CASE REPORT
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Life-threatening Arterioureteral Fistula Between Iliac Vessel and Ureteral Stump: A Case Report

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Abstract

Arterioureteral fistulas (AUFs) are a rare but life-threatening condition that cause massive hematuria. Radiation treatments, pelvic surgeries and chronic permanent ureteral stent use due to pelvic malignancies have been identified as the most important causes of AUFs. The number of cases in which arteriography and computed tomography angiography are insufficient and the pulsatile flow of the fistula can be observed only with ureterenoscopy is very rare in the literature. Here, we presented our 60-year-old patient with AUF, who has important risk factors such as pelvic surgery, radiotherapy and recurrent ureteral stent placement and whose pulsatile flow of the fistula was observed by ureteroscopy between the ureteral stump and iliac vessel

Keywords: Arterioureteral fistula, iliac vessel, ureteral stump

Introduction

Arterioureteral fistulas (AUFs) are a rare but life-threatening condition that cause massive hematuria. They usually occur where the ureters cross the iliac vessels (1). Radiation treatments, pelvic surgeries and chronic permanent ureteral stent use due to pelvic malignancies have been identified as the most important causes of secondary AUF (2) The most common symptom is intermittent hematuria, which can be life-threatening. AUFs are very difficult to diagnose. Failure to diagnose can be mortal. Although arteriography is defined as the gold standard diagnostic method (3), computed tomography (CT) angiography and antegrade/retrograde ureterography may also help in diagnosis despite their low detection rates. Treatment is mostly performed by endovascular interventions (4).

There are very few cases in the literature in which arteriography and CT angiography are insufficient, the pulsatile flow of the fistula can be observed only by ureterenoscopy. Here, we presented our 60-year-old patient with AUF who had significant risk factors such as pelvic surgery, radiotherapy (RT), recurrent ureteral stent placement, and pulsatile flow of the fistula observed by ureteroscopy.

Case Report

A 60-year-old female patient diagnosed with endometrial cancer 7 years ago applied to our clinic. It was learned that the patient had undergone total hysterectomy, bilateral salpingo-oophorectomy and pelvic lymph node dissection 7 years ago. She had received whole pelvic RT (57.8 Gy) after surgery. In the follow-up of the patient, bilateral hydroureteronephrosis (HUN) (right compensatory hypertrophic and grade 3-4 HUN, left partial atrophic grade 1-2 HUN) was detected due to RT. His creatinine (Cre) level was 4.6 mg/dL in July 2019. Bilateral 4.8 Fr Double J stents (DJS) were inserted. Her bilateral ureteral stents were changed regularly. Although the patient had bilateral DJS for 1 year, the patients became oliguric, the continuation of hydronephrosis and the development of Cre progression concluded that the drainage of ureteral stents were not sufficient, so right nephrostomy was deemed appropriate. So in July 2020, bilateral DJS was removed and only right nephrostomy was inserted. Five days after this procedure, the patient presented to the emergency department with intermittent macroscopic gross hematuria. CT angiography and digital subtraction angiography (DSA) was performed with the initial diagnosis of right renal AUF and pseudoaneurysm. No pathology was identified. The

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patient's hemoglobin (Hgb) was 7.8 g/dL. Her blood pressure was 95/62 mmHg, pulse rate was 101 beats/min, and her respiratory rate was 18/min. Blood clots were observed in the bladder and patient was hospitalized. Magnetic resonance imaging showed a 10X8 cm hematoma in the left perirenal, subcapsular area and clot in the collecting system (shown in Figure 1). As the patient's hematuria continued and Hgb value decreased to 5.6 g/dL, cystoscopy and left nephrectomy were performed under emergency conditions. On the cystoscopy, hematoma in the bladder and after washing, an appearance compatible with radiation cystitis was observed in the bladder. Retrograde pyelography was not applied to the patient, since the patient was operated under emergency conditions due to a significant decrease in Hgb and there was no hematuric urine flow from either ureteral orifice. As the hematuria persisted on the 2nd postoperative day, the patient underwent intravenous contrast-enhanced CT angiography with the preliminary diagnosis of right renal arteriovenous fistula, right ureteroiliac fistula and left ureteroiliac fistula. However, no findings compatible with any active extravasation or fistula were found. Then, right renal angiography and bilateral pelvic arteriography were performed, but no pathology was found to explain the current situation. It was decided to perform repeat cystoscopy and bilateral diagnostic ureterorenoscopy (URS). Cystoscopy was applied first, no active hematuria was observed. Then, the right URS was applied. Active hematuria or ureteral clots were not observed. Then, the left stump was entered through the left orifice. Pulsatile arterial flow was observed at the midureteral level (shown in Figure 2a). The arterial level associated with the ureteral fistula was determined by retrograde ureterography (shown in Figure 2b). The vascular plug was immediately placed at the level of attachment of the left common iliac artery to the ureter for embolization by interventional radiologists via an endovascular approach (shown in Figure 2c). The patient, who had no postoperative problem, was discharged on the 5th day after the procedure. The patient did not have hematuria or fistula recurrence for a year. The patient is currently being followed up with a right nephrostomy. There is no need for hemodialysis. The cre value is at the level of 3.4 mg/dL.

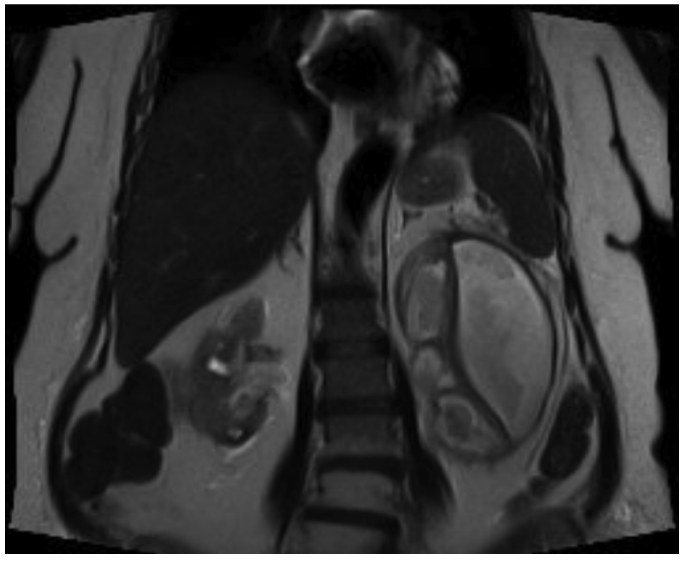

Figure 1. Left perirenal, subcapsular 10X8 cm hematoma and clot in the collecting system on magnetic resonance imaging

Discussion

AUFs are a rare but life-threatening condition that cause massive hematuria. They typically occur where the ureter crosses the iliac vessels, close to the iliac bifurcation (1). AUFs are more common in women (4). Fistulas on the left are more common than on the right (1). While these may be vascular aneurysms or pseudoaneurysm erosion primary fistula to the ureter (15%), they are mostly secondary fistulas (85%), and they appear after urological and gynecological cancers or after radiation (5).

While pelvic surgery and radiation therapy cause fixation of the ureter to the arteries due to ischemia, inflammation and fibrosis in the artery wall by damaging the vasavasorum, placing a ureteral stent creates a basis for fistula formation by increasing the pressure and causing abrasive erosion in the ureter wall and accelerating necrosis (6,7). Recently, an increase in the incidence of AUF has been observed in parallel with the prolongation of life expectancy, the widespread use of radiation treatments, pelvic surgeries and chronic permanent ureteral stents due to pelvic malignancies (2). As the radiation dose increases, the extent of arterial damage also increases, which means more frequent AUF (8). Considering that the average time between radiation therapy and the onset of AUF is 36 months, and the average time between ureteral stent placement and the onset of AUF is 18 months, it can be said that ureteral stent placement plays a greater role in the development of AUF (9). Ureteral stents act as support against AUFs. The use of large

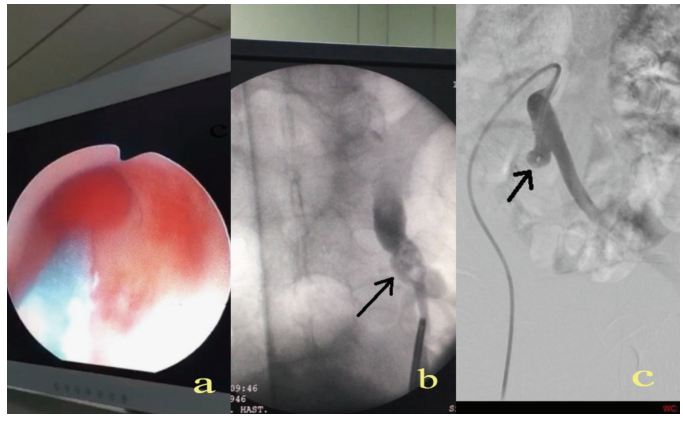

Figure 2.a. Pulsatile arterial flow at the midureteral level, **b.** Level of the artery associated with the ureteral fistula on retrograde ureterography, **c.** Vascular plug inserted through the endovascular approach at the level where the left common iliac artery connects to the ureter

diameter ureteral stents causes ischemia by putting pressure on the ureter wall. Considering that the 7F and 12F ureteral stents have the same flow rate, care should be taken to use the smallest diameter and softest material possible during ureteral catheterization in patients with risk factors (10). Our patient also had important risk factors for AUF, such as pelvic surgery, pelvic RT and recurrent urethral stent insertion.

Unfortunately, only 22% of the patients are diagnosed correctly before treatment (11). The most common symptom up to 74% is hematuria (1). This entity, which is generally encountered with massive hematuria, may result in death, unfortunately, as it is a rare condition among the causes of hematuria (11) and is diagnosed late and even often undiagnosed.

If hematuria occurs in patients undergoing pelvic surgery, radiation and ureteral stenting, AUF should be kept in mind and pelvic angiography should be performed when the diagnosis cannot be made with imaging methods. The probability of CT Urography showing positive findings in AUF is only 22% (1). Although contrast-enhanced CT can show the enhancing mass at the point where the ureter crosses the vessel wall in the presence of a pseudoaneurysm, bleeding usually occurs with a direct fistulous connection between the artery and the ureter. This situation makes it impossible to show AUF with cross-sectional imaging (6). Studies have shown that CT helps in diagnosis only in 42% of the cases (1). DSA remains the gold standard for the diagnosis of AUF. However, it can still show the accuracy of the diagnosis to be as low as 69% (1). For DSA to indicate an active fistula, there should typically be a flow rate faster than 3 mL/s. If it is considered that abnormal findings are not observed in approximately one-third of the patients with AUF even with DSA, angiographic examinations must be evaluated with different oblique projections. This is because small pseudoaneurysms can be overlooked because of overlapping the iliac arteries. Although the diagnostic value of angiography increases with provocative maneuvers such as manipulating, removing of ureteral stents or thrombolytic applications, it should be kept in mind that these maneuvers may lead to massive hematuria and emergency intervention may be required (7). As an alternative to arteriography, diagnosis can be made by antegrade or retrograde ureterography by direct contrast extravasation from the ureter to the arterial structures. In a study, the accuracy of this method in diagnosis was found to be 52% (1). Ureterography should be considered in patients in whom AUF is highly suspicious but angiograms cannot explain the cause. It should be remembered that the suspicion is important in these patients, as our patient could not be diagnosed with angiographic studies, but diagnosed by retrograde ureterography and ureteroscopy. A mean of 2.4 instrumental examinations for patients was required to achieve the correct diagnosis (1). Mortality due to AUF varies between 7% and 23% (3).

Although open surgery accompanied by vascular ligation and/or nephrectomy preserves its place as a form of treatment (2), less invasive endovascular interventions that require a multidisciplinary approach with the cooperation of urologists and interventional radiologists have been accepted as the main treatment for treating AUFs recently (3). Malgor et al. (12) found that patients treated with open repair had a higher rate of enterocutaneous fistula (60%) and rate of early complications (27%) than the endovascular treatment. Fox et al. (13) found no difference in hematuria recurrence in patients treated with open (33%) and endovascular repair (14%). In patients with a short life expectancy, urinary diversion with percutaneous nephrostomy may be an alternative in addition to ureteral occlusion. However, it should be kept in mind that this method may progress with recurrent bleeding without arterial occlusion or repair (14). Nevertheless, endovascular repair remains the treatment of choice in the appropriately selected patient, as many patients have been successfully treated with endovascular intervention and open surgical repair is uncomplicated. We also used the embolization method with the endovascular approach in our patient in accordance with the treatment method of our age. Following the right main femoral artery entrance, the left common iliac artery was selectively catheterized using a hydrophilic guide wire and a diagnostic catheter. The arterial branch associated with the left ureter stump was embolized with a vascular plug of appropriate diameter from its origin in a way not to block the main iliac artery flow.

Ethics

Informed Consent: Informed consent was obtained from the patient who participated in the study.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: E.K., A.E.Ç., Concept: E.K., Design: Y.Ö.İ., Data Collection or Processing: E.K., Analysis or Interpretation: Y.Ö.İ., Literature Search: M.H.Ö., A.E.Ç., Writing: M.H.Ö., A.E.Ç.

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