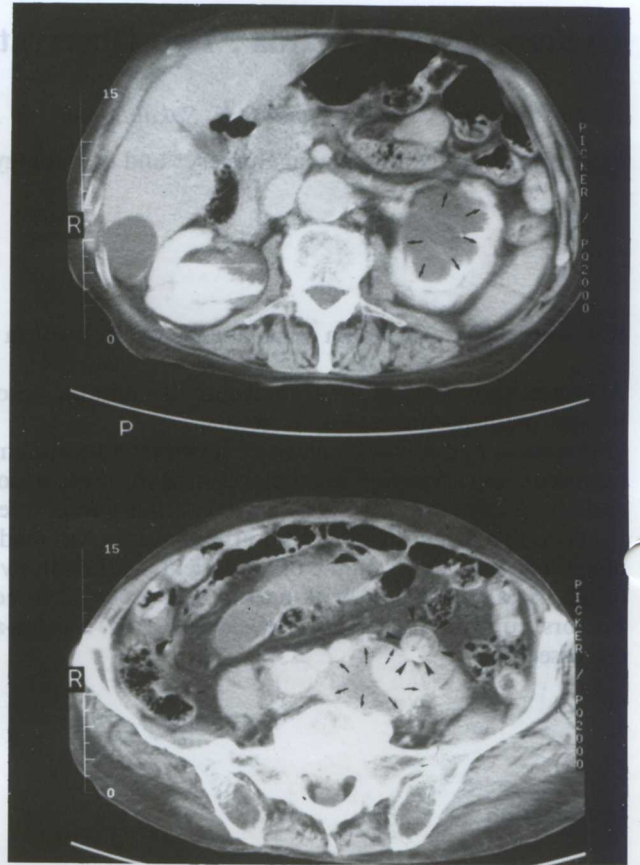


Fig. 2

Preoperative CT-scan (case 1) reveals huge uretero-nephrosis of the left kidney. The distended left ureter (arrow heads) is adherent to a left iliac artery aneurysm (partially thrombosed) (small arrows).

## Discussion

Uretero-arterial fistulas are exceptional (two cases in series of 2100 surgical aortoiliac revascularization procedures at our department in the same period, 1980-1996). These two cases illustrate some particular aspects of this rare condition. The first case had as predisposing factors an indwelling ureteral catheter since seven months, previous aortoiliac surgery and an iliac artery aneurysm. Correct preoperative diagnosis was made on CT-scan. Haematuria was controlled after exclusion of the iliac aneurysm. In the second case, the cause of haematuria was unknown before operation. It highlights the difficulty of demonstrating uretero-arterial fistulas. A renal source of bleeding was suspected and nephrectomy was performed. This operation ultimately proved to be inappropriate, since haematuria recurred from the lower ureteral stump.

The clinical course of uretero-arterial fistulas is characterized by prolonged intermittent gross haema-

turia, preceding the episode of life threatening haemorrhage in the urinary tract [3, 4]. Uretero-arterial fistulas are almost secondary to underlying predisposing factors, such as previous pelvic surgery (exenteration, uretero-enterostomy) [2, 4-10], previous aortoiliac surgery [3, 11-13], prolonged ureteral catheterization for obstruction [2-4, 6, 7, 9, 10, 14], previous irradiation [2, 4, 6-9, 14], and iliac artery aneurysm [1, 4, 15-17]. Spontaneous uretero-arterial fistula of a normal iliac artery and ureter, without prior surgery or endoscopic intervention, has not been reported. All these various favouring settings have in common a periureteral fibrosis and devascularization. The presumptive mechanism is pressure necrosis by transmission of arterial pulsation on an already compromised ureter, resulting in uretero-arterial fistula formation at the side where the ureter crosses the iliac vessels. A stiff intraluminal double-J stent rigidifies the ureter and predisposes to erosion into the adherent artery. The ureteral stent functions as a valve, temporarily tamponading the fistula. This explains that haematuria is often intermittent, before cataclysmic bleeding occurs. Removing the indwelling catheter often reactivates the bleeding [3, 4, 6, 7, 12].

Prolonged ureteral stenting is the main promoting factor, present in 65% to 85% of the reported cases [4, 7, 18]. Leaving for prolonged periods a stent in an ureter, jeopardized by ischaemic fibrosis, is not without danger. Ureteral stents should be of small caliber and soft (silicone), to minimize the risk of uretero-arterial fistula.

A correct preoperative diagnosis of uretero-arterial fistula is difficult to make for several reasons. Arteriography generally fails to demonstrate the uretero-arterial fistula, because of the intermittent course of bleeding. In only nine of the cases reported in the literature, arteriography was contributive to correct diagnosis [18]. Mobilization of the ureteral stent (over a wire) can dislodge a clot at the site of the fistula, provoking active bleeding. This manoeuvre can be helpful to unmask the fistula on angiography. Intravenous pyelography shows a hydroureteronephrosis and filling defects within the distended ureter, consistent with thrombus. It says nothing on the source of the bleeding. In rare cases, retrograde ureteropyelography reveals extravasation of contrast medium at the site of the fistula [2, 10, 13, 17]. Cystoscopy allows to exclude other causes of haematuria and reveals the side of ureteral bleeding, but not the cause. Computerized tomography is the most informative investigation: it shows the adherence between a distended, clot-filled ureter and the iliac artery [8, 13, 14, 17]. In view of the difficulties to evidence uretero-arterial fistulas preoperatively, a high index of suspicion is essential.

Correct preoperative diagnosis is determinant for the outcome. A favourable outcome was observed in 92%

of cases where the diagnosis of uretero-arterial fistula was confirmed before operation [2, 3]. On the other hand, a fatal outcome was observed in half of the cases with missed or erroneous diagnosis [18]. Inappropriate nephrectomy [5, 10, 12, 14, 15, 19] or renal embolization [20, 21], leaving the distal ureter and fistula in place, has been reported by other authors.

Treatment should be principally oriented on the arterial side of the fistula. Exclusion of the iliac artery by surgical ligation or endovascular embolization techniques [2, 6, 7, 9, 18, 21] stops the active bleeding. Local contamination and scarring create a hostile environment, rendering direct arterial repair hazardous. Where necessary, an extraanatomic cross-over femoro-femoral bypass is the safest mean to obviate limb ischaemia [2, 3, 5-9]. Recently, a new attractive therapeutic option was described by Kerns [18]. He excluded the uretero-arterial fistula by an endovascular vein-covered stent. The ureteral component of the fistula does not require treatment [18]. The distended ureter filled with blood clots should be left undisturbed and the indwelling double-J catheter should be left in place to assure adequate urinary drainage [2, 7]. Optimally, the ureteral stent should be removed after some months, if clinically indicated [18].

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removal of ureteral stents. The presumed mechanism is pressure necrosis by transmission of arterial pulsation on an already compromised ureter, resulting in uretero-arterial fistula formation at the site where the ureter crosses the iliac vessels. A still intraluminal double-J stent rigidifies the ureter and predisposes to erosion into the adjacent artery. The ureteral stent functions as a valve temporarily tamponading the fistula. This explains that haematuria is often intermittent, before catastrophic bleeding occurs. Removing the indwelling catheter often reactivates the bleeding [3, 4, 6, 7, 12]. Prolonged ureteral stenting is the main promoting factor present in 82% to 85% of the reported cases [4, 7, 18]. Leaving for prolonged periods a stent in an ureter, jeopardized by ischaemic fibrosis, is not without danger. Ureteral stents should be of small caliber and soft (silicone) to minimize the risk of uretero-arterial fistula.

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