Arterio-Ureteral Fistula: 11 New Cases of a Wolf in Sheep's Clothing

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Purpose: We provide insight into the presentation, diagnostics, treatment, and accompanying clinical difficulties and complications of an arterio-ureteral fistula and add 11 patients with arterio-ureteral fistula to the 90 described in the literature.

Materials and Methods: A retrospective search was done for confirmed cases of arterio-ureteral fistula that were treated at 3 medical centers.

Results: A total of 11 cases of arterio-ureteral fistula were reviewed from 1980 to 2006. A history of major abdominal surgery had an essential role in 10 of the 11 cases, especially vascular intervention in 8. All patients presented with varying manifestations of hematuria. Ureteral contrast studies and nonprovocative angiography provided the most valuable diagnostic information with all 5 and 3 of 5 showing positive results, respectively. In 4 of 11 patients (36%) the diagnosis of arterio-ureteral fistula was only made during laparotomy. Ten patients were treated with a classic open surgical approach (vascular and urological) and in 1 endovascular stents were inserted. Two of 11 patients (18%) needed acute surgical intervention because of hemodynamic instability. The in hospital mortality rate was 9%. At a mean followup of 17 months 3 other patients (27%) had died of causes unrelated to the arterio-ureteral fistula.

Conclusions: The diagnosis of arterio-ureteral fistula should be considered in patients with persistent hematuria who have a history of major abdominal vascular surgery even when diagnostic test results are negative for a fistula, because in 36% of our patients the diagnosis could only be made at operation. The in hospital morbidity and mortality rates are considerable and, therefore, a timely diagnosis and elective multidisciplinary treatment are preferred.

Key Words: ureter, arteries, fistula, hematuria, stents

Arterio-ureteral fistula is an uncommon diagnosis. A previous review of this disease identified only 90 patients in the literature to date.[1] These fistulas are often life threatening and due to their scarcity the diagnosis can easily be delayed or even missed. The mortality rate is 7.1% to 23% and strongly related to the diagnostic delay.[2,3] In most patients hematuria is the only precursor of this disease. To have a keen eye for the possibility of this entity is essential to adequately recognize and treat this cause of hematuria. We focused on the etiology, presentation, diagnostics, treatment and associated clinical pitfalls of AUF. This study was based on 11 case reports from 3 hospitals and the relevant literature.

MATERIALS AND METHODS

Patients

We retrospectively collected data on patients with proven AUFs at 3 hospitals with surgical training accreditation, including University Medical Center Utrecht, St. Antonius Hospital Nieuwegein and Free University Medical Center Amsterdam. The records of all performed surgical interventions were searched. Furthermore, urologists and vascular surgeons were asked whether they had ever encountered patients with an AUF. All retrieved cases were verified for the presence of an AUF during intervention.

Definitions

An AUF was defined as a confirmed abnormal passageway between an artery or a previously inserted vascular graft and the ureter. Data from the medical records on patients with a confirmed diagnosis were collected, including general patient characteristics, medical history, the diagnostic pathway, fistula specifications, vascular and urological treatment, followup and the eventual cause of death. End points were patient death or loss to followup.

Literature Search

The literature was searched in PubMed® and EMBASE™ using combinations of synonyms of the description of the diagnosis.

RESULTS

The search strategy resulted in 11 confirmed cases of AUF being gathered from 1980 to 2006. Mean age of the 11 patients at diagnosis was 61 years (range 33 to 84) and 8

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Characteristics of patients with AUF described from 1980 to 2006

No. women/men	8/3
Mean age (range)	61 (33–84)
No. history:	
Vascular	8
Onco-urological	2
Onco-urological, radiotherapy + ureteral stenting	1
Idiopathic	1
No. side:	
Rt	6
Lt	5
No. iliac artery involvement	11
No. indicative investigations/total No.:	27
Ureteral contrast study	5/5
Nonprovocative angiography	3/5
CT	2/4
Ultrasound	2/5
Pyelography	0/3
Cystoscopy	0/5
No. diagnosed during surgery	4
No. vascular treatment:	
Unilat bypass	7
Aortic bifurcation prosthesis	2
Defect oversewn	1
Endovascular stenting	1
No. urological treatment:	
Ureteroureterostomy over catheter	5
Nephroureterectomy	3
Ureteral catheter insertion	1
Ileal conduit revision	1
Kidney autotransplantation	1
Mean followup (mos)	17
No. cause of death (time):	
Fistula	1 (9 days postop)
Other	3 (mean 21 mos)

patients were women. The table shows an overview of all case characteristics.

The medical history revealed a preceding central vascular reconstruction in 8 patients. Surgery of the common iliac artery had been performed in 5 cases, including an interposition graft, endarterectomy, patch, bypass or combinations, an aortic bifurcation prosthesis had been placed in 2 and no specified surgical procedure for an abdominal aneurysm of the aorta had been performed in 1. Two patients had undergone previous extensive pelvic oncological surgical resection due to carcinoma of the cervix in 1 and carcinoma of the rectosigmoid in 1. In the former patient urinary diversion had been created, while in the latter a ureteral stent was present and radiation therapy (45 Gy) had been given. The latter woman was the only patient in our series in whom a ureteral stent was present. Because of retroperitoneal fibrosis a 6Fr ureteral stent had been in situ for more than 7 years.

Symptoms started in all patients with varying presentations of hematuria. Occasionally multiple episodes of hematuria had previously occurred. In 1 patient the first episode of hematuria was related to the start of anticoagulants for a lung embolism.

Before diagnosis the 11 patients underwent a total of 27 investigations, consisting of a ureteral contrast study, nonprovocative angiography, CT, ultrasound, excretory urography and cystoscopy. More than 1 investigation could have had a role of importance in a single patient. A flow of blood from the artery into the ureter or urine from the ureter to the artery was consider to prove the existence of an AUF. Differences in sensitivity were observed among these investigations. Additional evidence for the AUF diagnosis was found on all 5 ureteral contrast studies, including 1 antegrade and 4 retrograde studies, 3 of 5 angiographies, 2 of 4 CT images and 2 of 5 ultrasound studies. Three pyelographies and 5 cystoscopies provided no additional value.

The diagnosis in 4 patients could only be confirmed during the surgical procedure and in 1 repeat laparotomy was performed because the fistula was not detected during the first operation and hematuria persisted subsequently. During the diagnostic process ipsilateral hydronephrosis was observed in 7 patients and ipsilateral pseudoaneurysms were noted in 7. Pyoureter with pyonephrosis was present in 1 patient.

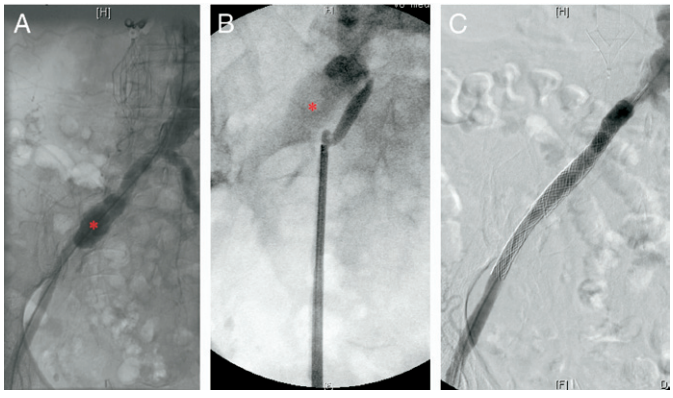
At diagnosis the AUF appeared to be on the right side in 6 patients and on the left side in 5. In all 11 patients the iliac artery was involved in the AUF, including the common iliac artery in 5 and in 6 this was not further described. In 4 of the 8 patients who had previously undergone central vascular reconstruction the vascular side of the AUF was specifically described to originate from the anastomosis of the native artery with previously inserted vascular graft material.

After the diagnosis 9 patients underwent elective surgery and in the other 2 the operation was performed in an acute setting because of hemodynamic instability. A combined vascular and urological approach was used in all 11 cases. Vascular interventions consisted of bypass grafting of the affected iliac artery in 5 patients, ligation of the affected vessels and staged bypass grafting in 2, insertion of an aortic bifurcation prosthesis in 2, suturing of the arterial defect in 1 and endovascular stenting of the iliac artery in 1 (see figure). In 3 patients previously inserted infected vascular graft material was resected first.

Urological interventions consisted of ureteral repair (ureteroureterostomy) over a catheter in 5 patients and insertion of a ureteral catheter in 1. Three patients needed nephroureterectomy, 1 required ileal conduit revision and another underwent kidney autotransplantation.

To prevent further fistularization omental interposition was used during the operation in 1 patient. In 1 patient embolization and then nephrectomy of the right kidney were performed, which later appeared to be unnecessary. Although the diagnosis of AUF was not considered, hematuria was persistently considered to be of renal origin.

Mean followup was 17 months (range 9 days to 4.5 years). Four patients died during followup. One patient died after 9



A, arteriography of right iliac artery shows aneurysm (asterisk) near iliac-ureteral crossing. *B*, retrograde contrast study of right ureter reveals extravasation of contrast material into right iliac artery aneurysm. *C*, arteriography demonstrates right iliac artery after placement of 2 endovascular stents.