

Case Report

Ureteroarterial Fistula Presenting as Gross Hematuria during Routine Nephroureteral Stent Exchange

Abstract

Ureteroarterial fistula (UAF) is an uncommon entity and underrecognized etiology of hematuria in patients with a history of pelvic malignancy, irradiation, and surgery. Herein we report two cases of UAF unexpectedly identified during routine nephroureteral stent exchanges which highlight salient points regarding diagnosis and management.

Keywords: Arterioureteral fistula, ureteral stent, ureteroarterial fistula

Introduction

Ureteroarterial fistula (UAF) is an uncommon entity whose incidence has been rising due to the increased use of chronic ureteral stents in patients with a history of pelvic malignancy, irradiation, and surgery. Despite this, it remains an underrecognized etiology of hematuria which often leads to delay in diagnosis and contributes to the mortality rate of 7%–23%.^[1] We report two cases of UAF unexpectedly identified during nephroureteral stent exchanges which highlight salient points regarding diagnosis and management. Institutional review board approval was obtained for this case report.

Case Reports

Case 1

A 75-year-old female with a left-sided ureteroileal anastomotic stricture managed with 14-Fr transstomal nephroureteral stents (UreSil, LLC, Skokie, IL, USA) for the past year presented for an exchange. Medical history was notable for stage II urothelial carcinoma of the bladder and urethra treated with neoadjuvant chemoradiation and cystourethrectomy with ileal conduit urinary diversion 4 years earlier. The patient had been hospitalized 4 days prior due to intermittent hemorrhage via the urostomy requiring transfusion. Initial workup including platelet count, coagulation panel, and computed tomography angiography (CTA) were unremarkable.

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During the exchange, massive hemorrhage via the urostomy with hypotension and tachycardia occurred following stent removal. A 14-Fr × 40 cm multi-side-hole biliary drain (UreSil, LLC, Skokie, IL, USA) containing its inner stylet was emergently placed to achieve hemostasis. A uretero-common iliac artery fistula was suspected given the apposition of these structures on prior CT. Pelvic angiography with the drain in place was unremarkable. Retrograde pyelogram following stylet removal demonstrated opacification of the left common iliac artery confirming a fistula [Figure 1a]. Emergent endovascular fistula exclusion with a 10 mm × 40 mm iCast Balloon-Expandable Covered Stent (Atrium, Hudson, NH) was performed. Repeat pelvic angiography and retrograde pyelography confirmed successful exclusion and stent patency [Figure 1b and c]. There was complete postprocedural resolution of hemorrhage. Given the concern for developing a future stent graft infection, the patient underwent an open left ureteronephrectomy and left common iliac artery reconstruction 1 week later. The postoperative course was complicated by graft disruption and infections ultimately resulting in the patient's demise 3 months later.

Case 2

A 38-year-old male with bilateral extrinsic obstructive uropathy secondary to a presacral mass presented for exchange of a partially dislodged right percutaneous nephrostomy. Medical history was notable

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Figure 1: (a) Retrograde pyelogram through the left multi-side-hole drain (arrows) demonstrates opacification of the left common iliac artery (arrowheads) confirming the diagnosis of ureteroarterial fistula. (b and c) Repeat retrograde pyelogram and angiogram following stent deployment (arrows) confirms fistula exclusion and stent patency

for stage IV colorectal adenocarcinoma treated with chemoradiation and pelvic exenteration 18 months earlier. The patient had experienced three episodes of hemorrhagic output from the right nephrostomy in the 3 weeks prior to presentation which were presumably due to catheter dislodgment. On presentation, the patient was tachycardic with a hemoglobin level of 3.4 g/dL and required transfusion. Platelet count and coagulation panel were unremarkable.

During the exchange, the guidewire traversed distal to the location of the ureteric obstruction. Gross pulsatile hemorrhage was appreciated from the 5-Fr Kumpe catheter (Cook Medical, Bloomington, IN, USA). Hemodynamic parameters remained stable. Contrast injection demonstrated opacification of the right external iliac artery consistent with a fistula [Figure 2a]. Intraprocedural review of prior cross-sectional imaging revealed a pseudoaneurysm of the right common iliac artery abutting the ipsilateral ureter which was likely due to tumor erosion. The Kumpe catheter was left *in situ* to achieve temporary hemostasis. The pseudoaneurysm was visualized during pelvic angiography and excluded with a 10 mm × 58 mm Lifestream Balloon-Expandable Vascular Covered Stent (BD Bard Peripheral Vascular, Tempe, AZ, USA) [Figure 2b]. Repeat pelvic angiography and antegrade pyelogram confirmed successful fistula exclusion [Figure 2c]. No further episodes of hemorrhage were noted until the patient's expiration 1 month later due to disease progression.

Discussion

The presented cases highlight several important aspects regarding UAF. Risk factors for the development of UAF include prior surgery and irradiation for pelvic malignancy and chronic ureteral stent placement. Intermittent hematuria is the most common presenting symptom.^[1] The degree of hematuria ranges from microscopic to massive hemorrhage requiring resuscitation.^[2] Fistulization is typically unilateral and involves the common or external iliac artery. Diagnosis

remains problematic as UAF is seldom considered as a cause of hematuria. CTA is rarely diagnostic, but can identify alternative etiologies of hematuria (neoplastic invasion, radiation necrosis, nephroureterolithiasis, and stent-related injury) or findings suggestive of UAF (pseudoaneurysm, collecting system thrombus, and ureter-iliac artery apposition). Angiography and pyelography are the most sensitive studies.^[2] Provocative maneuvers with endoureteral balloon or catheter-directed mechanical friction has been reported to increase diagnostic yield via disruption of tamponading thrombus.^[2]

Management of UAF is controversial but has evolved to predominantly involve endovascular repair due to the high operative risk associated with open surgery in a hostile abdomen. Robust data regarding long-term outcomes for endovascular repair are lacking though existing evidence suggests that it is noninferior to surgical repair.^[3] Stent-graft infection and reintervention for recurrent hematuria after endovascular treatment are rare but serious complications.^[3,4] The role of long-term prophylactic antibiotics is unclear, but their use may be prudent given the morbidity associated with infection.

Conclusion

The clinical suspicion for UAF should be heightened when encountering significant hematuria during nephroureteral stent exchange in patients with a history of irradiation and surgery for pelvic malignancy and chronic ureteral stent placement. Endovascular management is a viable option and may be comparable to open surgical repair. Long-term antibiotics use may be prudent, given the morbidity associated with stent infection.

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Conflicts of interest

There are no conflicts of interest.

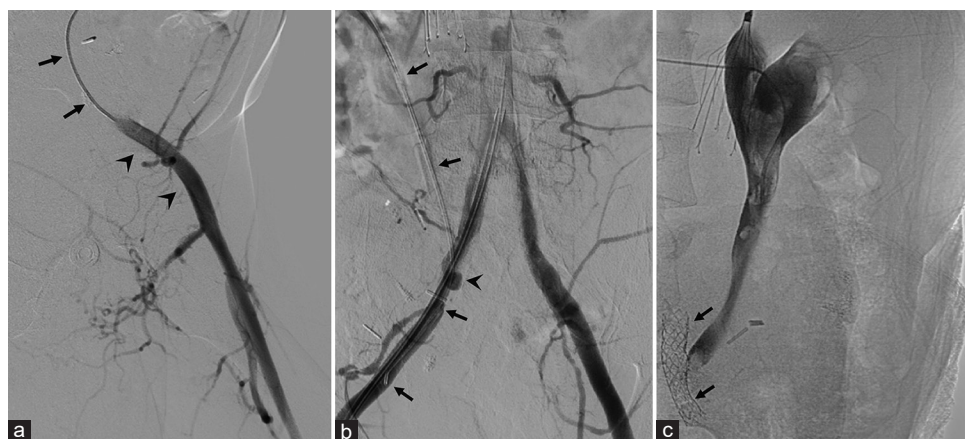


Figure 2: (a) Prone antegrade pyelogram through the Kume catheter (arrows) demonstrates opacification of the right external iliac artery compatible with a ureteroarterial fistula (arrowheads). (b) Supine pelvic angiography shows the right common iliac artery pseudoaneurysm (arrowhead) at the tip of the Kume catheter (arrows) in the ureteroarterial fistula. (c) Prone antegrade pyelogram demonstrates successful ureteroarterial fistula exclusion following stent deployment (arrow)

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