Endovascular Management of Ureteroarterial Fistula: A Rare Potentially Life Threatening Cause of Hematuria

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ABSTRACT

Ureteroarterial fistula is a rare, potentially life-threatening cause of hematuria characterized by an abnormal channel between a ureter and artery. The rarity of this condition, complexity of predisposing risk factors and intermittence of symptoms may delay or obscure its diagnosis. With a high index of suspicion and careful angiographic evaluation, embarking on this condition is not only possible but sets the stage for curative intervention. We report a case of a ureteroarterial fistula presenting with intermittent hematuria, successfully diagnosed at angiography and managed with endovascular stent graft placement.

CASE REPORT

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Clinical Presentation and Imaging

A 70-year-old debilitated female presents to the emergency department with a several day history of intermittent hematuria from her indwelling Foley catheter. Past medical history included extensive pelvic trauma, uterine cancer treated with surgery and radiation, colostomy for remote bowel perforation and indwelling ureteral stents for significant calculus disease burden. She was recently found to have a colovesicular fistula to her rectal stump. This was being managed non-operatively.

On initial evaluation, the patient was afebrile, mildy tachycardiac and dehydrated. Non-bloody stool was noted from her ostomy. Blood clots were identified within the Foley collection bag.

Her hemoglobin was low at 7.8g/dL (normal 12.1-15.0 g/dL) from her baseline ~11g/dL. WBC was elevated at 16.7 bil/L (normal 3.3-10.7 bil/L). Serum creatinine measured 5.10mg/dL (normal 0.60-1.40 mg/dL), significantly elevated from her baseline of ~1.6mg/dL (normal 0.60-1.40mg/dL). Urinalysis showed a large amount of blood with positive bacteria, leukocyte esterase and amorphous crystals.

Non-contrast CT of the abdomen and pelvis demonstrated marked bilateral hydronephrosis with multiple bilateral calculi and migration of the right ureteral stent such that its proximal loop was just distal to the ureteropelvic junction (Figure 1). The patient was admitted following fluid resuscitation, blood transfusion and initiation of antibiotics.

Cystoscopy performed the following day showed a blood clot within the colovesicular fistula localized to the posterior dome of the bladder. The fistula was fulgurated. Both ureteral stents where occluded by debris and subsequently removed. Bilateral percutaneous nephrostomy tubes were placed for urinary diversion and relief of hydronephrosis. Subsequent colonoscopy of the rectal stump and colon was unremarkable. No suspicious findings were noted on vaginal exam. Over the next week, her serum creatinine deceased but she continued to have blood tinged urine from the right nephrostomy tube. Following multiple transfusions, Vascular Interventional Radiology was consulted to aide in localizing the site of bleeding.

Delayed (24 hour) technetium-labed red blood cell scan demonstrated subtle radiotracer accumulation in the regions of the right kidney and urinary bladder (Figure 2). At this point, the decision was made to proceed with angiography.

Management

The patient was taken to the angiography suite the following day. While in the supine position, right common femoral artery access was obtained. Working through a 6F Pinnacle® sheath (Terumo®, Tokyo Japan), a non-selective aortoiliac angiogram performed via 5F Omniflush ® Pigtail catheter (Angiodynamics ® Latham New York) was unremarkable (Figure 3).

Attention was then turned to performing selective right internal iliac arteriogram. A C2 Glide ® catheter (Terumo ® Tokyo Japan) was noted to enter what was presumed to be the right internal iliac artery. However, when contrast was injected, it opacified an aperistalitic tubular structure in a nonvascular distribution (Figure 4a). Contrast extended into the urinary bladder, opacified the rectal pouch and spilled onto the angiography table from the patient's rectum (Figure 4b). It was concluded that the catheter had entered the ureter through a fistulous tract and that contrast flowed into her rectal pouch via the colovesicular fistula. A V18 microwire (Boston Scientific ® Natick Massachusetts) was then advanced through the C2 Catheter and coiled in the urinary bladder. The catheter was withdrawn slightly over the wire such that its distal tip was in the external iliac artery. Contrast injected over the wire opacified the right internal iliac artery and demonstrated its autonomy from the ureteroarterial fistula. The fistula site was localized to the distal right common iliac artery, just proximal to its bifurcation (Figure 5).

In anticipation of excluding the fistula with a covered stent, the right internal iliac artery was embolized with seven 6mm Nester ® coils (Cook Medical ® Bloomington Indiana) through a 2.8F Omega® coaxial microcatheter (Terumo® Tokyo Japan) (Figure 6). Post-embolization aortoiliac arteriogram performed through a 5F marker pigtail catheter (Angiodynamics ® Latham New York) demonstrated non-opacification of the right internal iliac artery (Figure 7). Then, a 9x 5mm Viahban ® covered stent (Gore ®, Neward Delaware) was deployed spanning the right common iliac artery and right external iliac artery excluding the origin of the embolized right internal iliac artery and UA fistula site (Figure 8).

Follow-up

Following endovascular therapy, the patient's hematuria ceased. Her hemoglobin stabilized to around 10g/dL (normal 12.1-15 g/dL) without need for any further transfusions. Serum creatinine trended down to her baseline over the following week. WBC count normalized upon completion of a course of Aztreonam. She was discharged from the hospital 1 week after endovascular intervention. The patient has since returned to Interventional Radiology for bimonthly percutaneous nephrostomy tube exchanges and denies any hematuria recurrence.

DISCUSSION

Etiology and Demographics

Ureteroarterial fistula is a potentially life-threatening cause of hematuria as the result of abnormal communication between an artery and ureter. The connecting channel typically involves the common or external iliac artery and the mid to distal ureter. Fistulas involving the internal iliac artery [1-2] and aorta [3] have also been reported.

This pathologic entity is uncommon with only around 90 cases being described in the literature [4]. The majority of cases have been described in women (86%) with an incidence higher in patients >50 year old [4-5]. Predisposing conditions include: the prolonged use of ureteral stents placed at the ureteroarterial crossing, radiation therapy in combination with surgery for genitourinary or gynecologic malignancy, presence of a ureteral stump after nephrectomy, vascular reconstructive surgery in the pelvis and ureterolithotomy complicated by urinary leak [4].

Clinical and Imaging Findings

The most common presenting symptom is recurrent hematuria [6] varying in amount from microscopic to frank bleeding. Thrombus occlusion of the fistula is felt to be responsible forepisodic nature of symptoms [1]. Flank pain, upper and lower urinary tract infections and hydrouteronephrosis may also be present. Laboratory evaluation is usually nonspecific and suggestive of infectious and or calculus etiology. Differentiation between myoglobinuria and hemoglobinuria must be made as both may lead to a false positive dipstick, and this can be confirmed with the absence of red blood cells on microscopic exam. Because of the intermittency and nonspecificity of symptoms, diagnosis can be elusive.

Diagnostic imaging, with ultrasound and CT, are considered low yield as close proximity of structures makes delineation of a discrete tract difficult. The utility of these noninvasive studies would be better served to rule out other conditions that may mimic similar presentation[6-7]. Theoretically, delayed phase excretory imaging (CT Urogram) may show evidence of abnormal communication between the two structures, however this has yet to be described. Cystoscopy may occasionally show pulsatile bleeding at one of the ureteral orifices. Extraction of a ureteral stent (if present), may provoke pulsatile bleeding during cystoscopy.

Though not very sensitive (<50%), iliac arteriography is the most specific diagnostic modality [7]. Angiography may occasionally demonstrate the extravasation of contrast material into the ureter. Provocative maneuvers such as stent removal over a guidewire, selective arterial injection using multiple oblique views, high-pressure balloon occlusion pyeloureterography, and manipulation of ureteral catheters (to dislodge an occluding thrombus) may be helpful if there is a high index of suspicion.

Treatment and Prognosis

Currently, there is no gold standard for the management of ureteroarterial fistulas. One study described success with coil

embolization of the ureterjust proximal to the fistula [3]. Other treatment options include surgical ligation, and intraoperative balloon occlusion followed by extraanatomic bypass of the common, external or internal iliac arteries [8-10].

Polytetrafluoroethylene-covered stent graft placement is a minimally invasive, endovascular procedure which can provide complete closure of the fistula while preserving antegrade arterial flow to the lower extremity and pelvis [9]. Self-expanding stents have demonstrated success [11]. Depending on the location of the fistula take off, the stent may require sacrifice of a distal iliac branch. In our patient, we coil embolized the internal iliac just distal to its take off to prevent a Type II endoleak via retrograde flow dissecting between the stent and common iliac intima.

Prior to 1980, the mortality rate associated with ureteroarterial fistulas was estimated to be 69% [5]. This was largely attributed to delay in diagnosis and treatment. Heighted clinical awareness of this condition with improved imagining and treatment techniques have reduced mortality rates to approximately 23%[5].

Differential Diagnosis

While the differential for intermittent hematuria is vast, several diagnoses are responsible for the majority of cases:

1. Cystitis, most commonly secondary to a urinary tract infection, is the most common cause [12].
2. Urethritis and prostatitis may present with intermittent hematuria as well as dysuria.
3. Nephrourolithiasis is the most common etiology arising from the upper urinary tract and is frequently associated with renal colic [12].
4. In young healthy individuals presenting with intermittent hematuria, recent strenuous exercise should be given consideration [13].
5. Patients with sickle cell disease may experience intermittent hematuria secondary to papillary necrosis.
6. With a known family history of kidney disease or cerebral aneurysms, polycystic kidney disease must be considered as a cause of intermittent hematuria [12].
7. IgA nephropathy is also consideration as it classically presenting with episodic hematuria beginning one to two days after a URI [14].
8. In addition to benign etiologies, malignancy may also be a cause. Patients over the age of 45 with gross intermittent hematuria have a genitourinary malignancy until proven otherwise. Bladder cancer in particular is known to cause intermittent hematuria [15].
9. Ureteroarterial Fistula as in this case should be considered when significant risk factors are present and the above stated conditions have been appropriately treated or excluded.
10. Other rare causes of intermittent hematuria include renal arteriovenous malformation, schistosomiasis, and endometriosis of the urinary tract [16]. Nutcracker syndrome, also known as Aorto-Mesenteric Left Renal Vein Entrapment Syndrome, has also been shown to cause intermittent hematuria [17].

TEACHING POINT

Ureteroarterial fistula is rare, potentially life-threatening condition and should be considered in the differential diagnosis in patients presenting with intermittent gross hematuria with a prior history of pelvic surgery/radiation, ureteral manipulation and chronic cannulation. Although currently no standard of care exists, endovascular fistula exclusion utilizing stent graft offers a reasonable treatment option.

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FIGURES

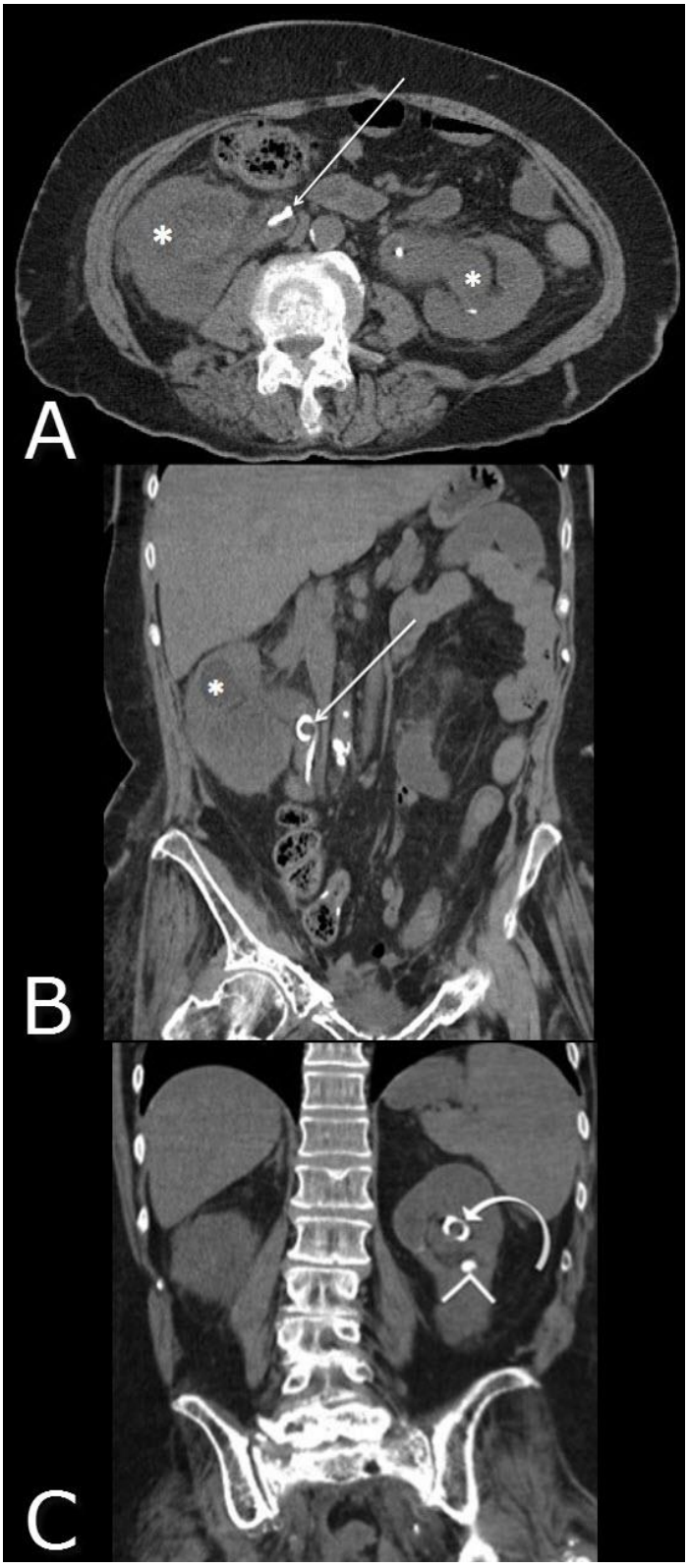

Figure 1 (right): Non contrast CT abdomen and pelvis at multiple levels in a 70-year-old female with a right ureteroarterial fistula. FINDINGS: Figure A) Axial Image demonstrates bilateral hydronephrosis (asterix) and displacement of the right ureteral stent (arrow). Figure B) Coronal image obtained at the level of the pubic symphysis demonstrates right hydronephrosis (asterix). The proximal loop of the indwelling ureteral stent is seen within the proximal right ureter, just distal to the ureteropelvic junction (arrow). Figure C) Coronal image at the level of the sacroiliac joint demonstrates the indwelling left-sided ureteral stent appropriately positioned with the left collecting system (curved arrow) and a calculus within the lower pole (arrowhead) reflecting this patient's known calculus disease burden. TECHNIQUE: 3mm coronal reformatted computed tomography images obtained on a Siemens® SOMATOM® 64 slice spiral CT scanner at 256mAS, 1.2 KV and 500 msec.

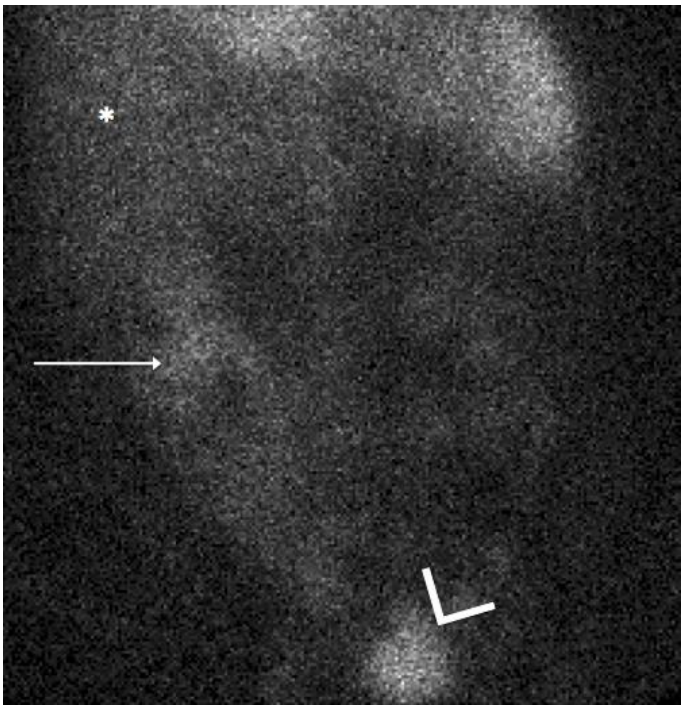

Figure 2: GI bleed scan in a 70 year old female with a right ureteroarterial fistula. FINDINGS: Delayed (24 hour) Tc 99m Tagged RBC scan demonstrates nonspecific radiotracer accumulation within the right upper quadrant (arrow) and in the region of the bladder (arrowhead). Note region of the Liver (asterix).TECHNIQUE: Tagged RBC scan images obtained in the PA projection 24 hours following injection of autologous RBC's tagged invitro with 21.7mCi of Tc 99m using an ULTRA-Tag Kit ®. Images obtained on a General Electric® Anger Camera.

Figure 3: Diagnostic pelvic angiogram in a 70-year-old female with a right ureteroarterial fistula. FINDINGS: Digital subtraction angiogram during aortoiliac arteriogram via 5F pigtail catheter within the distal abdominal aorta (right common femoral approach) demonstrates unremarkable opacification of the bilateral common iliac (straight arrows), internal iliac (arrowheads) and external iliac (curved arrows) arteries. TECHNIQUE: Diagnostic/therapeutic fluoroscopy in the PA projection (patient supine) Total contrast used: 70cc of Isovue 250, 75cc of Isovue 300. Total fluoroscopy time: 51minutes.

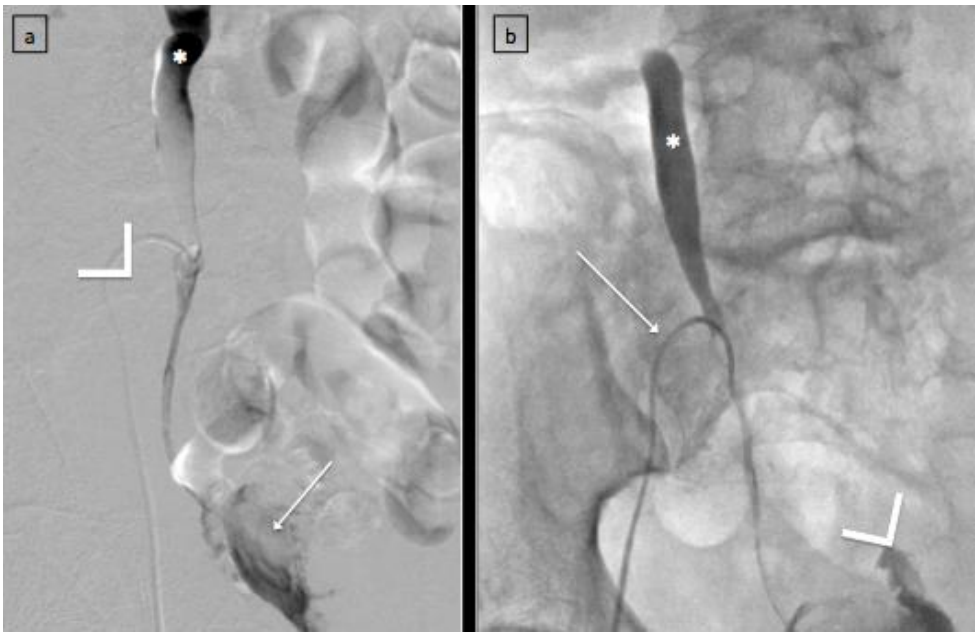

Figure 4: Diagnostic pelvic angiogram in a 70-year-old female with a right ureteroarterial fistula. FINDINGS: A) Digital subtraction angiogram obtained during presumed cannulation of the right internal iliac artery with a C2 catheter from a right common femoral artery approach (arrowhead). Following contrast injection, the catheter was noted to have entered a tubular structure (asterix). Contrast is seen flowing into the urinary bladder (arrow). B) Fluoroscopic spot image redemonstrates a C2 glide catheter (arrow) entering the ureter (asterix) via fistulous communication with an iliac artery. Contrast is seen filling the rectal pouch (arrowhead), presumably through the patients known colovesicular fistula. TECHNIQUE: Diagnostic/therapeutic flouroscopy. Total contrast used: 70cc of Isovue 250, 75cc of Isovue 300. Total fluoroscopy time: 51 minutes.

Figure 5: Diagnostic pelvic angiogram in a 70 year old female with a right ureteroarterial fistula. Digital subtraction angiogram obtained during contrast injection through a C2 glide catheter (curved arrow) with its distal tip in the right external iliac artery over a V18 wire coiled within the urinary bladder (arrowhead). Contrast opacification of the right internal iliac artery (arrow) indicates its autonomy from the fistula traversed by the wire. TECHNIQUE: Diagnostic/therapeutic Flouroscopy. Total contrast used: 70cc of Isovue 250, 75cc of Isovue 300. Total fluoroscopy time: 51minutes.

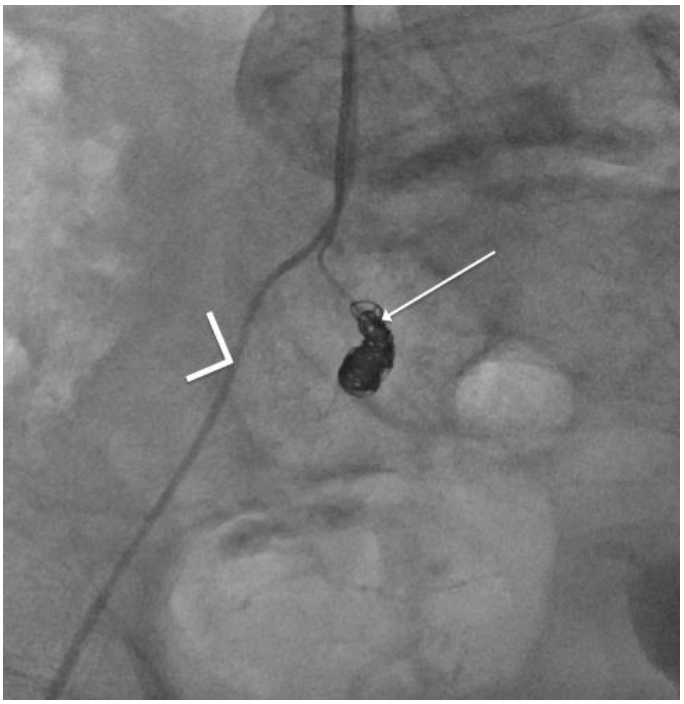

Figure 6: Flouroscopically-guided endovascular therapy in a 70-year-old female with a right ureteroarterial fistula. FINDINGS: Fluoroscopic spot image during coil embolization of the proximal internal iliac artery (arrow) performed in anticipation of excluding the nearby ureteroarterial fistula with a covered stent. TECHNIQUE: Diagnostic/therapeutic fluoroscopy in the PA projection (patient supine) Total contrast used: 70cc of Isovue 250, 75cc of Isovue 300. Total fluoroscopy time: 51minutes.

Figure 7: Flourscopically-guided endovascular therapy in a 70-year-old female with a right ureteroarterial fistula. FINDINGS: Aortoiliac digital subtraction angiogram through a marker pigtail catheter within the distal abdominal aorta (arrow) immediately following coil embolization of the right internal iliac artery (arrowhead). Notice the absence of flow within the right internal iliac artery distal to the embolization coils compared to normal filling of the left internal iliac artery (asterix).TECHNIQUE: Diagnostic/therapeutic fluoroscopy obtained in the PA projection (patient supine). Total contrast used: 70cc of Isovue 250, 75cc of Isovue 300. Total fluoroscopy time: 51minutes.

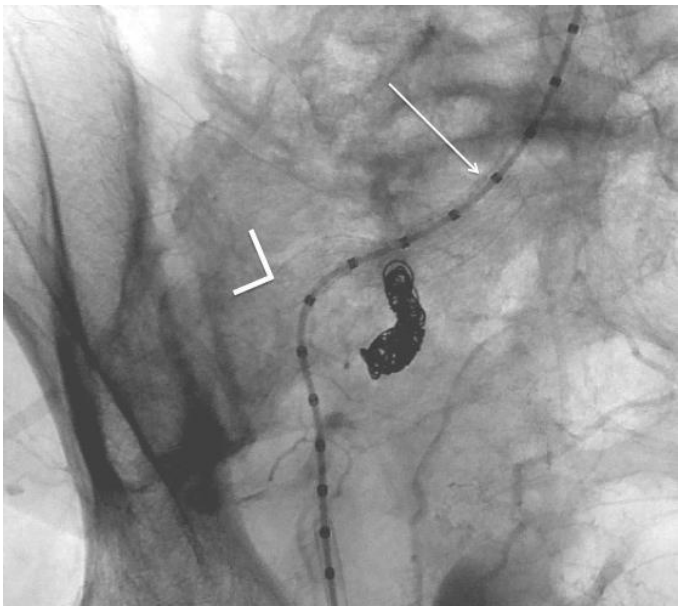

Figure 8 (left): Fluoroscopically guided endovascular therapy in a 70 year old female with a right ureteroarterial fistula. FINDINGS: Fluoroscopic spot image obtained following successful deployment of a 9x5mm Viahban ® covered stent with its proximal apposition site with the distal common iliac artery (arrow) and distal apposition site within the external iliac artery (arrowhead). TECHNIQUE: Diagnostic/therapeutic fluoroscopy obtained in the PA projection. Total contrast used: 70cc of Isovue 250, 75cc of Isovue 300. Total fluoroscopy time: 51minutes.

Etiology	Communication between the iliac artery and mid-to-distal ureter
Incidence	Rare; ~90 cases reported in the literature
Gender Ratio	More common in females
Age Predilection	More common in elderly
Risk factors	Long standing ureteral stents, pelvic surgery/radiation/vascular reconstruction, ureterolithotomy complicated by urinary leak
Treatment	Endovascular stent graft, embolization, surgical ligation, intraoperative balloon occlusion followed by extraanatomic bypass
Prognosis	23% mortality rate
Findings on imaging	**CT:** Non-specific; occasionally show pseudoaneurysm, hydroureteronephrosis, and signs of arterial graft infection. Excretory phase imaging may show abnormal contrast flow into artery at the fistula site. **Iliac Arteriography:** most specific but low sensitivity; fistula may not spontaneously fill with contrast; however, if there is a high index of suspicion, endovascular cannulation should be attempted

Table 1: Summary table for ureteroarterial fistulas

Etiology	Non-Imaging Findings	US	CT	MRI	Angiography
UA fistula	-Uncommon -History of pelvic surgery, radiation and/or indwelling GU stenting -Bleeding can be intermittent	-Not typically utilized -Intravascular US may demonstrate pulsatile flow into ureter	-Hydroureteronephrosis, -Excretory phase imaging may show active extravasation of contrast from the artery to ureter -Arterial phase imaging may show early opacification of ureter	-Unknown; -Theoretically, may show T2 hyperintense, enhancing tract between artery and ureter	-May not spontaneously fill with contrast -With high index of suspicion, cannulation should be attempted
Cystitis	-Urinary urgency, frequency and dysuria -Positive Leukocyte esterase and Nitrites on Urinalysis	-Nonspecific bladder wall thickening -Layering, mobile debris	-Bladder wall thickening -Perivesicular fat stranding	-Bladder wall thickening with T2 prolongation -Perivesicular fat stranding	N/A
Nephro-uretero-lithiasis	Presents with colicky flank pain	-Echogenic foci with acoustic shadowing - "Twinkle artifact" on color Doppler	-Hyperdense foci within the collecting system/ureter system -Indinavir stones are radiolucent and usually undetectable	-May show signal void in ureter/collecting system on heavily T2-weighted images -Abrupt change in ureter caliber suggests obstruction	N/A
Bladder Cancer	-Presents with painless hematuria.	Echogenic mass adherent to the bladder wall without posterior shadowing	-Sessile or pedunculated mass protruding into the bladder lumen -Typically enhances more than surrounding bladder wall - Look for bladder wall invasion and/or enlarged pelvic lymph nodes	-T2 prolongation greater than normal wall but lower than urine -Post contrast enhancement -Useful in assessing bladder wall invasion and locoregional metastasis	N/A
Prostate Abscess	-Suprapubic pain -Fever and dysuria in older males -Urinary retention	-Ill defined, loculated hypoechoic mass with fluid-debris level -may show increased flow on color Doppler	-Enlarged, edematous gland -Unilocular or multilocular hypodense collections +/- fluid fluid levels -Peripheral rim enhancement post contrast	-Heterogeneous T1 hypointense, T2 hyperintense collections -Peripheral rim enhancement	N/A
Autosomal Dominant Polycystic Kidney Disease	Family history of renal failure	-Enlarged kidneys -Multiple anechoic cysts with well-defined, imperceptible walls and posterior acoustic enhancement; -Cysts complicated by hemorrhage or infection will show internal echogenic material without flow	-Enlarged kidneys with innumerable cysts measure fluid density with thin imperceptible wall without enhancement -Complicated cysts may be hyperattenuating with non-enhancing septations or with calcifications -Cysts common in other organs such as liver and pancreas	-Enlarged kidneys with simple cysts demonstrate low T1 and high T2 signal -Enhancement of solid component or septae should raise suspicion for renal cell carcinoma -Must be cautious in administering gadolinium to avoid NSF	N/A
Renal AVM	- Hypertension, - Renal insufficiency - High output heart failure - Flank bruit on exam - History of renal biopsy	-Serpentine, anechoic tubule with turbulent flow on color Doppler -Aneurysmal dilation of draining vein, -Decreased resistive index (<0.4)	-Intrarenal vascular mass with decreased enhancement of renal parenchyma due to blood shunting -Enlarged feeding vessel with diminished nephrogram and atrophy distal to AVM	-Flow voids on non-contrast T1/T2 -Early enhancement of draining veins on arterial phase imaging -On MRA, dilated tortuous vessels with multiple fistulous connections	-Tortuous, dilated vessel -No nephrogram due to blood shunting -No capillary phase following contrast injection -Early contrast filling of renal vein and IVC
Nutcracker Syndrome	-Flank pain -Hematuria -Proteinuria -Perirenal varices -Possible left varicocele	- Large left renal vein diameter ratio between distended and narrowed portions -Parallel orientation of Aorta and SMA -Elevated peak systolic velocity within narrowed segment	-Reduced Aorta: SMA angle (<45^{o}) -Left renal vein stenosis - Collateral vessels - Early enhancement of left gonadal vein on portal venous phase imaging	On MRA, findings similar to CECT	May demonstrate a pressure gradient >3 mm Hg on renal venography

Table 2: Differential diagnosis table for intermittent hematuria.

MRI= Magnetic Resonance Imaging, MRA= Magnetic Resonance Angiography, CECT= Contrast Enhanced Computed Tomography, GU= Genitourinary, IVC= Inferior Vena Cava SMA= Superior Mesenteric Artery, NSF= Nephrogenic Systemic Fibrosis

ABBREVIATIONS

AVM = Arteriovenous malformation
CECT = Contrast Enhanced Computed Tomography
CT = Computed Tomography
F = French
GU = Genitourinary
IVC = Inferior Vena Cava
MRA = Magretic Resonance Angiography
MRI = Magnetic Resonance Imaging
NSF = Nephrogenic Systemic Fibrosis
PCN = Percutaneous Nephrostomy
SMA = Superior Mesenteric Artery
URI = Upper respiratory infection

KEYWORDS

Ureteroarterial fistula; intermitted hematuria; selective iliac angiography; colovesical fistula

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