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## Arterio-ureteral Fistula – a Systematic Review

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### Introduction

A rare but nonetheless important cause of recurrent gross haematuria is a fistula from an artery into the urinary outflow tract. The condition is difficult to diagnose and the morbidity and mortality high. One important prerequisite for a better prognosis is to be aware of the condition. The aim of this review, therefore, is to summarise present knowledge.

### Method

The literature was searched through Medline and through references in previous publications. Except for two Italian, four Spanish and five Japanese papers the literature has been scrutinized and the review is based on this collective experience. A patient reported by Hodges<sup>1</sup> referred to in several later publications seems to be ureteral obstruction by an aneurysm and not a regular fistula with bleeding into the ureter. Although stated in the title nothing in the case report indicates the presence of an arterio-ureteral fistula in

the patient described.<sup>2</sup> Post-traumatic cases have been excluded.<sup>3–5</sup> One patient with a fistula between the iliac vein and the ureter has been described,<sup>6</sup> but is not discussed further here. Eighty cases have been reported.

### Etiology and Pathogenesis

Arterio-ureteral fistula can be classified into primary (15%) and secondary (85%) to treatment procedures. There were 45 female and 35 male patients, of median age 58 years (range 18–83 years). Three cases occurring during pregnancy will be discussed separately.<sup>7–9</sup>

#### Primary fistula

Of the ten primary fistulae, nine have been caused by an aortoiliac aneurysm.<sup>10–18</sup> The patient without an aneurysm had an arteriovenous malformation.<sup>19</sup> Three aneurysm patients also had previous pelvic surgery,<sup>12,16–18</sup> but the report points to a primary fistula. One case was a classical aneurysm rupture with shock and haematuria. Peroperatively an internal iliac aneurysm rupture was found, extending into the ureter.<sup>18</sup>

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**Table 1. Operations with secondary arterioureteral fistulas.**

	No of patients
Pelvic surgery with radiation	42
Vascular reconstructive surgery	18
Other pelvic procedures	6
Surgery for stenosis of ureteropelvic junction	1
Transplantation	1

### *Secondary fistula*

The majority of fistulas are secondary (Table 1), mainly to radiation and extensive surgery for urological or gynaecological cancer. The delay between surgery and haematuria was a median of 2 years (2 months to 30 years) for the patients with malignant disease and 10 years (3 months to 25 years) for the patient with previous vascular surgery, all of whom had been reconstructed with a synthetic graft. Most patients had a complicated course during or after the first procedure. Many cases had outflow obstruction causing hydronephrosis with need for ureteral catheters and stents, often with frequent changes (70%) or reoperations. Ureteral necrosis may predispose to development of a catheter fistula.<sup>20</sup> Repeated dilatations of ureteral strictures had been performed in many of the patients in combination with catheterisation and stenting. Twenty-nine patients (42%) had various types of urinary diversion. In one patient the fistula was seen between a graft artery aneurysm and the native ureter after kidney transplantation.<sup>21</sup>

### *Fistula during pregnancy*

All three cases date from some time ago and all patients had demonstrated a massive urinary tract infection with septic complications. Two of them had had ureteral catheterisation, and all three demonstrated massive urinary haemorrhage. Two patients bled to death and the third died of septic complications. In none of them had the fistula been suspected pre-mortem.

### **Symptoms and Signs**

The dominating symptom is gross haematuria, which was seen in all patients. It can be intermittent and during periods of haematuria, the bleeding may be massive with hypotension and even shock. In one patient the haematuria started during heparinisation

for treatment of deep venous thrombosis.<sup>22</sup> Haematuria was occasionally combined with abdominal or flank pain. A few patients had symptoms of urinary tract infection and presumed pyelonephritis. In patients with a stent or catheter, bleeding may have been provoked or exaggerated while changing the stent/catheter.<sup>23</sup>

### **Investigations**

Often the diagnosis was not considered, which may lead to repeated negative investigations and diagnostic delay.

Cystoscopy can localise the bleeding to one of the ureteral orifices, and if there is a ureteral catheter or stent, extraction may provoke bleeding. If orificial bleeding is pulsating a arterio-ureteral fistula is likely. Massive bleeding from the ureter at cystoscopy can temporarily be blocked with a balloon catheter.<sup>24-26</sup>

Angiography usually is negative but it is important to obtain oblique projections in order to better identify small pseudoaneurysms, which otherwise can be overlooked. On rare occasions extravasation of blood may be demonstrated. The fistula may also be seen but this is exceptional.<sup>27-34</sup> In patients with a ureteral stent, visualization of the fistula may be provoked by stent removal<sup>31,35,36</sup> or by moving the catheter back and forth.<sup>24</sup>

CT of the abdomen and pelvis is usually negative or non-specific and the bleeding is only rarely seen. If contrast is used the fistula may be visualised.<sup>37</sup> The non-specific findings include the presence of a pseudoaneurysm, signs of graft infection or hydronephrosis-ureter.

In some cases a retrograde pyelography has been of diagnostic value. Some authors have reported contrast around the vascular graft which may be considered an indirect but strong sign<sup>38-41</sup> and occasionally direct visualization of the fistula has been reported.<sup>42-44</sup>

In many cases the bleeding has been extensive enough to warrant emergency surgical exploration and the arterio-ureteral fistula has been diagnosed on the operation table.<sup>12,38,42,45-61</sup>

In four cases diagnosis was made first at autopsy.<sup>12,29,62,63</sup>

### **Treatment**

In situations with massive and life-threatening bleeding without a correct diagnosis of an arterio-ureteral

**Table 2. Treatment options used for arterial lesion in case of arterio-ureteral fistula.**

Open procedures
Local reconstruction (arteriorrhaphy, patch closure, interposition graft, bypass)
Ligature with or without extra-anatomic reconstruction
Ligature of the internal iliac artery
Autotransplantation
Endovascular procedures
Embolisation of the internal iliac artery and a polar renal artery
Embolisation with or without extra-anatomic reconstruction
Graft covered stent

fistula, most patients have been treated with nephro-ureterectomy. Obviously the source of bleeding is not removed if only a nephrectomy is done and in such cases a secondary ureterectomy may be necessary before adequate haemostasis is obtained.<sup>31,41,48,64</sup>

The treatment plan must deal both with the arterial lesion and the urinary outflow and a report will follow regarding the treatment options used. One problem with the evaluation of different treatments is that the follow-up to short or poorly documented.

Either a nephrectomy was performed or equally often an attempt to reconstruct the ureter or performing a ureterostomy. There are some patients where a pyelonephrostomy was performed<sup>41,56-59,65</sup> and in a couple of cases the ureter was simply ligated.<sup>64,66</sup>

The various arterial treatment procedures used in case of arterio-ureteral fistula are summarised in Table 2. In two reports it is not indicated how the arterial lesion was handled.<sup>48,54</sup> Surgical exploration and a local reconstruction with just closure of the defect or using a patch or a graft has dominated but in a few cases closure of the artery or removal of an earlier graft without reconstruction has been performed with only mild symptoms of ischaemia.<sup>39,40,55,60</sup> A local reconstruction has sometimes been covered with omental tissue.<sup>4,12</sup> Local ligation and extra-anatomic reconstruction (most often a femoro-femoral cross-over bypass) seem to be a logical procedure because of the potential risk for an infection as there has been a contact with the urinary tract. In a few patients the fistula has emanated from the internal iliac artery and in these cases simple ligation has been performed. In one patient the nephrectomised kidney was auto-transplanted.<sup>51</sup>

In recent years there has been an increasing use of endovascular procedures such as embolisation of the internal iliac artery,<sup>31,42,67</sup> embolisation of a rare case with fistula from a polar renal artery to the ureter,<sup>32</sup> embolisation or permanent balloon occlusion of the common/external iliac system in combination with a femoro-femoral cross-over bypass<sup>24,25,31,34,35,68</sup> or the use

of a graft covered stent.<sup>26,27,33,67,69</sup> However, a recurrence also may be a problem with the latter treatment.<sup>69</sup>

## Results

Of the 57 patients treated interventional seven died during the first postoperative month (procedure mortality 12%). Before 1980 the mortality was 60% and after 1980 7.1%. A correct pre-operative diagnosis is of great importance. Of those 41 operated on with a diagnosis of or a strong suspicion of a fistula the mortality was zero. Of those 18 where diagnosis became clear at surgery seven (39%) died. In 22 cases there is no follow-up defined. Of the remaining, eleven died at a median of 6 months postoperatively (range 2–11 months), the majority from their underlying malignancy or infectious complications. The median follow-up time of the survivors was 10 months (range 1–54). In all patients, during their follow-up, the haematuria ceased.

## Comments

Arterio-ureteral fistulation is rare, especially primary cases which are seen almost exclusively as a complication to aortoiliac aneurysmal disease. The secondary or iatrogenic fistulas, after various types of pelvic interventions, have increased rather rapidly in frequency, probably as a consequence of an increased number of vascular reconstructions and more advanced and extensive treatments for pelvic malignancies. An increasing use of indwelling rigid ureteral catheters and stents as well as urinary tract diversionary procedures also contributes. One type of fistula has disappeared, the ones seen during pregnancy with pyelonephritic obstructed kidneys. Flexible catheters and antibiotic treatment seem to have prevented this catastrophic complication.

In the majority of patients, the anatomic localisation of the fistulas has been at the arterial crossing in the pelvic rim, that is between the ureter and the common iliac artery. In one case the fistula has gone to the bladder.<sup>70</sup> A few have engaged the internal iliac artery and in one case a polar renal artery after Acusise endopyelotomy.<sup>32</sup>

There are several situations with a risk for arterio-ureteral fistula development:

- prolonged use of ureteral stents or catheters, especially rigid ones;
- a ureteral stump after nephrectomy;

- vascular reconstructive surgery, especially with anastomosis or pseudoaneurysm in the pelvis;
- radiation therapy, although only in combination with surgery;
- surgery for malignant diseases, especially uterine cancer or transitional cell cancer of the bladder;
- ureterolithotomy complicated with a urinary leakage;
- rigid drainage tubes placed towards the ureteroarterial crossing.

The pathophysiology is not clear in all details but the inflammatory reaction caused by surgery, radiation and/or urinary leakage may cause a relative fixation of the ureter to the artery or a vascular graft with repeated pulsatile trauma. Both a synthetic vascular graft and a rigid ureteral stent may increase the risk for pressure necrosis. In some patients the ureter is incorporated into fibrotic perigraft tissue causing ureteral obstruction with a hydroureter and sometimes ureteral fistulas.<sup>45,71–75</sup> The ureter may be rather thin-walled. Many of the conditions with risk for an arterio-ureteral fistula have as a common denominator ischaemia of the ureter (pressure, necrosis, surgical mobilisation, radiation, chronic infection, fibrosis). Meticulous surgical technique to preserve ureteral vascular supply is therefore important.

The main symptom is haematuria, often intermittent, and after a varying number of bleedings a massive life-threatening bleeding might occur, which also in fact may be fatal, before diagnosis and treatment. The reason why some patients experience pain is probably due to clot formation within the renal pelvis and ureter.<sup>23</sup> The initial small bleedings may be compared with herald bleedings seen in patients with arteriointestinal fistula.<sup>76,77</sup> During quiescent periods, a clot occludes the communication between the artery and the ureter and intestine respectively. After degeneration by proteolytic enzymes the bleeding will recur.

Because of the rare occurrence, diagnosis is difficult and in patients with gross haematuria a high index of suspicion is therefore important and the possibility must be considered in the differential diagnostic thinking. A preoperative diagnosis is of great importance to decrease mortality.

Unfortunately there are no certain diagnostic methods. Especially in patients with synthetic grafts they may be seen bathing in contrast at retrograde pyelography. To be seen at angiography there must be a certain amount of bleeding. In patients with a ureteral catheter or stent, bleeding may be provoked by movement or extraction of a balloon catheter. If this is to be tried there must be the possibility to block

the haemorrhage by a balloon catheter and there must also be a vascular surgical backup. When the suspicion has been brought up it is important to be diagnostically aggressive preoperatively.

What type of arterial reconstruction should be used depends on whether there is an infection or not. This is, however, difficult to exclude with a fistulation to the urinary system. To be on the safe side an extraanatomic reconstruction is therefore probably correct whether the fistula is closed by an open procedure or through endovascular embolisation (coil springs, detachable balloons etc). In a few patients endovascular graft covered stents have been used which is an attractive treatment option in the short run but follow-up time is still very short. Late infectious complications might occur. There is also a risk for refistulation. Only few cases treated in this way has been reported and with short-term follow-ups.

One problem with the majority of publications is the poor routine for reporting results, another is the rather short follow-ups in those who are followed. The haematuria has been effectively stopped with all treatment options but the prize has been a high postoperative morbidity and mortality although it has decreased considerably during the two last decades. Without treatment, however, the mortality is 100%.

To avoid the complication, chronic treatment with rigid ureteral catheters and stents should be avoided, especially in patients with previous pelvic irradiation. When performing aortofemoral reconstructions with synthetic grafts the tunnelling procedure should aim at placing the ureter in front of the graft to avoid compression of it between the native artery and the graft.

In conclusion, arterio-ureteral fistula is a challenging complication where resources should be optimised with a close cooperation between the vascular surgeon, interventional radiologist and urologist.

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