Uretero-Arterial Fistula – Not So Rare?

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Abstract
The first uretero-arterial fistula (UAF) was reported in 1908 by Moschcowitz [1]. In 2009, a systematic review identified 139 cases. Since then a further 23 cases were described with 19 cases originating from a single center. It has been recognized as a very rare condition in the past. However, more recently, the increasing incidence of UAF has led us to believe that this condition is more frequent than previously described. Aging population, improved cancer survival and extensive multimodal pelvic cancer treatments have been recognized as culprits for the increased incidence of UAFs. We present a case of fistulous communication between the internal iliac artery and ureter in a patient with a potential risk factor previously not described in the literature.

Case Report
A 77-year-old man who had obstructive uropathy due to retroperitoneal fibrosis presented to the emergency department, with visible hematuria associated with suprapubic pain and difficulties in passing urine. He was admitted to the urological ward and a 3-way urethral catheter with continuous irrigation was inserted. The patient had a history of chronic kidney disease (stage IV), hypertension, ischemic heart disease and previous cardiac stenting, transient ischemic attack, hyperparathyroidism.

Following admission, he continued to have intermittent visible hematuria, which intensified 4 days later where Hb level dropped to 80 g/l requiring transfusion of 4 units of blood. Subsequent CT revealed ureteric stents in situ with mild left sided hydronephrosis and no retroperitoneal bleed. The bladder was partially filled with blood clot. It was not possible to identify the lower end of the stent due to the clot.

The patient was taken to the operating theater for urgent cystoscopy and bladder wash out. During cystoscopy, the stents were removed. A perforation was noted on the posterior bladder wall.
Open surgical exploration was then undertaken. Perforation repair was done using double layer 2.0 Vicryl closure. Postoperatively the patient was transferred to intensive therapy unit.

Six hours later he became hemodynamically unstable with a systolic blood pressure of 40 mmHg accompanied by gross hematuria. Therefore an emergency cystoscopy was performed. A large amount of blood was noted emerging from the left ureteric orifice. In view of this it was decided to perform an emergency nephrectomy. The procedure was somewhat difficult due to multiple perinephric and retroperitoneal adhesions. At this stage, the patient developed cardiac arrest and died.

Post mortem examination revealed a 2 mm hole in the lower third of the left ureter connected to the adjacent severely atheromatous aorta forming a fistula. Both aorta and ureter were encased by thick fibrous tissue (fig. 1).

**Discussion**

**Risk Factors and Pathophysiology of UAF**

The commonest risk factors associated with UAFs were ureteric or vascular stenting, history of abdominopelvic cancer or vascular surgery and radiotherapy. In the literature, neoplastic conditions associated with development of UAF included bladder, cervical, colorectal, endometrial, prostate and vaginal cancers as well as lymphomas and sarcomas including osteosarcoma. The pathological changes described in the post mortem examinations allowed investigators to formulate the mechanism responsible for UAF evolvement. High pressures and pulsations present in the artery and subsequent ischemic microvascular damage of the vasa vasorum lead to the necrosis at the junction were artery crosses ureter. Together with surgery or radiation related fibrosis and fixation of stented ureter at the crossover junction, pressure necrosis develops. We believe that in our case, the idiopathic retroperitoneal fibrosis resulted in partial obstruction of the adjacent ureter leading to the subsequent requirement for repeated stenting. This then played an important role in predisposing to local damage and ischemia culminating in fistula formation.

**Symptoms and Diagnosis**

Commonest presenting symptom includes hematuria (up to 62%), which is usually intermittent and can be microscopic or visible. Other symptoms include associated urinary tract infection (10%) and lumbar or loin pain (17%), incidental finding of hydronephrosis (10%) or presence of a pseudo or true aneurysm of the external or common iliac artery (38%) [2, 5].

When UAFs are suspected, in a context of clinical triad of pelvic surgery, radiation and ureteric stenting, the best diagnostic modality seems to be pelvic angiography with or without provocation [6]. Retrograde pyelography and cystoscopy due to low sensitivity of around 60%, is not recommended [7]. In order to demonstrate the UAF on retrograde pyelography requires a sufficient pressure gradient from ureter to artery, which may not be possible in the presence of hematuria. Some authors report semi-rigid ureteroscopy as a diagnostic modality [8]. From our experience, we do not recommend ureteroscopy because it can dislodge a tamponading clot or tear the fistula, leading to massive hemorrhage. CT scan has been reported as low to moderately sensitive diagnostic tool in the literature [9].

**Treatment**

Over the years treatment options evolved due to advancement of minimally invasive techniques in the field of interventional radiology. There are multiple options available when considering UAF’s repair. In principle, the treatment must include repair of the ureter and artery. The vascular pathology can be managed with open surgical repair, simple ligation of vessel, bypass, arterial embolization and repair of artery with graft material [3]. However, the most optimal treatment option seems to be endovascular treatment due to low risk of hematuria recurrence and low risk of graft infection [10]. The options for repair of the ureteral defect include ureteric stenting, nephrectomy with ureteric ligation or nephroureterectomy, nephrostomy insertion, ureteral resection with primary anastomosis or ileal ureteric reconstruction.
Commonly experienced complication in these patients is poor wound healing owing to prior pelvic surgery and radiotherapy.

**Conclusion**

In summary, uretero-iliac fistula is a clinical presentation that physicians need to consider in patients with the triad of radical pelvic surgery, pelvic radiation, idiopathic retroperitoneal fibrosis and situations requiring repeated ureteric stenting. Treatment methods have evolved over the years and the prompt diagnosis and management increases the chances of survival in these high-risk patients.

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