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Case Report

Endovascular treatment of arterioureteral fistulae involving both external and internal iliac arteries; Report of a rare case

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Abstract: Arterioureteral fistula (AUF) is a rare condition that may lead to lethal hematuria if not treated promptly. A 65-year-old female presented at our emergency department with massive hematuria and associated hypotension. She had a history of cervical cancer treated with radical hysterectomy followed by adjuvant chemotherapy and radiotherapy. Although computed tomography scan findings and renal angiography showed no signs of active bleeding, the symptom of hematuria aggravated. Even after prophylactic embolization of both renal arteries, hematuria persisted. On cystoscopy, there was a jet flow of bleeding from the right orifice of the ureter. AUF was highly suspected and subsequent intra-operative angiography was performed. Fistulous connections were observed and managed with endovascular repair. We report a rare case of AUFs showing connections with both the right external and internal iliac arteries.

Keywords: Arterioureteral fistula, covered stent, embolization, hematuria

Introduction

Arterioureteral fistula (AUF) is a rare but lethal condition if not treated promptly. Because the only symptom is hematuria in most of the patients, diagnosis of the disease can be delayed or missed. If hemodynamic instability occurs due to massive hemorrhage, mortality rates reach 23% [1]. Risk factors include pelvic surgery, radiation therapy, peripheral vascular disease, and chronic ureteral stenting [2]. We present a case of a patient with arterial-ureteral fistulae involving both the external and internal iliac arteries with a literature review. To the best of our knowledge, there are no previous reports of AUFs involving both the unilateral external and internal iliac arteries.

Case report

A 65-year-old female was transferred to our emergency department from another institution for management of massive hematuria and associated hypotension. She had been diagnosed with cervical cancer 15 years ago and had received radical total abdominal hysterectomy with multiple cycles of adjuvant chemotherapy and radiation therapy. One year prior to the visit, she had an episode of acute kidney impairment on both sides due to bilateral hydronephrosis. Hydronephrosis had been managed with ureteral double J stent insertion and the last exchange had been done approximately 2 months previously. She was admitted to another institution for evaluation of gross hematuria which developed 2 weeks before admission to our hospital. On computed tomography (CT), hematomas around the right ureter and in the right pelvis were detected. Hemoglobin (Hb) levels dropped to 5 g/dl and she received eight units of red blood cells (RBCs). The right double J stent was then removed and a right percutaneous nephrostomy (PCN) was performed. Renal angiography was performed but there were no signs of extravasation. Despite these measures, gross hematuria persisted with associated hypotension.

Imaging studies were performed again at our institution. Findings on the abdominal CT scan

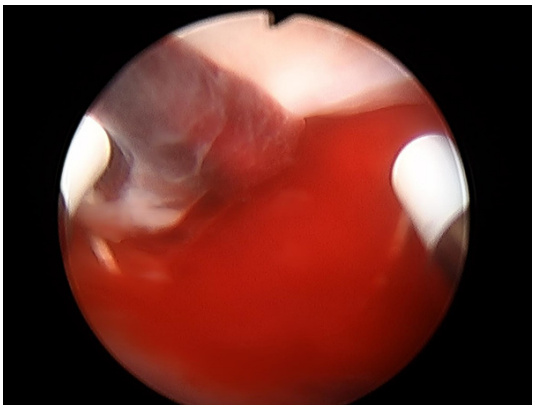

Figure 1. Preoperative cystoscopy showed massive bleeding into the urinary bladder from the right ureter.

showed no evidence of active bleeding. Renal angiography was performed to identify the cause of hematuria, but, no evidence of extravasation was found. Prophylactic embolization was performed with polyvinyl alcohol particles for the left renal artery and with Gelfoam for the right renal artery. But symptoms aggravated, and there was massive hematuria and a decrease in hemoglobin levels to 2.3 g/dl. The SBP was barely maintained, ranging from approximately 50 to 70 mmHg, even with the administration of norepinephrine and vasopressin.

On cystoscopy, a pinkish jet flow at the right ureteric orifice was noted. Due to massive bleeding, the urinary bladder was filled with a large amount of blood (**Figure 1**). Right nephrectomy was performed immediately, however, severe hypotension with massive hematuria persisted. Additional dopamine, dobutamine, and epinephrine were infused with simultaneous transfusion of packed RBCs, fresh frozen plasma, and platelet concentrates. In order to rule out arterial bleeding, the vascular surgeon gently dissected and temporarily clamped the right common iliac artery. The amount of hematuria reduced and the blood pressure gradually increased. A 5 F introducer sheath was inserted through the right common femoral artery under ultrasound guidance. Angiogram showed a suspicious fistulous connection of the right internal iliac artery with the right ureter (**Figure 2A**). A catheter was inserted into the right internal iliac artery, followed by the insertion of five 8 mm-Nester coils. After coil embolization, the amount of bleeding decreased. However, another fistulous tract was found between the right external iliac artery and the right ureter (**Figure 2B**). A 10 × 50 mm Viabahn covered stent (W.L. Gore, Flagstaff, AZ, USA) was inserted into the fistulous tract. Balloon angioplasty was performed with a 9 × 40 mm balloon catheter (**Figure 2C**). After successful placement of the covered stent and balloon angioplasty, the angiogram showed no bleeding through the fistulous tract (**Figure 2D**). After successful control of the AUFs, bleeding control in the operation field was achieved.

Postoperatively, the patient was closely observed in the intensive care unit with application of continuous renal replacement therapy and blood transfusion. Vital signs became stable and the laboratory findings improved. The patient recovered and was sent to the general ward.

Discussion

AUF is defined as an abnormal communication between a major artery and the ureter. AUF is classified into the primary type and secondary type depending on the cause of the fistula. Primary type (15%) is caused by anomalies of the arterial system, such as aneurysms, vessel malformation, or aberrant vessels that enter into the ureter. Secondary type (85%) is much more common and is caused by prior pelvic interventions [3]. The known common risk factors for the secondary type of fistula are a previous history of pelvic surgery (64%), combined pelvic radiation (49%), and ureteral stent placement (69%) [4]. There are three reported cases of pregnancy-associated AUF where patients had severe urinary tract infection with septic complications [3]. The most commonly involved arteries were the common iliac artery (57.1%), the external iliac artery (21.4%), the internal iliac artery (8.3%), graft/anastomosis site (10.7%), and the inferior mesenteric artery (1.2%) [4]. To our knowledge, AUFs involving both external and internal iliac arteries simultaneously are very rare, and we could not find the report of this kind.

Considering the rarity of AUF, it is difficult to suspect this condition; thus making it difficult to establish the diagnosis. Hematuria is usually the only symptom with the severity varying from minimal to life-threatening. Some patients present with accompanying flank pain or other

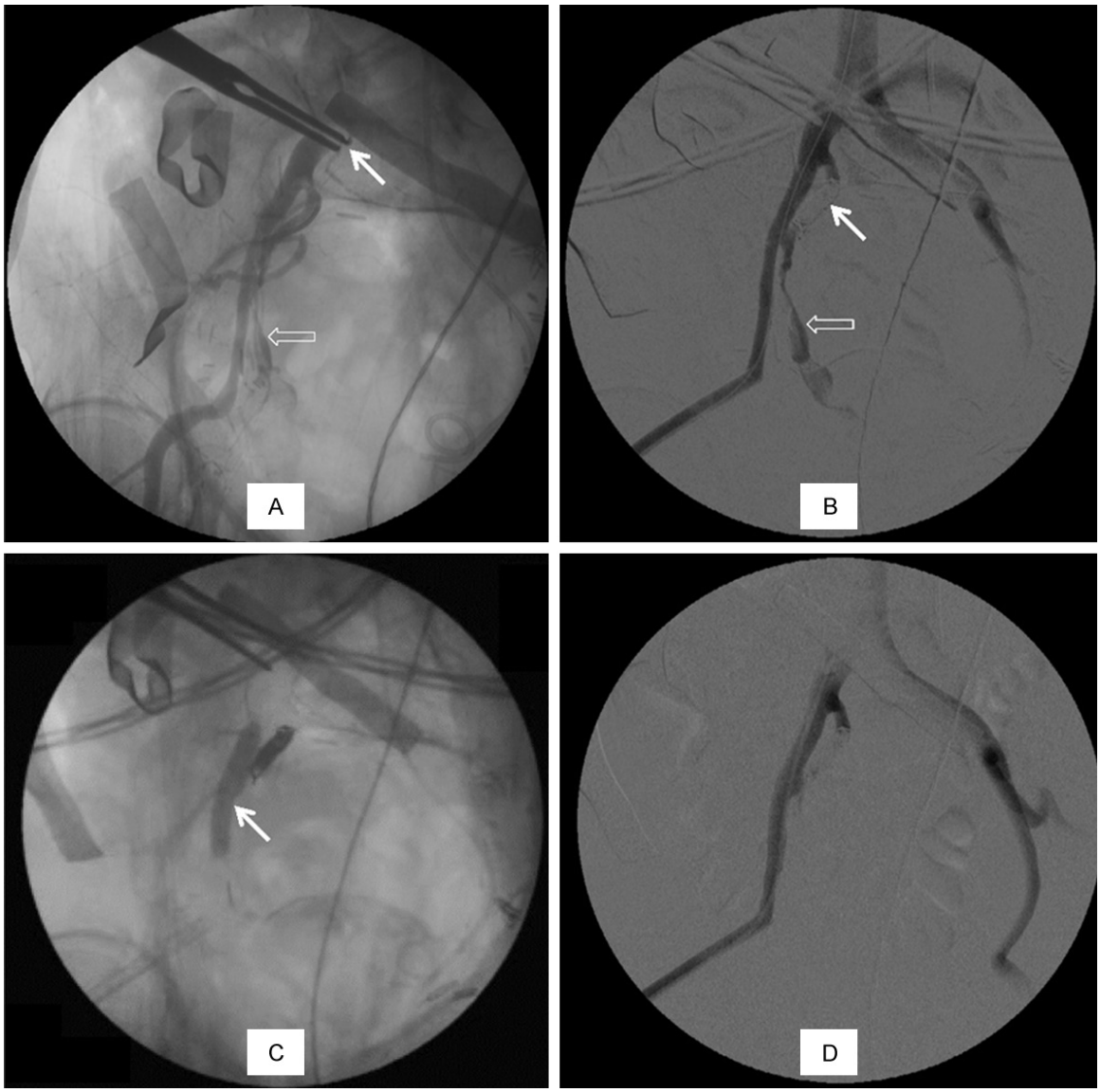

Figure 2. A: With clamping of the right common iliac artery (arrow), the initial angiogram showed contrast filling in the right ureter from the right iliac artery (open arrow). B: After coil embolization of the right internal iliac artery (arrow), the angiogram showed persistent contrast filling in the ureter from the right external iliac artery (open arrow). C: Ballooning was performed after placement of a 10 × 50 mm Viabahn covered stent in the right external iliac artery (arrow). D: Completion angiogram showed no contrast leak.

signs of infection. Most patients with hematuria undergo cystoscopy, but cystoscopy had a diagnostic value in only 4% of patients in a previous study [1]. CT scan can only be helpful in 7 to 9% of cases [1, 4]. Angiography is the best modality to diagnose AUF. Approximately 69% of 78 cases were diagnosed by angiography. In 20 to 26% of cases, the diagnosis of AUF was considered only after open surgery had been performed, as in our case [1, 4].

The reported mortality for AUF varies from 7.1% to 23%. But if the diagnosis is delayed, mortality rises up to 58% [5, 6]. Treatment of AUF is performed by open surgical repair of the fistula or endovascular treatment. Open repair can be challenging due to a previous history of pelvic surgery, radiation or hemodynamic instability. With the development of covered stents, endovascular approach has become the treatment of choice for AUF [7, 8]. Coil embolization with femoral crossover bypass was a common practice before the introduction of stent grafts [7, 8]. Coil embolization can still be an option for treatment of a fistula involving the internal iliac artery [7, 9]. Stent grafts can be less invasive,

while providing rapid control of hemorrhage. But, there is also a risk of infection in the fistulous communication between the ureter and the artery. Therefore, prophylactic antibiotics should be considered [8].

In conclusion, although AUF is a rare condition, it can be life-threatening and difficult to diagnose, so a high degree of suspicion for AUFs is important. When massive hematuria occurs in a patient with a history of pelvic surgery, radiation, or ureteral stent, the diagnosis of AUF should be considered. Angiography may be the diagnostic test of choice and endovascular stent graft coverage is the recommended treatment whenever possible.

Disclosure of conflict of interest

None.

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