



Uretero-iliac artery fistula: a challenge diagnosis for a life-threatening condition: monocentric experience and review of the literature

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Abstract

Introduction Uretero-iliac artery fistulae (UIAF) are the consequence of chronic inflammatory events that create a fibrous and poorly vascularized uretero-vascular adhesion. They often occur in patients with a history of surgery, pelvic radiotherapy, and chronic ureteral stenting. The presentation is usually massive gross hematuria with acute anemia unto to hemorrhagic shock, representing a life-threatening condition. High mortality rate is reported (7–23%).

Materials and methods We present four cases in three patients, treated in our Institution from 2013 to 2018, and reviewed the published literature. UIAF was defined as the ratified presence of an abnormal communication between the ureter and any artery. In all patients, the UIAF was initially evaluated by contrast-enhanced computed tomography (CT) angiography. The management strategy was defined individually based on the specific risk profile of each patient.

Results In all cases, ureteral-iliac artery fistula occurred in female patients with previous surgery or radiation and with presence of indwelling ureteral stent. In every case the hematuria was massive and life-threatening. Diagnosis was delayed because of the poor diagnostic accuracy of CT scan, leading to over-treatment. Angiography resulted the best diagnostic tool. The endovascular treatment proved good outcomes in terms of early complications, with no case of mortality.

Conclusions In case of gross hematuria during ureteral stent change in female patients with history of previous pelvic surgery and radiation, UIAF must be suspected and immediately treated, since it represents a urological emergency. Angiography can be useful to confirm the diagnosis and endovascular treatment with vascular endoprosthesis is the best therapeutic option.

Keyword Ureteroiliac fistula · Ureteral stricture · Hematuria · Iliac endoprosthesis · Pelvic surgery

Introduction

Uretero-iliac artery fistula (UIAF) is a recognized but uncommon condition due to multiple factors. Few cases are described in the literature (< 150) [1], but incidence is increased in the last years, since at the beginning of 1990s only 20 cases were described [2]. Diagnosis and management are still a challenge for urologist [2]. Uretero-arterial fistulas can develop with aorta, common iliac artery, external iliac artery and hypogastric artery. Aortic fistulas are usually associated with aneurysms; hypogastric fistulas are very uncommon. Fistulas between ureter and common or external iliac artery are usually associated with a history of pelvic surgery, pelvic irradiation, chronic ureteral stenting and vascular disease [3]. The presentation is usually massive gross hematuria with acute anemia unto to hemorrhagic shock, representing a life-threatening condition. High mortality rate is reported (7–23%) in literature [4]. However, endovascular techniques have improved morbidity and mortality [3,

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4]. Gold standard for diagnosis are angiography and computed tomography (CT) scan but, however, these methods have a low sensibility when fistula is too small, and the diagnosis can be misunderstood; in these cases, patients often undergo explorative laparotomy [5]. The aim of this study is to describe four cases of UIAF in three patients treated by classical and endovascular approach.

Materials and methods

This single-center, retrospective observational study was based on data obtained from patients' medical records from 2013 to 2018 in our institution. We collected four consecutive cases in three patients who were diagnosed with UIAF. UIAF was defined as the ratified presence of an abnormal communication between the ureter and any artery. In all patients, the UIAF was initially evaluated by contrast-enhanced CT angiography. The management strategy was defined individually based on the specific risk profile of each patient. Clinical characteristics, diagnosis, management and follow-up of patients affected by UIAF were recorded in an Excel database and were analyzed retrospectively using descriptive analysis.

Results

All patients were female. Mean age was 66 (57–83). All patients underwent radical hysterectomy and radiation therapy for cervical carcinoma 13 (11–15) years before. Two patients underwent radical cystectomy with Bricker diversion for actinic cystitis. All patients had bilateral ureteral stent for stenosis of ureter (one patient) or ureter-iliac anastomosis stenosis (two patients). According to our database, we do not know exactly how many years the patients had stents. Presentation was the same for all four cases: massive gross hematuria during periodic change of ureteral stent. In our Institution we used silicone stents and we perform the periodical change every 3 months with a fluoroscopy-guided out-patient procedure; the technique used is the placement of a guidewire into the previous stent, the subsequent removal of the stent and the placement of the new stent on the guidewire. In every case, the massive hematuria presented during the removal of the stent after the guidewire placement. Immediate management was the stabilization of vital parameters with infusion of colloidal solutions and adrenergic drugs. Blood transfusions were necessities in all cases. Hematuria was temporary auto-resolved in all cases. Two patients were submitted to a CT-scan with delayed ureteral phase, finding clots in renal pelvis which, along with hemodynamical instability, raised the suspicion of active bleeding from the renal parenchyma, without identifying an arterial

vessel to be embolized. For these reason, a right radical nephrectomy was performed (Fig. 1). After radical nephrectomy, both 2 patients had a massive hemorrhage from the drain in the first post-operative day and angiography finally reported the fistula between distal tract of right ureter, left in place during nephrectomy, and right common iliac artery. One of these patients 3 years after developed a contralateral fistula in the left side, diagnosed with massive hematuria during change of the left ureteral stent, but angiography and CT scan did not show the fistula. The other patient had diagnosis through angiography (Fig. 2) during second episode of massive hematuria because in the first presentation angiography and CT scan resulted negative. In all four cases, after the detection of the UIAF, a vascular procedure was performed, and an endo-prosthesis was put in common iliac artery to solve the fistula; endovascular procedures resulted safe, without complications; observation of patients was continued in hospital for 18 (5–21) days. Ureteral stent was removed in all cases and a nephrostomy tube was placed to drain the kidney. After placement of endoprosthesis, one patient underwent massive deep vein thrombosis despite low molecular weight heparin prophylaxis; an inferior caval vein filter was placed and removed after 3 months of anticoagulant therapy. Follow-up is of 49 (25–66) months and resulted uneventful in all patients (Table 1).

Discussion

In our experience, UIAF occurred in all cases in female patients with previous medical history of hysterectomy and radiation therapy for cervical carcinoma. In the literature,

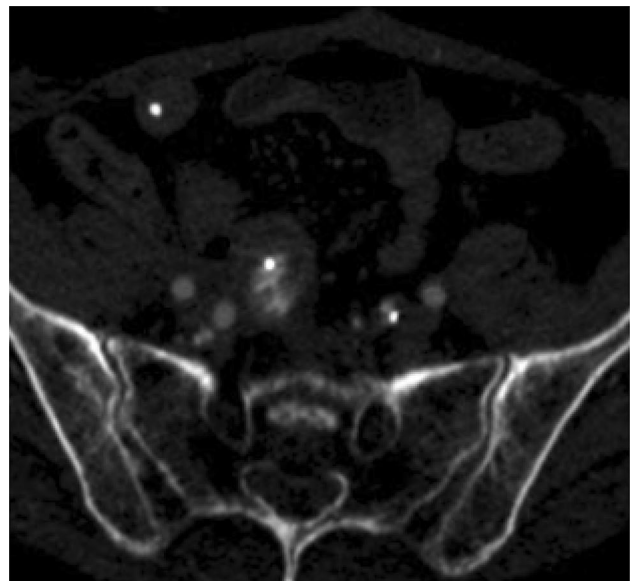


Fig. 1 Intra-ureteral contrast medium in areterial phase of CT-Scan

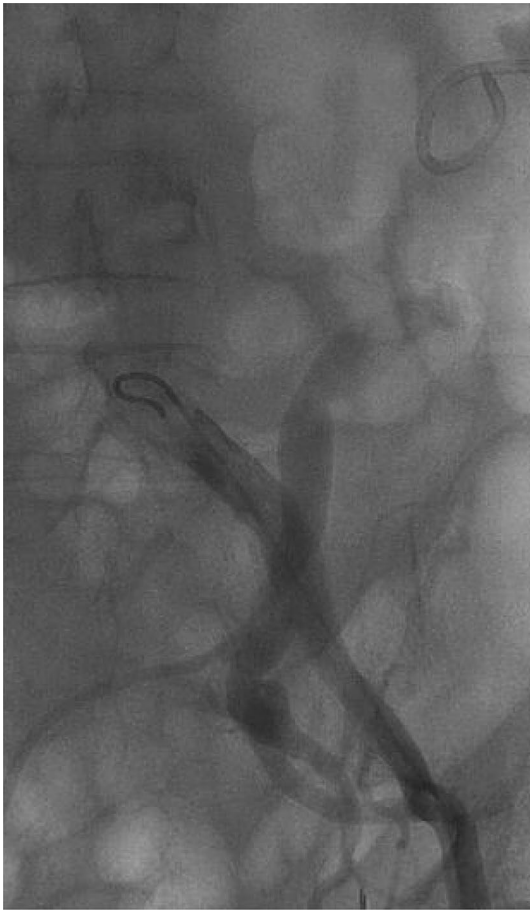


Fig. 2 Left ureter-iliac artery fistula detected during angiograph

57% of UIAF occur in women at a mean age of 58 years [6]. Two patients underwent also a Bricker diversion for actinic cystitis. All patients had an indwelling stent for

stenosis of ureter or ureteral-iliac anastomosis. We know that radiation is a risk factor for stenosis of uretero-iliac anastomosis in Bricker diversion and the ureteral stent is the most adopted treatment. The presence of an indwelling stent and periodical change is a risk factor for inflammation, traumatic lesions, infections and hematuria; the risk is increased in radiated patients [7]. The ureter crossing iliac vessels makes a turn and is narrower than other tracts: it may suffer more traumatism during stent change. Furthermore, radiated tissues are less resistant and softer and are predisposed to more damage. In our cases those factors can have contributed to lesion the ureter forming fistula with the common iliac artery. Maybe also the constancy and the hardness of stent may had contributed to increase the damage. Gross hematuria was episodic. One possible explanation is the increase of blood pressure in some moments, for example due to anxiety for the change of stent, made as outpatient procedure without anesthesia, a possible valve mechanism of fistula can explain passage of blood with arterial pressure from artery to ureter overcoming the flap of valve. Another possible explanation is the presence of stent compressing the fistula and preventing the continuous passage of blood. Those reasons can explain the difficulty to detect fistula with angiography when hematuria is temporary solved; the false-negative result during diagnosis is due to the closing of the flap of valve in that definite moment. In our experience blood refluxed in renal pelvis providing a false positive for kidney hemorrhage and led to radical nephrectomy in two cases; also, chronic pyelonephritis (probably due to a chronic presence of a stent) increased the false imaging of a bleeding kidney. During nephrectomy, ureter was cut proximal to the cross with iliac artery because absence of suspicious of the fistula and without

Table 1 Clinical characteristics, diagnostic modalities, management strategies, and clinical outcomes in the three cases study patients affected by UIAF

	Patient 1, 1st case	Patient 1, 2nd case	Patient 2	Patient 3
Age	66	68	57	73
Hx of pelvic radiation therapy	Yes	Yes	Yes	Yes
Hx of pelvic surgery	Yes	Yes	Yes	Yes
Urinary diversion	Ileal conduit	Ileal conduit	No	Ileal conduit
Level of stenosis	Anastomosis	Anastomosis	Pelvic urether	Anastomosis
Indwelling urethral catheter	Single-J	Single-J	Double-J	Single-J
Side	Right	Left	Right	Left
1st CT scan	Negative	Negative	Negative	Negative
Angiography	Positive	Negative	Negative	Positive
Therapy	Endoprosthesis	Endoprosthesis	Endoprosthesis	Endoprosthesis
Diversion subsequent	Nephrostomy tube	Nephrostomy tube	Nephrostomy tube	Nephrostomy tube
Follow-up (months)	66	42	63	25
Recurrence	Controlateral	No	No	No

Table 2 Revision of the literature and comparison with our experience

Authors	Araki et al.	Krambeck et al.	Okada et al.	Fox et al.	Our experience
Cases	2	7	11	20	4
Risk factors					
Chronic indwelling stents	100%	100%	91%	84%	100%
Pelvic surgery	100%	100%	73%	100%	100%
Pelvic radiation	0%	100%	45%	74%	100%
Diagnosis					
CT scan	Negative	Negative	Positive in 55%	–	Negative in 100%
Angiography	Positive in 100%	Positive in 63%	Positive in 45%	–	Positive in 50%
Treatment					
Endovascular graft stent	100%	100%	100%	70%	100%
Open repair	0%	0%	0%	30%	0%
Follow-up					
Follow-up (months)	12	–	18	15	49
Recurrence	0%	–	36%	–	0%
Mortality	0%	0%	0%	10%	0%

an history of urothelial cancer. For this reason, after the passage in the ureter, blood went in the renal loggia and drained, leading to repeat CT scan and finally diagnose the fistula. The patient with late contralateral fistula and the other patient with negative first angiography, underwent immediate placement of preventive endoprosthesis relying only on clinical suspicion. Angiography in our experience represents the most valid diagnostic option in emergency because of the possibility to detect immediately the fistula, meanwhile the CT scan can delay diagnosis and treatment. Laparotomy was unnecessary for diagnosis and treatment in our experience, but in many cases reported in the literature it can be an effective method to solve the condition [8]. In cases with achieved hemodynamic control, a multidisciplinary discussion with radiologist, interventional radiologist, vascular surgery is mandatory to settle and solve the problem [9, 10]. Endoprosthesis seems to be very effective: with a mean follow-up of 31 months we did not observe any complication. In addition, in the literature is resulted to be the preferred option [11, 12]. Okada et al. reported 36% of recurrence of hematuria in a series of 11 patients underwent placement of endoprosthesis for UIAF, with need for a surgical approach in two patients [13]. The bigger series reported in the literature, by Fox et al. comparing retrospectively endovascular versus surgical approach, did not show a difference between the two methods, reporting the same rate of recurrence of hematuria, complications and mortality with a follow-up of 15 months [14, 15] (Table 2). Open surgical treatment is still described in the literature, especially in patients with first failed an endovascular treatment or enteric contamination, abscess, and local sepsis, but we did not experience this treatment in our experience. One important topic is

the management of urological condition. After a placement of endoprosthesis, is advisable to remove indwelling ureteral stent to avoid further complications. Nephrostomy tube [16, 17] is the preferred option. In patients with discussed risk factors a softer and smaller stent or a preventive nephrostomy tube can be the best prevention method. Several limitations of our study should be observed. Our data were retrospective in its design and had a low level of evidence. This is, however, inevitable in studies of highly rare status, resulting in a small sample size. Therefore, future studies on larger cohorts of patients are desirable.

Conclusion

Our study identifies different factors that were present in all our patients and might be related to an increased risk of developing UIAFs: concomitant presence of a medical history of previous pelvic malignancy treated with surgery or radiation-therapy, urinary diversion and indwelling ureteral stent in female patients. Gross hematuria during stent change in those patients must be suspected of ureteroarterial fistula and the clinical suspicion is the key factor for immediate diagnosis of this urological emergency. Selective angiography is useful to confirm the diagnosis while subsequent endovascular endoprosthesis represents a safe approach with reduced post-procedural complications.

Compliance with ethical standards

Conflict of interest The authors all declare no competing financial or personal interests.

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