Xanthogranulomatous pyelonephritis with reno-colic fistula: A rare complication of urinary tract infection

Onn LV, Bickle I, Chua HB, Telisinghe PU, Chong CF, Chong VH


**Abstract**

Urinary tract infection (UTI) is one of the most common presentations in general practice and, in most instances, occurs in a single episode and is easily treated with a course of anti-microbial therapy. However, when a patient has recurrent UTIs, it is important to consider evaluation for any underlying causes. We report the case of a 32 year old female who had recurrent UTIs; this was a case of recurrent UTI secondary to xanthogranulomatous pyelonephritis from renal stones with resultant reno-colic fistula formation.

**Introduction**

Urinary tract infection (UTI) is one of the most common presentations in general practice and is easily treated with a course of anti-microbial therapy. However, when a patient has recurrent UTIs, it is important to consider evaluation for any underlying causes. We report a case of recurrent UTIs secondary to xanthogranulomatous pyelonephritis (XGP) from renal stones with resultant reno-colic fistula formation.

**Case Summary**

A 32-year-old female with a background history of type 2 diabetes mellitus, hypertension, and hyperlipidemia was admitted for evaluation of recurrent UTIs. Prior to this, she had had multiple outpatient clinic visits with recurrent urinary symptoms over the past year. Each time, she had been treated for UTIs with courses of antibiotics. Despite these, the frequency of her visits seemed to be increasing. Several times, urine cultures had...
isolated *Escherichia coli* (*E. coli*). At the latest presentation, she experienced increasingly persistent left flank pain and mild fever, but without urinary symptoms. She denied any respiratory symptoms or any significant weight loss. There was no family history of kidney disease or any cancers. On examination, she was febrile and had a mildly tender, enlarged left kidney. Chest examination was normal. Her blood work showed mild leucocytosis (white cell count 13.6 x 10⁹, [Normal range 3.6-10.2 x 10⁹]), mild acute kidney injury (Serum Urea 8.0 [3.2-7.4], and creatinine 130.4 µmol/L [63.6-110.2]), and elevated C-reactive protein (C-reactive protein 5.45 mg/dL [<0.49]). She was started on a broad-spectrum intravenous antibiotic (amoxicillin-clavulanic acid at 1.2gm three times a day).

An ultrasonography scan of the abdomen showed an enlarged left kidney with severe hydronephrosis, and multiple calculi in the renal pelvis with reduced renal cortical thickness. These findings were confirmed by computed tomography ([Figure 1a](#)), but the scan also revealed intravenous contrast in the descending colon. Antegrade pyelogram confirmed the presence of a renocolic fistula ([Figure 1b](#)).

After discussion, the patient proceeded with a laparoscopic assisted left nephrectomy. During surgery, the enlarged left kidney was noted to be surrounded by inflammatory changes attached to part of the descending colon. The area with the fistula was identified, and the kidney and affected part of the descending colon were resected. Gross examination of the resected specimen showed a grossly enlarged and inflamed left kidney with a fistula seen between the inflammatory mass and the colon. Histology showed large amounts of fibrosis and inflammatory cells, along with a large number of foamy histiocytes ([Figures 2 a and b](#)). The findings were all consistent with XGP. Post-surgery, the patient remained well without any further episode of UTI during a follow-up of several years.

**Figure 2: a) Resected kidney showing two glomeruli with peri-glomerular fibrosis, and an interstitium infiltrated by large number of foamy histiocytes, lymphocytes, and plasma cells (H&E stain x 20), and b) A magnified view showing destruction of the renal parenchyma and tubules.**

**Discussion**

XGP is an uncommon inflammatory disorder that is characterized by the abundance of foamy histiocytes. It was first described by Sclagenhauer in 1916¹, and, although cases continue to be reported, it is now much less common than it was in the pre-antibiotic era. XGP is commonly associated with urinary tract pathology, such as recurrent UTIs, lithiasis (nephrolithiasis or urolithiasis), or urinary obstructions. It is a condition that affects adults and children.¹⁻³

Manifestations of XGP are classified into three categories: Stage 1 (localized disease confined to the renal parenchyma), Stage 2 (involvement of the perinephric fat), and Stage 3 (extension into the perinephric space).² Renocolic fistula, the extreme spectrum of Stage 3, is generally rare with very few reported cases.¹⁻⁴

The underlying mechanism of fistula formation is through a combination of ongoing inflammatory processes and increases in the pressure on the renal calyceal system secondary to stones. Superadded infections lead to necrosis of the thinned cortex, and this can lead to perinephric abscess formation, which subsequently perforates through structures. In most cases, as in our case, the adjacent left colon is affected, resulting in the
formation of a renocolic fistula. Among the reported complications associated with fistula formation, renocutaneous and renobronchial fistulae are the most common.

The clinical presentations of XGP are similar to pyelonephritis or UTI, but are typically recurrent, and, in the later stage, an enlarged kidney is invariably present. In our case, both stones and the renocolic fistula are causes of the recurrent UTIs. The most common organisms isolated are *Escherichia coli* (*E. coli*) or *Proteus species*. In the more advanced stages, kidney function deteriorates, and the kidney can become non-functional.

Diagnosis of XGP can be a diagnostic challenge as the clinical and radiological manifestations resemble malignancies and other infections. Manifestations as renal and Wilms tumors have been reported. Coexistence of XGP with renal neoplasm has also been reported. Parekh et al. reported the coexistence of XGP and renal tubulopapillary adenomas. Sarcoïdosis affecting the kidney has also been reported to mimic XGP. Other causes of a renocolic fistula include renal tuberculosis and diverticulitis.

Radiological manifestations are variable. The classical radiological change reported is the ‘Bear Paw’ sign, with pockets of abscesses resembling the paw of a bear. However, such a manifestation may not always be present. In our case, there were pockets of abscesses that did not have the classical features of the ‘Bear Paw’ sign. Radiological manifestations depend on the stage of disease. Inflammatory or septic changes are localized to the kidney in Stage 1, but adjacent organs are involved in Stages 2 and 3 of the disease. In a case of XGP presenting with septic emboli, the renal lesions were lobulated and localized and were misdiagnosed as renal cell carcinoma. In most cases, the diagnosis of XGP is only made after surgery. During the intraoperative period, the diagnosis of XGP can also be difficult as the gross appearance of XGP, a yellowish mass with focal necrosis and hemorrhage, can resemble renal cell carcinoma.

The prognosis of XGP depends on the stage of disease at diagnosis. Adequate treatment of early stages (Stages 1 and 2) with antimicrobials, with or without surgery, is generally curative. However, the condition may reoccur if not adequately treated. For those patients at Stage 3 of the disease, especially those in the extreme spectrum where adjacent organs are involved, surgery is required, followed by adequate antimicrobials. XGP with septic extensions or emboli has been reported. Cases of psoas abscess and lung abscesses secondary to XGP have also been reported.

Management of XGP often involves nephrectomy and excision of the fistula. Use of antimicrobials alone is generally not an effective cure, as in our patients. Although rare, recurrence in the remnant kidney can occur, and the patient should continue to be monitored.

In conclusion, our case of XGP with renocolic fistula highlights that frontline clinicians should consider XGP in patients who present with recurrent UTIs. Although benign, the condition can have malignant-like manifestations through infiltrations of the adjacent structures. Imaging should be done early, and the presence of enlarged kidney(s), especially those with stones, should be evaluated for XGP. In addition, adequate treatment of the stones, if detected early, would have prevented the progression to XGP.

References


