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A Case of Uretero-Arterial Fistula

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Abstract

Initially recognized as a rare condition, uretero-arterial fistulas are increasing in incidence. In this report, we present a case of uretero-arterial fistula arising from chronic ureteral stenting followed by surgical repair. Patient presentation, clinical considerations, treatment approach, and complications unique to our case are discussed.

Keywords: Angiogram, fistula, stent, uretero-arterial fistula, vascular

Introduction

The uretero-arterial fistula is a rare entity thought to be increasing in incidence over recent years with an aging population and increased life expectancy. Since it was originally described in 1908 by Moschcowitz, approximately 150 cases have been described, with a majority of cases described after 1970.[1] Most cases involve patients with a previous history of cancer with multimodal pelvic cancer treatments and chronic ureteral stenting. Management options include both endovascular and open surgery.

Case Presentation

A 71-year-old female with a history of uterine cancer status post radical hysterectomy and radiation therapy developed radiation cystitis with bilateral hydronephrosis requiring chronic bilateral internal double J ureteral stents which were exchanged multiple times over the past 5 years. She presented to the hospital with gross hematuria. Ureteral stents were removed with the placement of nephrostomy tubes at an outside hospital before her presentation. Her hemoglobin on presentation measured 5.4 g/dL, and 4 units of packed RBCs were administered. Urine culture was positive for vancomycin-resistant enterococcus. Based on initial concerns of hemorrhagic cystitis, a cystoscopy was performed but did not demonstrate active bleeding from the bladder. However, bloody efflux was noted from the right ureteral opening. Bilateral nephrostomy tubes were placed for urinary diversion.

Two days later, the patient experienced another episode of gross hematuria. CT angiogram (CTA) showed a uretero-arterial fistula between the right common iliac artery and the right ureter (Figure 1), and interventional radiology was consulted. An aortoiliac angiogram was performed via right femoral artery access using a multi-side hole catheter, which demonstrated the subtle arterial ureteral fistula arising from the right common iliac artery (Figure 2A). The fistula was then excluded through the deployment of 2 overlapping 10 mm x 38 mm Atrium iCast stent grafts (Figure 2B). While the first stent graft excluded the arterial ureteral fistula, a decision was made to place an overlapping second stent graft given the presence of an endoleak which could have resulted in the persistence of the fistula (Figure 2C and D).

Given a history of chronic urinary tract infections and percutaneous nephrostomy tubes, there was a concern for infection of her iliac artery stents. The patient decided to proceed with surgical repair of the fistula with urinary diversion and chose to forego major urological reconstruction, opting to have right ureter ligation with continued right-sided nephrostomy tube.

The surgical repair involved extensive lysis of adhesions in the pelvic region, which was complicated by enterotomies resulting in small bowel resection. The common iliac artery was dissected down to the area where the ureter appeared to be plastered. The ureter was ligated above this level with a 2-0 silk, and a metallic clip was applied. The ureter was adherent over the iliac vessels. The ureteral segment distal to this adherent area was also transected and over-sewn with a silk suture. The remnant ureter over the vessel was sharply dissected off, and the artery was opened with resection back to healthy margins. The previously placed iliac artery stents were removed. The right-sided great saphenous vein was then used for patch

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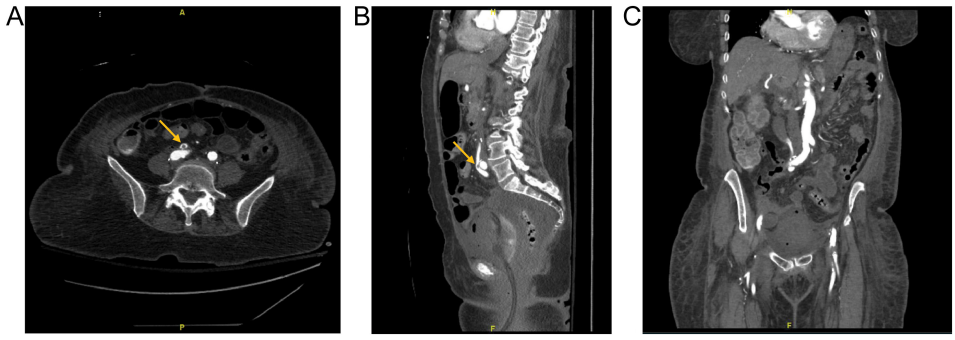

Figure 1. Right ureteral-iliac arterial fistula demonstrates a thin communication between the right iliac artery and mid ureter (yellow), where the thrombus is seen as a filling defect in the right mid ureter. (A) Axial, (B) sagittal, and (C) coronal views.

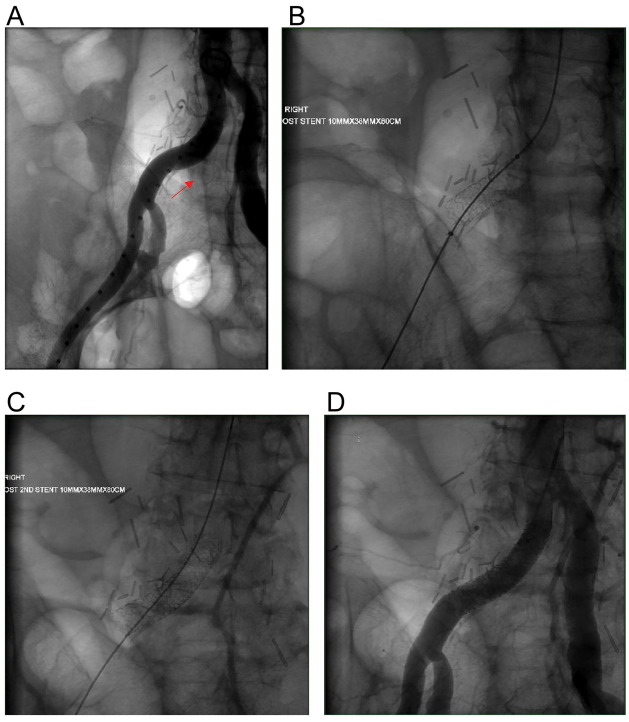

Figure 2. (A) Aortoiliac angiogram showing subtle uretero-arterial fistula (red arrow). (B) After deployment of the first stent. (C) After deployment of the second stent to exclude endoleak. (D) Final arterioiliac angiogram with the exclusion of uretero-arterial fistula.

angioplasty. The abdomen was then closed in a standard fashion. No drains were left behind.

Postoperatively, the patient was admitted to the intensive care unit. Vasopressor support was weaned off, and she was extubated on postoperative day 1. Due to poor oral intake and severe deconditioning, Dobhoff placement with tube feeds was initiated. She had a worsening oliguric acute kidney injury but responded appropriately to the furosemide stress test with adequate urine output and improved renal function. She did not require hemodialysis and was discharged to rehab on postoperative day 10.

The patient presented 2 months later to the emergency department with abdominal pain, nausea, and poor intake. Computed tomography (CT) of the abdomen and pelvis without contrast showed no evidence of bowel obstruction but fluid collection in the retroperitoneum near the surgical site, possibly a seroma or hematoma (Figure 3). There was another fluid collection near the femoral vein likely a seroma/abscess at the saphenous vein harvest site. Bowel anastomosis and Iliac artery patch angioplasty sites seemed intact. Shortly afterward, the patient had an episode of bilious vomiting and aspiration, which led to cardiac arrest with pulse less electrical activity (PEA) arrest. Unfortunately, a return of spontaneous circulation was not achieved, and the patient passed away.

Discussion

The uretero-arterial fistula is a rare entity. It was initially described by Moschcowitz in 1908 in a 36-year-old patient post bilateral ureterolithotomy. It was a unique case where bilateral uretero-arterial fistulas were thought to have resulted between the ureters and external iliac vessels secondary to ureteral drain pressure injury. The patient underwent bilateral ligation of the external iliac arteries and proceeded to have a favorable outcome.[1]

Most of the patients described in the literature with uretero-arterial fistulas have a history of pelvic cancers, multimodal pelvic treatments for cancer, chronic ureteral stents, and a previous history of vascular surgery.[2,3] Uretero-arterial fistulas are classified as primary (15%) or secondary (85%).[3,4] Primary uretero-arterial fistulas are native while secondary uretero-arterial fistulas can form after surgery, vascular graft placement, irradiation, retroperitoneal fibrosis, or ureteral stenting. Anatomically, the fistula can exist between the native ureter and common, external, or internal iliac arteries. Fistulization can also occur with the aorta in cases involving urinary conduits.[5]

The pathophysiology of secondary uretero-arterial fistulas is complex and appears to involve a combination of inflammation, ischemia, and fibrosis.[3,6] Ultimately, the uretero-arterial fistula forms most often where a ureter crosses an artery (usually the common or external iliac) or anastomosis, and an associated pseudoaneurysm is reported in 38% of cases.[3,7]

Patients most commonly present with hematuria, which can be microscopic or macroscopic and range from intermittent to massive, often declaring itself at ureteral stent exchange/removal.[3,7] Diagnosis of uretero-arterial fistulas can be difficult, and the clinician must have a high degree of suspicion. Contrast-enhanced CT was reported to be helpful in 42% of cases[7,8] and can show an enhancing pseudoaneurysm or

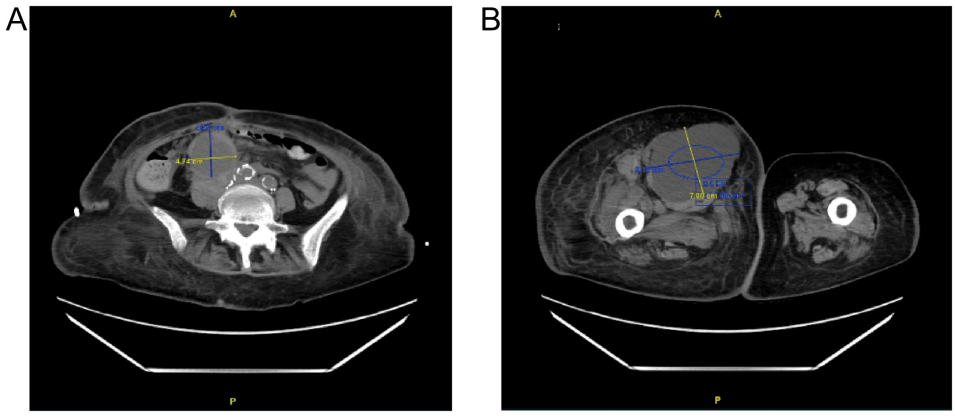

Figure 3. (A) Right retroperitoneal fluid collection lateral to the right proximal common iliac artery. (B) Right thigh fluid collection abutting the right femoral artery and vein.

hydronephrosis with ureteral clot; however, the uretero-arterial fistula will not be readily apparent. If there is a high degree of suspicion and a CT scan is negative, cystoscopy should be considered, which will show bleeding from the ureteral orifice. Nevertheless, contrast-enhanced CT is important for endovascular and surgical planning for localizing where the ureter crosses the arterial structures. Provocative angiography has been described in difficult cases but involves an elevated risk of uncontrolled bleeding.[9] It involves a combination of arteriography and ureteral stent manipulation.

A retrospective chart review from 1975 to 2004 at the Mayo Clinic revealed 8 uretero-arterial fistulas in 7 patients, all of whom presented with hematuria with risk factors including chronic indwelling stents, previous pelvic external beam radiotherapy, and pelvic surgery.[10] Provocative angiography was diagnostic in 63% of the cases.[10] When performed, cystoscopy revealed lateralized, pulsatile hematuria in all cases.[10] Prior to the development of the indwelling ureteral stent in 1978, few uretero-arterial fistulas were reported, supporting the role that these stents play in the development of uretero-arterial fistulas.[11] In particular, larger-diameter stents tend to apply greater radial pressure, leading to ischemic changes in the ureteral wall that could predispose the individual to uretero-arterial fistulas.[10] In the study, the authors presented a treatment algorithm recommending that (1) poor surgical candidates undergo endovascular stent with nephrostomy tube, (2) marginal surgical candidates undergo iliac occlusion ± arterial bypass with ureteral stent or nephrostomy tube, and (3) surgical candidates undergo primary surgical repair of the artery with nephrectomy and ureteral ligation or nephroureterectomy in patients with normal renal function.[10] Nephrostomy tubes in patients with marginal renal function can be attempted combined with arterial repair to preserve remaining renal function.[10]

A more recent report[12] in 2017 discusses a 51-year-old female with a history of cervical cancer post hysterectomy and radiation who developed bilateral ureteral strictures that were treated with chronic indwelling stents. A right uretero-arterial fistula subsequently arose, which was managed with an endovascular stent to the external iliac artery. Two years later, the patient presented with hematuria and hematochezia secondary to a uretero-arterial-enteric fistula, which was successfully treated with the placement of nephrostomy tubes.

Unfortunately, no clear society guidelines exist currently for diagnostic protocols and management of uretero-arterial fistulas. Treatment options remain controversial with several articles and reviews favoring endovascular management.[9,13] However, failure of endovascular management with late complications including recurrent bleeding, stent-graft infection, and pelvic abscess are described that require open surgery.[14]

Open surgical repair has been attempted in cases of local pelvic sepsis, iliac artery aneurysm, occluded femoral vessels, and failure of endovascular intervention with stent-graft infection or recurrent bleeding.[14] Surgical intervention is associated with high morbidity and mortality, as most cases of uretero-arterial fistulas have a previous history of pelvic irradiation, open surgical intervention, and concurrent inflammatory changes. Causes of morbidity reported after the open repair include recurrent hemorrhage, enterocutaneous fistula, bowel perforation, sepsis, and bowel obstruction.[13,14] Options for surgical intervention include ureteral repair, shunts or nephroureteral resection along with fistula take down with primary arterial repair, patch angioplasty with vein patch, ligation of the iliac artery with extraanatomical bypasses including iliofemoral or femoral-femoral bypass, and arterial ligation of an internal iliac artery is involved.[13,15]

Endovascular treatment is now commonly preferred as a bridge to surgery or used alone as definite therapy.[9] Management involves stent-graft placement for occlusion of uretero-arterial fistulas. Coil embolization has also been attempted for the management of uretero-arterial fistulas, especially if the internal iliac artery is involved.[13,15] Complications with endovascular intervention include recurrent hemorrhage, stent-graft infection, lower extremity morbidity, and rarely stent-graft thrombosis.[9,13]

Conclusion

Uretero-arterial fistula is a rare entity occurring most commonly in patients with previous pelvic or vascular surgeries, cancer, or irradiation. Management remains controversial and is dependent on a case-by-case basis. Endovascular intervention is favored in most cases as definitive therapy or as a bridge to open surgery for controlling life-threatening hemorrhage. Open surgical intervention is associated with significant morbidity, as most cases present a hostile surgical field with previous irradiation, surgery, and local sepsis. However, open

intervention is indicated in cases of local sepsis or failure of endovascular intervention.

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