

Oncology

Silent arterio-ureteral fistula: Diagnosis and endovascular management

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ABSTRACT

We report a patient with history of recurrent invasive rectal adenocarcinoma complicated by obstructive uropathy requiring nephro-ureteral catheter placement. Two years later during a regular catheter exchange, the patient developed unusually bloody urine raising suspicion for possible vascular injury. CT angiogram and conventional angiogram were negative. However, an antegrade nephrostogram revealed a fistulous communication between the right ureter and the right internal iliac artery. Subsequently, the artery was sacrificed using detachable coils. We discuss the rare encounter of a silent arterio-ureteral fistula, the value of antegrade nephrostogram as a diagnostic tool, and the management options in such scenario.

1. Introduction

Arterio-ureteral fistula (AUF) is a severe and rare medical condition characterized by an abnormal connection between a ureter and the common iliac, internal iliac, or external iliac arteries. The majority of AUFs are either oncological or iatrogenic, brought on by urological operations, trauma, or surgical procedures. The growing prevalence of ureteral stents usage is also contributing to the rising occurrence of AUFs.¹ The symptoms of AUFs are often vague and can resemble the presentation of other urological or vascular disorders, making the diagnosis challenging. Contrast-enhanced computed tomography (CT) can detect various useful findings such as extravasation, pseudoaneurysms, hydronephrosis, and opacification of ureters, which suggest the presence of AUF.²

In our case, we describe the discovery of a silent AUF that remained asymptomatic until incidentally suspected during a routine nephro-ureteral catheter exchange. Intriguingly, it was not seen on CT angiogram and conventional angiogram however confirmed on antegrade nephrostogram, and eventually successfully managed by endovascular sacrifice of the internal iliac artery.

2. Case presentation

The patient is a female with history of recurrent rectal adenocarcinoma, initially treated with systemic chemotherapy (Capecitabine + Oxaliplatin) and radiation therapy followed by surgical resection. Seven years later the patient developed isolated pelvic recurrence with invasion of the cervix, vagina, and uterus, requiring concurrent chemo-

radiation followed by resection. At the time of surgery, double-J stents were placed by urology for ureteral identification. Prior to stent removal, the patient developed obstructive uropathy requiring the placement of bilateral percutaneous nephro-ureteral catheters (PCNU). The catheters were regularly exchanged every 2 months to ensure patency and reduce the risk of infection. Two years later during a routine exchange, it was noted that the patient had dark bloody urine draining from the right catheter site and through her urethra. The patient remained hemodynamically stable.

3. Investigations

An urgent CT Angiogram was performed to evaluate for possible injury and potential source of bleeding. The CTA was negative, however, the right PCNU was seen coursing in close proximity to the right internal iliac artery (Fig. 1). Due to the high clinical suspicion for a possible AUF, the patient was scheduled for a pelvic angiogram.

4. Treatment

Under general anesthesia, the patient was placed in a prone position to facilitate performing the angiogram and simultaneous nephrostogram through the PCNU access. The left radial artery was used to access the arterial system. The right internal iliac artery was sub-selected, and the initial angiogram showed no evidence of vascular injury or an AUF (Fig. 2). At this stage, the right nephro-ureteral catheter was removed, and a 21 cm sheath was placed across the ureter. Pull-back nephrostogram through the sheath showed a fistulous communication between the

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<https://doi.org/10.1016/j.eucr.2024.102712>

Received 18 February 2024; Accepted 15 March 2024

Available online 21 March 2024

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right mid-ureter and the main internal iliac artery (Fig. 3). Attention was shifted back to the arterial side and selective catheterization of the proximal anterior and posterior divisions of the right internal iliac artery was performed. Sequential coil embolization was done using multiple detachable coils (Fig. 4A). Follow-up angiogram showed satisfactory occlusion of the right main internal iliac artery (Fig. 4B). Follow-up nephrostogram showed total occlusion of the fistula (Fig. 5). The right nephro-ureteral catheter was then converted to a nephrostomy to avoid further manipulation and injury. The patient remained hemodynamically stable after the procedure and was discharged from the hospital the next day.

5. Outcome and follow up

Clinical follow-up one month later showed no episodes of bleeding. Follow-up CT scan showed stable position of the coils with no evidence of injury or fistula.

6. Discussion

The causes of AUF can be divided into primary (15%) and secondary (85%) types.³ The primary causes are associated with arterial disease processes like aneurysms, vascular malformations, or abnormal vessels that erode into the ureter.⁴ Secondary causes are usually a result of medical or surgical interventions. Inflammation and fibrosis from pelvic surgery combined with radiation and ureteral stent placement result in fixation of the ureter to the nearby artery.⁵

Patients with a history of pelvic malignancy such as our patient usually are subjected to these risk factors raising their risk for AUF.¹ Radiation causes ischemic injury to the arterial wall by damaging the vasa vasorum of the iliac artery or aorta, and the severity of the ischemic injury is dose-dependent.⁶ Additionally, many of these patients are receiving anticancer treatments that hinder the body's natural healing

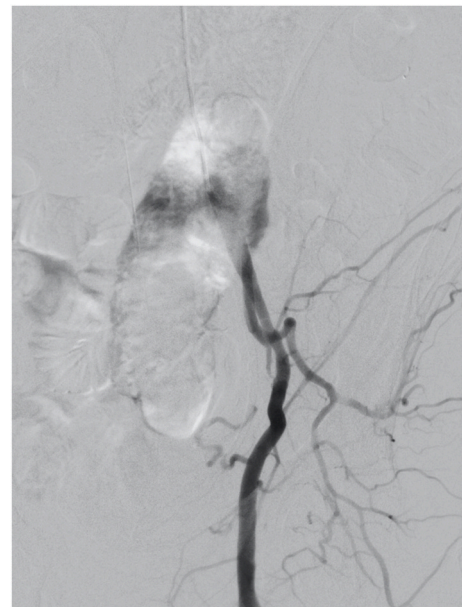


Fig. 2. Initial selective angiogram of the right internal iliac artery shows no evidence of arterio-uretral fistula.

processes.⁷ Pressure necrosis and fistula formation are brought on by high arterial pressure being transmitted to the adjacent wall of the artery and ureter.⁷

In a systemic review published by Kamphorst et al. hematuria was the first presentation in 99% of the patients, with 76% having massive hematuria. The remaining 1% presented with symptoms of claudication or leg ischemia. 13% of patients had associated flank pain.² Our patient

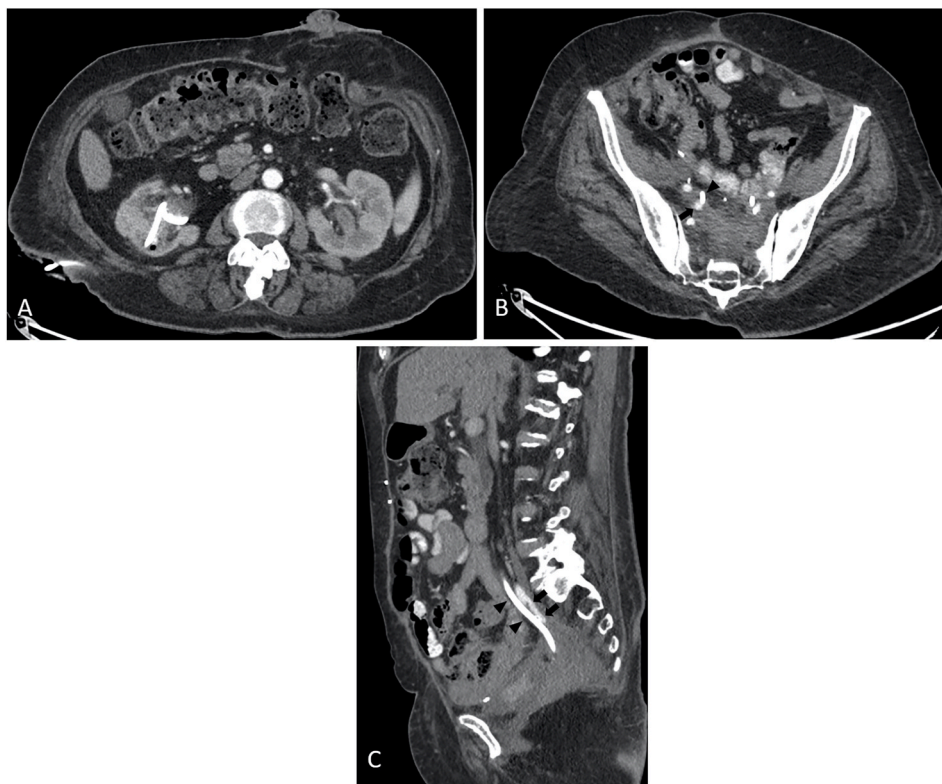


Fig. 1. (A) Axial cut arterial phase CT angiogram of the abdomen and pelvis shows the right nephro-uretral catheter proximal pigtail within the renal pelvis, no evidence of contrast extravasation or vascular injury. (B,C) Axial and sagittal cuts arterial phase CT angiogram of the abdomen and pelvis shows the right Nephro-uretral catheter (Arrowheads) coursing in close proximity to the right internal iliac artery (Arrows).



Fig. 3. Right nephrostogram through a sheath shows simultaneous opacification of the ureter (Arrow heads) and the right internal iliac artery and its distal branches (Arrows) indicating the presence of an arterio-ureteral fistula.

was completely asymptomatic and the AUF would have been completely missed if it wasn't for the change in urine color noted during regular PCNU exchange raising the clinical suspicion for AUF. The key to an early diagnosis and a successful course of treatment is a high index of suspicion in patients with the proper risk factors.⁷

Most patients presenting with hematuria undergo cystoscopic evaluation, which can localize the bleeding site. Pulsatile bleeding is characteristic of AUF. Angiography on the other hand, is considered the best imaging modality for confirming AUF with a sensitivity of 62% followed by CT angiography with a sensitivity of 48%.² In 25 case reports, "provocative angiography" has been described as the manipulation of a vascular catheter or ureteral stent at the suspected AUF site. In approximately 80% of these cases, the provocative procedure was

helpful in confirming AUF.² In our case antegrade nephrostogram confirmed the diagnosis of AUF and localized the site of the fistula hence facilitating proper management, and hereby avoiding provocative arterial manipulations which could potentially increase the risk of massive bleeding.

AUF was traditionally treated surgically by separating the fistulous connection between the artery and ureter, by ligating the artery and inserting an extra-anatomic bypass, sewing directly over the fistula, or placing a vein patch and interposition graft.⁴ In the past few years, the management of AUFs has transitioned from open surgical repair to minimally invasive endovascular treatment.² This is most likely due to increased awareness of this condition, improved imaging techniques, faster treatment response times, and the minimally invasive nature of endovascular procedures.⁸ In our discussion of management options, two primary approaches emerge: the placement of a covered stent and the embolization of the culprit vessel. In our particular case, we opted to sacrifice the internal iliac artery, rather than pursuing the placement of a covered stent. This decision was grounded in the goal of achieving complete occlusion of the fistula and preemptively averting any potential recurrence stemming from further degradation of the arterial wall.



Fig. 5. Follow-up nephrostogram confirms total occlusion of the fistula and stable position of the coils (Arrows) within the right internal iliac artery.

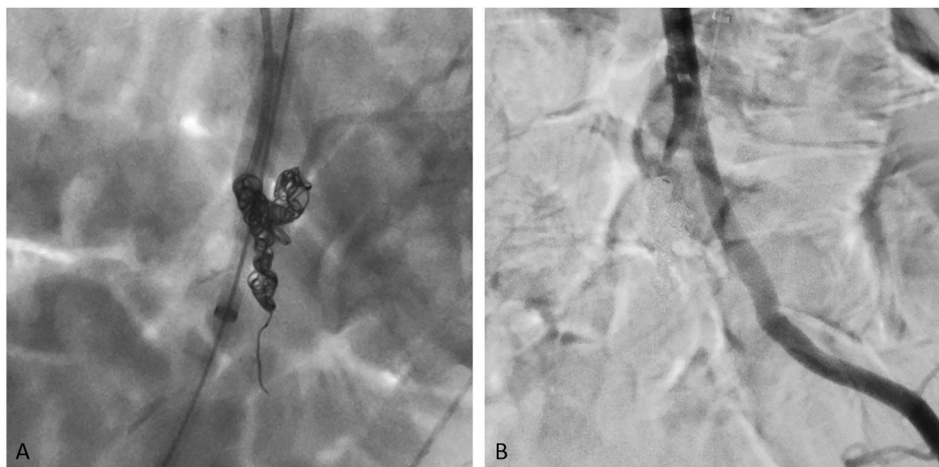


Fig. 4. (A) Fluoroscopic image shows coils deployed within the main right internal iliac artery. (B) Angiogram of the right common iliac artery showing total occlusion of the internal iliac artery.

Moreover, placing a covered stent would necessitate initiating the patient on a regimen of long-term anticoagulation, to reduce the risk of stent thrombosis and the potential development of distal emboli; events that could precipitate ischemic complications.

Sacrificing the main internal iliac artery secured the closure of the fistula while concurrently fostering the development of collateral circulation from both the ipsilateral external iliac artery and the contralateral internal iliac artery branches and preserving the flow to distal pelvic structures.

7. Learning points

- Clinical suspicion of arterio-ureteral fistula (AUF) should not be dismissed, especially in high-risk patients with pelvic malignancy.
- Antegrade nephrostogram is a useful diagnostic tool to confirm the diagnosis of AUF in cases of negative CT and angiogram.
- In addition to considering the fistula's location, it is crucial to account for ongoing tissue radionecrosis and breakdown when devising the management approach in cancer patients. This helps prevent the potential recurrence of the fistula in the future.

Ethics approval

Our institution does not require ethical approval for reporting individual cases or case series.

8. Informed consent

Informed consent for patient information to be published in this article was not obtained because the patient passed away and the next of

kin is not reachable for consent. The patient information is fully anonymized.

CRediT authorship contribution statement

Ahmed Awad: Conceptualization, Data curation, Investigation, Validation, Writing – original draft, Writing – review & editing. **Bader Abou Shaar:** Investigation, Writing – original draft, Writing – review & editing. **Rodrck c. Zvavanjanja:** Supervision, Validation, Writing – review & editing.

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