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Case Report

Ureteroarterial fistula treated by endovascular stent placement ☆,☆☆,★

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ABSTRACT

A 69-year-old woman with changes secondary to pelvic radiotherapy presents repeated episodes of massive hematuria associated with double J catheter replacements. After several imaging tests, an uretero-arterial fistula is confirmed by angiography and treated with a coated stent. Uretero-arterial fistula poses a diagnostic challenge, requiring a multidisciplinary approach through clinical suspicion and interventional procedures.

Introduction

Uretero-arterial fistula is a rare but potentially life-threatening condition, which pose a diagnostic challenge due to its low incidence, scarce clinical expression and difficult diagnosis [2,3]. The purpose of this work is to raise the awareness of this pathology among radiologists, as well as its diagnosis and endovascular management.

Case report

Our patient was a 69-year-old woman with history of cervical cancer treated with hysterectomy and double adnexectomy, lymphadenectomy and omentectomy along with adjuvant pelvic radiotherapy. As a result of radiotherapy, the patient developed atrophy of the left kidney and later right obstructive uropathy, requiring placement of an indwelling double J catheter with scheduled replacements every 6 months.

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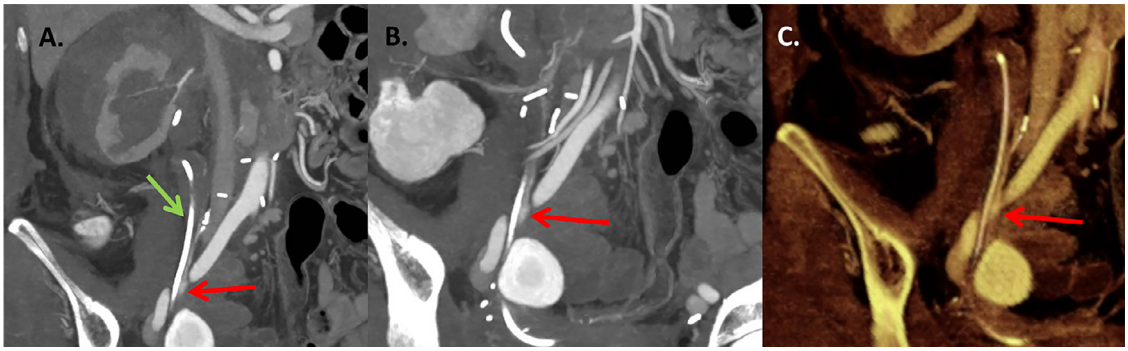

Fig. 1 – Arterial phase contrast-enhanced CT with MIP (A, B) and 3D (C) reconstructions (June 23rd). Intimate contact between the ureter with double J catheter (green arrow) and the right external iliac artery, showing a focal linear contrast extravasation (which wasn't identified in first approach). These findings allow the diagnosis of uretero-arterial fistula (red arrow). Note the right renal hematoma and urinoma on image A.

In last scheduled catheter replacement (June 21st), there was an intraoperative ureteral bleeding immediately after the removal of the double J catheter; a high-debit, pulsatile and incoercible bleeding that stopped only minutes after a new catheter was placed. The episode caused anemization of up to 6.1 g/dL of hemoglobin and secondary hemodynamic instability. Two red blood cell concentrates were transfused and urgent computerized tomography (CT) scan was performed, with and without intravenous contrast, with acquisition in arterial and venous phases. CT scan showed severe right ureterohydronephrosis with diffuse urothelial enhancement but no signs of active bleeding.

Two days later, the patient presented a progressive worsening of renal function with acute elevation of reactants (leukocytosis >20,000, PCR 400, Procalcitonin 85). An urgent nephrostomy was performed, which was carried out without complications. After 24 hours, a new episode of bleeding through the nephrostomy of up to 400 mL was observed. A new CT scan was indicated on June 23. The CT scan (Fig. 1) showed a large right urinoma and a renal subcapsular hematoma, but with no evidence of contrast extravasation from renal arteries, thus suggesting a parenchymatous cause for the bleeding. However, in the arterial phase, contrast appeared in the lumen of distal ureter. So, bleeding of renal or ureteral artery origin was suspected.

In the light of these findings, a selective arteriography was performed, showing two 3 mm in diameter pseudoaneurysms located in the right renal subcapsular region, adjacent to the nephrostomy site. Given their size, we considered them incidental findings, probably secondary to manipulation, not justifying the clinical-radiological findings. Nonetheless, they were embolized.

In spite of patient's initial favorable evolution, during a new attempt to remove the double J in the operating theatre on July 3, the patient presented intermittent anemic hematuria and even a massive bleeding episode (identical to the previously observed) which severely worsened the patient's clinical status.

These findings forced the meeting of a multidisciplinary committee consisting of urologists, interventional vascular radiologists and vascular surgeons who reviewed the case record and the images. Images showed a ureter crossing the right external iliac artery. So, uretero-arterial fistula was considered as the first diagnostic possibility.

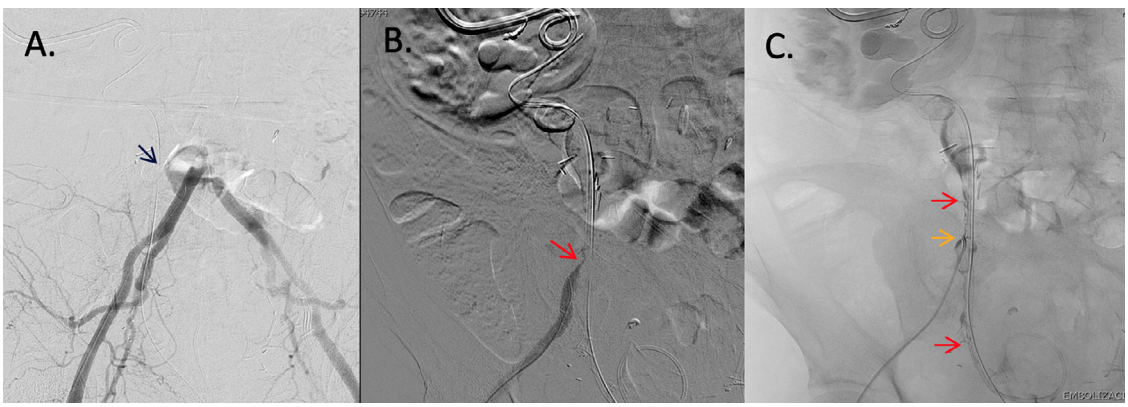

Fig. 2 – Initial angiography on July 4 (A) from aortic bifurcation where both iliac branches are well contrasted, without evidence of fistula. Double J catheter in right ureter (black arrow). Selective arteriography (B and C) of the right external iliac showing contrast extravasation to the ureter (red arrows) after "to-and-fro" movements. Tip of the Cobra catheter inserted at the proximity of the fistulous trajectory (C, orange arrow).

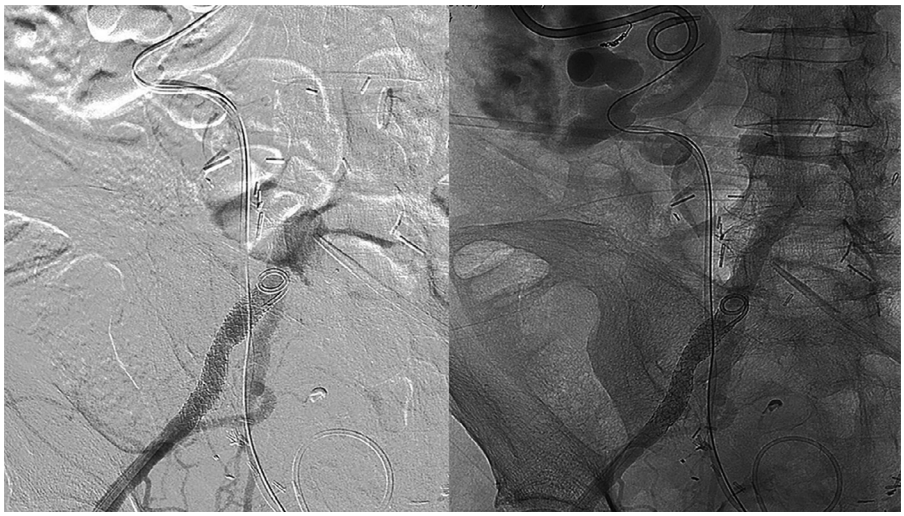

Fig. 3 – Angiography after endovascular treatment with expandable PTFE-coated stent in external iliac artery at the junction with the ureter, preventing occlusion of the right hypogastric artery. There is no contrast flow through the fistulous trajectory.

Accordingly, a diagnostic angiography and several provocation tests at the crossing point were performed. After several attempts it was possible to channel the fistulous tract through a Cobra catheter demonstrating the existence of an ureteroarterial fistula (Fig. 2).

A polytetrafluoroethylene covered prosthesis (BeGraft peripheral Stent Graft System; Bentley InnoMed GmbH, Hechingen, Germany), 10 mm in diameter and 57 mm in length, expandable with a balloon, was implanted using a 0.035" guide in the right external iliac artery. The closure of the fistulous trajectory was checked. No treatment of the ureteral component was attempted.

Hematuria progressively improved until it disappeared. Having a good diuresis and hemodynamic stability, our patient was finally discharged with a balloon nephrostomy. One month later the double-J catheter was removed without complications.

Discussion

The interest of this case lies in its rarity. Uretero-arterial fistula is a very uncommon condition (90 cases reported in English-language literature) [2] that is not only challenging to diagnose but also is potentially lethal (7%-23% mortality rate) [3,4]. Its incidence is increasing (5.7) as improvement in treatment of pelvic cancers have managed to increase life expectancy. Uretero-arterial fistulas are classified in primary (15%) and secondary (85%) [1,2]: first ones are due to primary pathology of the arterial system, such as aneurysms, vascular malformations or aberrant vessels. Secondary uretero-arterial fistulas are more frequent in women about 60-year old with history of abdominal surgery combined with pelvic radiation (mainly for gynecological, rectal or urological cancers), and carrying indwelling ureteral catheters [1], as in our case. Usually starts with inflammation and fibrosis of the ureteral wall caused by the catheter, which causes ureter fixation to adjacent artery. Radiation causes ischemia of the arterial wall which becomes more friable and prone to erosion by the ureteral catheter [2]. Eventually high blood pressure is transmitted to the uretero-arterial wall causing necrosis and thus creating a fistula. The most frequently involved arteries are the common and external iliac arteries, although fistulas are also described between aorta and internal iliac artery.

The clinical presentation in this pathology is not very representative, as it usually manifests itself with hematuria, hydronephrosis [1] and/or pain. Hematuria can be very variable (from intermittent to hemorrhagic shock) [1]. Although it is usually spontaneous, worsening with manipulation of the ureteral catheter is very characteristic. Therefore, uretero-arterial fistula should be considered in all massive and pulsatile hematuria appearing during ureteral catheter replacement.

Role of imaging tests in the diagnosis of uretero-arterial fistula is limited. CT scans [9] are usually negative, nonspecific and difficult to interpret [2,3]. The presence of pseudoaneurysms at the fistula site (up to 38% of cases) [3], clots in the excretory system and hydronephrosis favor suspicion. The most sensitive diagnostic technique is selective iliac arteriography [1–3,9]. However, its sensitivity is low, around 50% (probably due to occlusion of the fistulous orifice by a clot), and it is often necessary to resort to provocation tests such as friction of the ureteral catheter by “to-and-fro” movements and movements of the intraarterial guide(6).

Although there are no long-term follow-up studies after endovascular treatment of uretero-arterial fistulas [4], most authors agree that placement of coated stents is the treatment of choice [1–3,5,6,8], without needing adjuvant ureteral therapy. With this technique, mortality due to bleeding in uretero-arterial fistula has decreased from 60% to 13% [3].

In conclusion, uretero-arterial fistula is a rare condition that should be considered when a massive, pulsatile ureteral

bleeding appears during catheter removal in a patient with history of pelvic radiotherapy [1,10]. This diagnosis should be confirmed by selective arteriography. The treatment of choice is the endovascular placement of a coated stent [7].

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