

Case Presentation

A Highly Uncommon Variation of Spontaneous Urinary Extravasation as an Unusual Presentation of Distal Ureteric Calculus

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Abstract

Urolithiasis is a leading cause of acute abdominal pain and one of the most common conditions seen in emergency departments worldwide. Spontaneous Urinary Extravasation (SUE) is a relatively uncommon manifestation of distal ureteric urolithiasis and has a wide spectrum of clinical presentations depending on the site of urine leakage. Perforation could occur at any level from the calyx to the bladder but it is usually seen at the fornices or upper ureter. It is important to distinguish true rupture of the ureter from the forniceal tear with backflow extravasation because clinical presentations, outcomes and treatments are different. In this case study, a 29-year-old man was admitted to our hospital with sudden-onset of left fossa iliaca pain, fever and lower urinary tract symptoms after a short pain-free period following spontaneous cessation of an acute renal colic. He also had a microscopic haematuria and pyuria on his urinalysis. Contrast Enhanced Computed Tomography of the abdomen (CECT) showed an impacted tiny calculus (< 5mm) in the left intramural ureter and a urine extravasation from both the calyceal fornix and the upper ureter. This unique case is a highly uncommon variant of stone-induced SUE because of a special association between these two phenomena, raising both diagnostic and management challenges.

Keywords: Spontaneous urinary extravasation; Urolithiasis; Forniceal rupture; Ureteral rupture

Introduction

Urinary extravasation results from rupture of the urinary collecting system at any level from the calyx to the urethra [1]. It usually occurs after traumatic, iatrogenic, or tumor-induced tear [2,3]. However, Spontaneous Urinary Extravasation (SUE) is an uncommon condition, found in only 0.08% to 1% urograms [3,4]. It occurs in the absence of external trauma, prior ureteric manipulation, external compression, destructive kidney disease, previous surgery and stone-induced pressure necrosis [5,6]. Reviewing the literature, SUE is commonly associated with ureteral obstruction and attributed to ureteral calculus in approximately 50% of cases [3]. However, SUE is a relatively uncommon manifestation of distal ureteric calculi and has a wide spectrum of clinical presentations depending on the site of urine leakage. Perforation could occur at any level of the excretory tract but it is usually seen at the fornices and upper ureter. Spontaneous forniceal rupture is the most commonly described phenomenon leading to SUE, while spontaneous ureteral rupture is an extremely rare urological disorder; with only a few cases reported worldwide.

It is important to distinguish rupture of calyceal fornix from true ureteral rupture, as the latter can be more severe, requiring more aggressive treatment. Herein, we report a case of spontaneous rupture of both the fornix and the ureter secondary to a tiny intramural ureteral calculus and review the current literature, discussing the clinical and radiological dilemma of both phenomena.

Case Presentation

A 29-year-old previously healthy man presented to the Emergency Department complaining of a sudden onset of left iliac fossa pain, nausea, vomiting and fever, associated with increased urinary frequency and urgency of urination. He reports having experienced recurrent episodes of intractable left renal colic 10 hours before, all of which spontaneously resolved. He had an unremarkable medical history; there was no history of abdominal trauma, surgery or previous urological or kidney diseases.

On physical examination, he has a temperature of 38, 8 °C, a pulse rate of 114 beats / min, and a blood pressure of 110/56 mm Hg. His abdomen was nondistended and the bowel sounds were normal. However, he had left costovertebral angle tenderness and painful left hemi-abdomen without any signs of peritoneal irritation. The rest of physical exam was unremarkable.

Urine dipstick testing was positive for both microscopic haematuria and leukocyte esterase activity. Laboratory investigations showed moderate leukocytosis (13300/ μ l) with predominant neutrophils (96%), elevated C-reactive protein 34 mg/l and normal serum creatinine. Urinalysis revealed pyuria with microscopic haematuria and urine culture was sterile.

Unenhanced computed tomography of the abdomen showed a left intramural ureteric calculus that was less than 5mm in diameter with ipsilateral perirenal, peripelvic and retroperitoneal fluid collections,

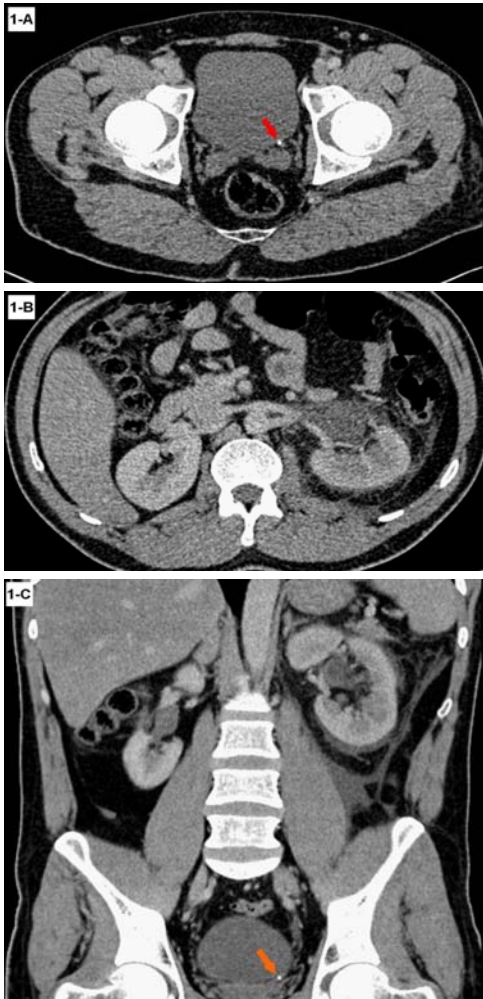


Figure 1: SUE from the left upper urinary tract in 29-year-old man. Axial images (A,B) and coronal reformat (C) of unenhanced CT scan of abdomen shows an impacted calculus in the left intramural ureter (red arrows) with perirenal, peripelvic and retroperitoneal fluid collections extending down to the left iliac fossa along with the ipsilateral psoas muscle.

extending down to the left iliac fossa along with the ipsilateral psoas muscle (Figure 1A, 1B and 1C).

Contrast Enhanced Computed Tomography (CECT) scan of the abdomen demonstrated in the excretory phase (10 minutes after contrast medium injection) an extravasation of contrast medium outside the left pelvicalyceal system; with the coronal reformats and 3D volume rendering reconstructions of the delayed phase, extravasation of contrast medium around the left upper ureter and ureteropelvic junction as well as around the middle calyx was clearly demonstrated (Figure 2A, 2b and 2C). The patient had neither delay in the left kidney function nor hydronephrosis. However, there was moderate left hydroureter without ureteral luminal enhancement (Figure 3A and 3B).

Based on the imaging results, the patient was diagnosed with spontaneous urinary extravasation caused by rupture of calyceal fornix and ureteral rupture, which seems to be related to the calculus impaction in the intramural ureter.



Figure 2: SUE from the left upper urinary tract in 29-year-old man. Axial image (A) and coronal reformats (C, D) of delayed phase of CECT scan of abdomen shows extravasation of contrast medium around the left upper ureter and ureteropelvic junction (red arrows) as well as around the middle calyx (yellow arrows). Please note that there was neither delay in the left kidney function nor hydronephrosis.

The patient was initially given intravenous antibiotic coverage and taken at the operating room the next morning. He underwent retrograde pyelography study under fluoroscopy which confirmed the extravasation of contrast at the level of the ureteropelvic junction and the calyceal fornix (Figure 4).

To restore urinary tract continuity, the patient underwent successful uretero-renaloscopic lithotripsy and double-J catheter placement. The duration of ureteral catheter stenting was 21 days and his catheter was removed, without any complication. His ruptured ureter healed completely and the patient was discharged with good clinical results.

Discussion

Urolithiasis is a leading cause of acute abdominal pain and one of the most common conditions seen in emergency departments worldwide.

The spontaneous cessation of renal colic in our patient is thought

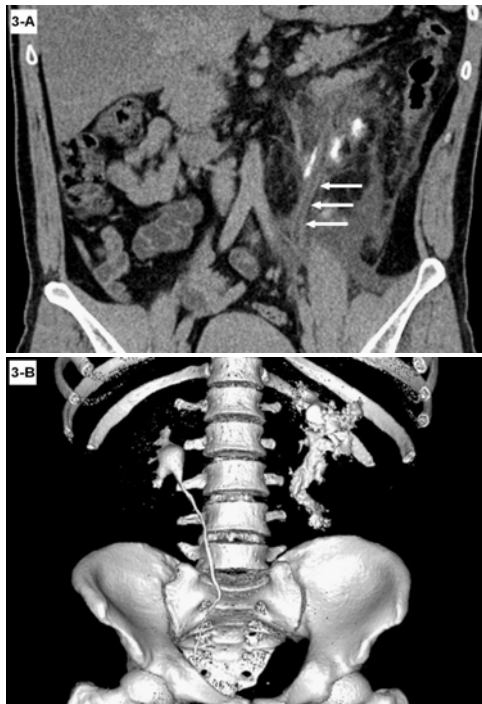


Figure 3: SUE from the left upper urinary tract in 29-year-old man. Coronal reformat (A) and 3D volume rendering reconstruction (B) shows moderate left hydronephrosis without ureteral luminal enhancement (white arrows).

to result from the decompression of the obstructed kidney, related to an impacted ureteric calculus, via rupture of the urinary tract at both the calyceal fornix and the upper ureter.

Spontaneous urinary extravasation is an uncommon presentation of intramural ureteral calculi that are less than 5 mm in diameter. Traditionally, they are likely to pass spontaneously or after medical expulsive therapy, or may mimic cystitis, urethritis, or prostatitis by causing suprapubic pain, urinary frequency and urgency, dysuria, stranguria, or gross hematuria [7].

Spontaneous rupture of the ureter is an extremely rare entity, with urine extravasation occurring into peripelvic and retroperitoneal spaces, while spontaneous forniceal rupture is a most commonly described phenomenon, which leads to urine extravasation into perinephretic space.

In our patient, urine extravasation is thought to result from two different underlying mechanisms. **First**, stone-induced erosion of the ureteral wall at the ureteropelvic junction may result from a downward-moving calculus that obstructs the distal ureter, with elevation of the intraureteric pressure, which subsequently leads to rupture of the ureter at this first weakest point [8,9]. **Second**, the concomitant elevation of the intrapelvic pressure secondary to obstruction may lead to backflow extravasation by rupture of the calyceal fornix, which is the least supported area with a very thin pelvicalyceal system in this point [5,10].

The clinical manifestation of SUE is diverse, ranging from mild flank discomfort to unremitting abdominal pain such as acute abdomen according to the site and the degree of urine leakage.

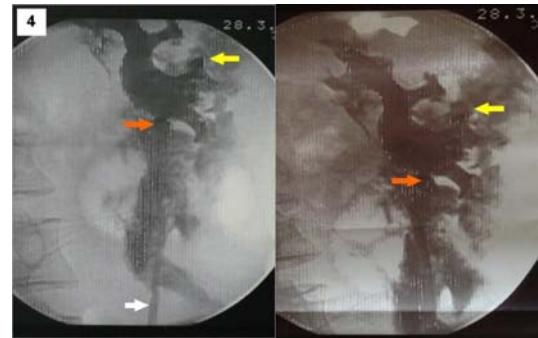


Figure 4: SUE from the left upper urinary tract in 29-year-old man. Fluoroscopic view showing the ureter (white arrow) with the exact sites of rupture of the excretory system; the calyceal fornix (yellow arrows) and the upper ureter (red arrows).

Sometimes, it is associated with nausea, vomiting, dysuria, urinary frequency and hematuria [11].

Clinically, patients who have just sustained a spontaneous forniceal rupture present with mild symptoms such as flank pain, nausea, and vomiting [5,12]. In contrast, most patients with actual rupture of ureter are more unwell with an acute abdomen, a high temperature and leukocytosis than those with backflow extravasation and misdiagnosis as acute appendicitis or diverticulitis is not uncommon [5,12,13].

In our patient, the largest amount of the extravagated urine comes from the ureter and was in close proximity to the peritoneum. As a result of chemical peritoneal irritation, his clinical condition deteriorated overnight as fever, acute abdomen pain and gastrointestinal symptoms developed after a short pain-free period following the cessation of renal colic.

The positive findings on his urinalysis and the associated lower urinary tract symptoms as well as the recent medical history of renal colic were the helpful hints of suspicious urological problem in the present case. However, the absence of these symptoms is insufficient to exclude the diagnosis [13].

When extravasation persists, the urine that collects outside the urinary tract can take the form of an encapsulated collection—a urinoma—or remain as a free fluid known as urinary ascites. With time, it may lead to several serious consequences including infected urinoma, retroperitoneal abscess formation and subsequent irreversible renal impairment; these complications can progress to urosepsis and death [6].

Due to its non specific presentation and possibility for catastrophic complications, SUE should be kept in the differential diagnosis of patients presenting with complex symptoms after renal colic.

Diagnosis is usually made when imaging reveals leakage of contrast, often with extravagated urine, from the site of rupture.

A plain abdominal radiographs are usually nondiagnostic and noncontributing because distended bowels frequently obscure the kidney and ureteral calculus. However, it may show the loss of a retroperitoneal landmark (eg, psoas shadow), antalgic posture of the vertebral column, a stone, or paralytic ileus [6,14,15]. Intravenous

urography has been used extensively for diagnosis in patients presenting with a history renal colic [16,17].

More recently, ultrasound and CT scan have become invaluable in diagnosing urine extravasation.

Ultrasonography is a real-time, inexpensive, repeatable, and radiation-free diagnostic tool. It is easily accessible and time-saving in the emergency department. In cases of ureteral rupture, ultrasonography can detect well a small fluid collection in the perirenal, pararenal and retroperitoneal spaces, and hydronephrosis, and exclude other abdominal pathologies [12,13]. Color duplex Doppler sonography parameters, including the resistance index and mean pulsatility index of the interlobular arteries, give an indirect assessment of the intrinsic pressure of the renal pelvis [18].

Given the high index of suspicion for underlying urological problem as well as the symptomatology worsening in our patient, we have already opted to use CECT of the abdomen to gain a panoramic view of the entire excretory system. In fact, intravenous contrast-enhanced CT is the most informative modality and has a higher sensitivity than other imaging modalities [19].

CT scan delayed phase images obtained 5-20 minutes after injection of contrast medium show contrast extravasation in the peripelvic, perinephric, or retroperitoneal spaces [15,20]. The exact site and the degree of leakage can also be readily assessed by CT [19]. Multiplanar reconstructions may further help in delineating the exact site of rupture [10]. False negative results are reduced based on the results of abdominal plain radiography in the excretory phase [10]. Not only is a CT scan less time-consuming than an intravenous pyelogram, but it can also detect urological pathological conditions that may be missed on excretory urography, such as tumor, infection, and renal infarction. Moreover, it also helps in the diagnosis of diseases other than urogenital problems, including gastrointestinal diseases such as acute appendicitis, diverticulitis or cholecystitis, and vascular disease including abdominal aortic or iliac artery aneurysm [13]. The easy availability and lack of preprocedural preparation make it a more feasible alternative for diagnosis than intravenous urography [6].

In our patient, we diagnosed forniceal and ureteral rupture via the extravasation of contrast medium from the perforated middle calyceal fornix ureter and ureteropelvic junction respectively with delayed contrast-enhanced CT.

The association of both phenomena, however, is a very rare condition and one of them is likely to be misdiagnosed. Moreover, there was some confusion between the backflow extravasation and true rupture of the ureter in the medical literature [13]. As in our case, differentiation between these two conditions may be made by some radiological observations; the presence of contrast material around the calyx highly suggests the forniceal rupture [5, 12], while non-visualization of the ureter in the affected side usually indicates rupture of the ureter [12]. If the radiological picture remains unchanged for a long period of time, it always points to the possibility of the ureteral rupture, while in cases of the forniceal rupture; contrast extravasation usually disappears in 24-48 hrs after the onset of an attack [5,12]. In our case, contrast media injection under fluoroscopy was performed 24 hours lasting the onset of the pain but both radiological findings remained unchanged.

It is important to distinguish rupture of calyceal fornix (backflow extravasation) from true ureteral rupture, as the latter can be more severe. In fact, ureteral rupture complicated with retroperitoneal abscess and sepsis can result in death [21]. On the other hands, forniceal rupture generally resolves spontaneously without any sequelae and it is commonly considered as one of the physiological protective mechanism for preservation of renal function in patients with urinary obstruction. However, peri-renal abscesses can occur as delayed complications in approximately 10% [22].

A clear distinction between these two conditions becomes very important since also treatment is different.

Previously, the treatment of choice for spontaneous rupture of the ureter was open surgery, including nephrectomy or incision with drainage. However, with advances in technology, minimally invasive endourological procedures such as double-J catheter placement, percutaneous drainage and ureteroscopic retrieval of the stones have gained popularity. Many studies have highlighted the significant benefits of this approach [13,19].

Recently, successful conservative management with supportive measures alone; including analgesics, antibiotic coverage and hydration under careful monitoring has been reported in the literature [11,14]. These studies suggest that nonoperative management should be considered first for patients in stable clinical condition to avoid unnecessary surgery and its associated complications, but advised that endourological intervention may still be necessary if conservative management fails.

In contrast, backflow extravasation is usually managed conservatively with analgesics and relief of obstruction. Antibiotic coverage is mandatory in chronic state to prevent infection. When forniceal rupture is associated with ureteric stones, treatment often requires endoscopic lithotripsy and ureteral stenting [6,12,15]. Small urinomas tend to resolve spontaneously this supportive management. When patients have larger urinomas or persistent leaking of the collecting system, the combined use of a Percutaneous drainage catheter with both a nephrostomy catheter and a ureteral stent or with a nephroureteral catheter diverts the urine away from the area of the leak and promotes healing of the collecting system [6,12,15].

However, given the potential for catastrophic complications such as retroperitoneal abscess, we would recommend endourological repair in all those patients, able to undergo endoscopic management to avoid both early and late complications.

Conclusion

Due to its non specific presentation and possibility for catastrophic complications, spontaneous urinary extravasation should be kept in the differential diagnosis of patients presenting with complex symptoms after renal colic. Spontaneous rupture of the fornix associated to spontaneous ureteral rupture is a very rare entity. Distinction between two phenomena can be made using delayed contrast-enhanced imaging techniques. It is important to distinguish isolated forniceal tear with backflow extravasation from that associated with true rupture of the ureter because clinical presentations, outcomes and treatments are different.

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