Nephrocutaneous fistula as the initial manifestation of asymptomatic nephrolithiasis: A call for radical management

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Abstract
Renal stones are a common affliction presenting in an acute setting. We report a case of asymptomatic renal stone in an elderly gentleman presenting initially as a discharging lumbar sinus managed by subcapsular nephrectomy and radical excision of the fistula tract. Nephrocutaneous fistula is most commonly associated with tuberculosis, xanthogranulomatous pyelonephritis, and rarely with complicated calyceal stones, and its occurrence with asymptomatic pelvic stones is rare. We present the points in favor of radical open surgery in the management of such patients.

Key Words: Chronic pyelonephritis, nephrocutaneous fistula, nephrolithiasis, nephrectomy

INTRODUCTION
Renal stones are a common urological emergency and frequently present as an acute colic. Rarely urolithiasis may remain asymptomatic for a prolonged period and present later as an advanced or complicated disease. Nephrocutaneous fistula is one such rare complication that has been scarcely reported, limiting our experience regarding its management to isolated case reports.

CASE REPORT
A 60-year-old gentleman presented to the outpatient department with complaints of a wound on the left lumbar region associated with foul smelling discharge for the past 6 months. The patient had no other complaints associated with the wound. The wound had appeared initially as a swelling which burst spontaneously after taking antibiotics, 6 months ago. Since then the patient had been undergoing treatment in the form of antibiotic and anti-inflammatory agents off and on with regular dressings. The discharge used to reduce while the patient took the medication but increased again after stopping therapy. There was no history of associated abdominal pain, bowel complaints, or urinary disturbances. The patient gave history of trauma to the left leg 1 year back leading to fracture of the neck of femur for which he underwent placement of a hip screw. Examination revealed a single wound on the left lumbar region [Figure 1] associated with mucoid discharge. There was no surrounding erythema or tenderness and the abdominal examination was essentially normal. Urine culture grew E. coli sensitive to nitrofurantoin, and was repeated after 1 week of therapy to ensure sterility of the urinary tract. An X-ray of the abdomen and KUB region was suggestive of a solitary renal calculus and a radio-opaque hip screw in the left hip joint. A sinogram was performed using 60% urograffin which revealed a complex fistula communicating with the left pelvicalyceal system [Figure 2]. An intravenous pyelogram performed with the intent of assessing the functional status of the kidney showed a non-functioning left kidney with a solitary pelvic calculus on the left side and a normally functioning kidney on the right side. A renal nuclear scan with DTPA showed poor function of the left kidney.

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left-side function with a GFR of 8 ml/min and a differential function of 11%. Based on these findings the patient was planned for nephrectomy and excision of the sinus tract. The patient was operated in a flank position by open flank approach, and dense adhesions were present in the renal fossa. A subcapsular nephrectomy was performed along with radical excision of the sinus tract, most of which was located in the subcutaneous plane [Figures 3 and 4]. A drain was placed and the incision was closed in layers. The patient had an uneventful postoperative course and was discharged on the 5th postoperative day. On follow up for a period of 6 months, the patient had no fresh complaints.

**DISCUSSION**

Nephrocutaneous fistula is a rare complication of renal stones and its presentation as the initial manifestation of nephrolithiasis is uncommon. These fistulae arise in a setting of chronic disease such as xanthogranulomatous pyelonephritis, tuberculosis of the kidney, chronic stone disease, and occasionally post trauma or surgery. Fistulas between kidney and adjacent organs like the pleura, colon, and lungs are also uncommon but renal communication with the skin is the rarest of all renal fistulae. Most fistulae are short and form along the path of least resistance, thereby presenting in the region of either the superior or inferior lumbar triangle. The formation of a renal abscess has been described as an initial step in formation of spontaneous fistulae; however, renal surgery or trauma can lead to formation of such fistulae without passing through this stage.

Our review of the literature revealed xanthogranulomatous pyelonephritis to be the most common cause of nephrocutaneous fistula, followed closely by tuberculosis of the kidney. Fistula formation after surgery was the next leading cause of fistula formation. Spontaneous fistula in a case of asymptomatic renal stone is uncommon. Chronic infections of the kidney like xanthogranulomatous pyelonephritis and tuberculosis are notorious in their presentation, and occasionally lead to
formation of fistulae with adjacent structures. These conditions are usually recognized post surgery, on histopathological examination of the tissue. Tuberculosis is recognized by altered shape of the kidney and the presence of giant and epitheloid cells among inflammatory exudates. Xanthogranulomatous pyelonephritis reveals the presence of foamy lipid laden xanthoma cells arising in a chronically inflamed kidney. In our case, the biopsy of the specimen did not reveal any foci of tuberculosis or xanthogranulomatosis. The only changes seen were those of chronic pyelonephritis.

It is important to emphasize that most cases are associated with non-functioning kidneys due to damage of functional parenchyma by long standing inflammation. Unlike fistulous communications with other organs, which become symptomatic early in the course of illness, fistulae to the skin represent a late stage of the chronic pathology. By the time fistulae manifest, the renal tissue has been rendered non-functional by the underlying pathology. All cases reported in the literature were associated with nephrolithiasis and chronic urinary tract infection,[5] which by itself is capable of damaging the entire kidney.

In an era of minimally invasive surgery the treating urologist is often tempted to undertake sub‑radical measures to manage renal fistulae.[6] Nephrocutaneous fistula is one condition that deserves radical excision of the fistula along with removal of the non functioning kidney. A definitive diagnosis regarding the pathology can often be made only after histopathology and leaving the fistula tract in situ may be a cause of recurrence and morbidity. In our patient, excision of the fistulous tract did not significantly add to the morbidity or length of stay. It is thus better to excise the pathology in its entirety in a single procedure.

Early surgical intervention in the form of nephrectomy and removal of the sinus tract can prevent morbidity and mortality in these patients.[7] It is the association of this condition with chronic complicated pathologies that makes less invasive measures prone to recurrence. One exception to radical management is the fistulae arising in an acute setting like trauma or surgery.[8] Such cases have been managed conservatively with good results.

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REFERENCES

1. Sherman SC, Limkakeng A. Xanthogranulomatous pyelonephritis with a nephrocutaneous fistula. J Emerg Med 2005;29:337-8.
2. Qureshi MA. Spontaneous nephrocutaneous fistula in tuberculous pyelonephritis. J Coll Physicians Surg Pak 2007;17:367-8.
3. Bryniak SR. Primary spontaneous renocutaneous fistula. Urology 1983;21:516-7.
4. Maheshwari PN, Shah HN. Nephrocutaneous fistula through percutaneous nephrolithotomy scar: Rare presentation of genitourinary tuberculosis. Urology 2005;66:655-6.
5. Antunes AA, Calado AA, Falcão E. Spontaneous nephrocutaneous fistula. Int Braz J Urol 2004;30:316-8.
6. Kijvikai K, Dissaranan C, Chalermsanyakorn P, Matchariyakul C, Kochakarn W. Hand‑assisted laparoscopic nephrectomy for xanthogranulomatous pyelonephritis with nephrocutaneous fistula after failed flank exploration. Surg Laparosc Endosc Percutan Tech 2006;16:263-6.
7. Ansari MS, Singh I, Dogra PN. Spontaneous nephrocutaneous fistula — 2 unusual case reports with review of literature. Int Urol Nephrol 2004;36:239-43.
8. Doddamani D, Hemal AK, Ansari MS. Ureteral stent — help or hindrance? In healing of post traumatic nephrocutaneous fistula. Int Urol Nephrol 2001;33:621-3.

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