

Bilateral Xanthogranulomatous Pyelonephritis With Renocolic and Duodenal Fistulas

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Review began 10/11/2026

Review ended 10/18/2026

Published 10/25/2026

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DOI: 10.7759/cureus.118557

Abstract

Xanthogranulomatous pyelonephritis (XGP) is an uncommon destructive inflammatory disease of the kidney associated with chronic urinary obstruction, calculi, and infection. Bilateral disease is exceptionally uncommon and creates a particular therapeutic problem because definitive nephrectomy may compromise native renal function.

We describe a 65-year-old man with chronic kidney disease and recurrent nephrolithiasis who presented with shortness of breath and severe microcytic anemia without prominent urinary symptoms. Serum creatinine was 4.67 mg/dL, increased from prior values of approximately 1.3-1.8 mg/dL. Imaging demonstrated bilateral staghorn calculi, marked parenchymal destruction, and characteristic bear-paw morphology, with more extensive disease on the right. Bilateral nephrostomy tubes were placed for decompression, after which serum creatinine improved to approximately 3 mg/dL. Repeat CT demonstrated persistent bilateral disease, multiple right perirenal fluid- and gas-containing collections, and a fistula between an abscess anterior to the right kidney and the proximal transverse colon. The patient subsequently underwent right nephrectomy, during which a duodenal fistula was also identified. Histopathologic re-review demonstrated lipid-laden foamy macrophages with suppurative inflammation, confirming XGP. Left nephrectomy was deferred at the patient's request.

This case highlights a systemic presentation of advanced bilateral XGP without prominent urinary symptoms, its potential for enteric fistulization, and the need to balance definitive source control against preservation of native renal function.

Categories: Urology, Internal Medicine, Nephrology

Keywords: bilateral renal disease, duodenal fistula, nephrectomy, renocolic fistula, staghorn calculi, xanthogranulomatous pyelonephritis

Introduction

Xanthogranulomatous pyelonephritis (XGP) is an uncommon chronic inflammatory disease in which renal parenchyma is progressively destroyed and replaced by inflammatory tissue containing lipid-laden macrophages. Chronic urinary obstruction, renal calculi, and infection are frequently associated with the disease, and computed tomography (CT) is central to identifying the characteristic renal abnormalities and defining extension beyond the kidney [1,2]. Typical presentations are insidious and nonspecific and may include flank or abdominal pain, fever, weight loss, and lower urinary tract symptoms, although urinary symptoms can be minimal [1,2].

Most XGP is unilateral. In a systematic review of 1,159 patients, only four (0.4%) had bilateral involvement among the included cohorts [1]. Bilateral cases have also been reported outside those cohorts, including selected patients treated with bilateral partial nephrectomy [3]. This distinction is clinically important because nephrectomy remains a common definitive treatment for diffuse destructive XGP, whereas removal of both kidneys necessarily eliminates native renal function.

We report a patient whose evaluation for severe anemia and shortness of breath unexpectedly revealed bilateral radiographic XGP with extensive right-sided disease, perirenal abscess formation, and enteric fistulization. This case highlights two practical challenges: recognizing advanced XGP when urinary symptoms are not prominent and planning definitive source control when bilateral disease limits the usual nephrectomy-based strategy because preservation of contralateral renal function becomes a central consideration.

Case Presentation

A 65-year-old man with hypertension, type 2 diabetes mellitus, peptic ulcer disease, chronic kidney disease stage 3, and recurrent nephrolithiasis was referred to the hospital by his primary care physician after outpatient testing showed severe anemia. Prior serum creatinine values had fluctuated between approximately 1.3 and 1.8 mg/dL. He had experienced multiple previous episodes of nephrolithiasis that

How to cite this article

Din N (October 25, 2026) Bilateral Xanthogranulomatous Pyelonephritis With Renocolic and Duodenal Fistulas. Cureus 18(10): e118557. DOI 10.7759/cureus.118557

generally resolved with spontaneous stone passage and had subsequently been lost to follow-up. His primary complaint was shortness of breath. He denied chest pain, fever, and abdominal pain, and urinary symptoms were not prominent in the recorded presentation.

Resting vital signs were stable, although orthostatic hypotension was present. He appeared uncomfortable and in moderate distress. Examination showed abdominal distention without palpable organomegaly.

Laboratory evaluation demonstrated microcytic anemia with a hemoglobin of 6.6 g/dL, serum creatinine of 4.67 mg/dL above his previously documented baseline, aspartate aminotransferase of 49 U/L, and C-reactive protein greater than 160 mg/L. Urinalysis showed pyuria, and urine culture grew *Proteus mirabilis*. He received two units of packed red blood cells, after which his hemoglobin stabilized, and antimicrobial therapy was given for the urinary infection.

Because of the marked renal dysfunction, renal ultrasonography was obtained. Both kidneys were enlarged and contained large staghorn calculi, with extensive dilated cystic-appearing calyceal spaces and marked thinning of the remaining renal parenchyma (Figure 1).

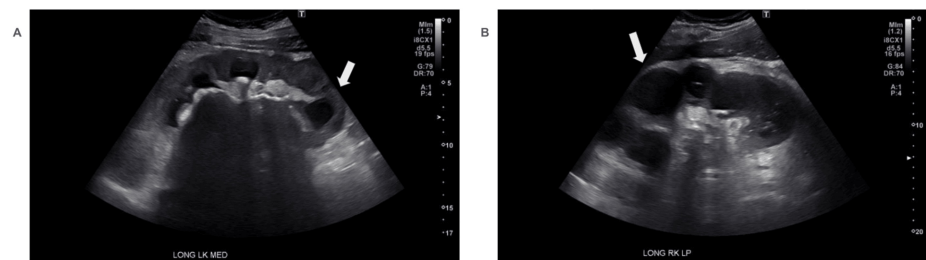


FIGURE 1: Bilateral Renal Ultrasonographic Findings

(A) Long-axis ultrasound image of the left kidney demonstrating marked distortion of the renal architecture, dilated calyceal spaces, and a large echogenic calculus with acoustic shadowing (white arrow). (B) Long-axis ultrasound image of the right kidney demonstrating a large echogenic calculus with acoustic shadowing (white arrow) and marked architectural distortion.

Noncontrast CT of the abdomen and pelvis confirmed markedly enlarged bilateral kidneys with large staghorn calculi. Multiloculated low-attenuation regions surrounding the calculi produced the characteristic bear-paw appearance of XGP (Figure 2). The right kidney was larger and more extensively involved than the left. Bilateral perinephric inflammatory changes were present, with gas within the right-sided inflammatory process. The initial examination also raised concern for fistulous communication with adjacent bowel, although the anatomy was not clearly delineated.

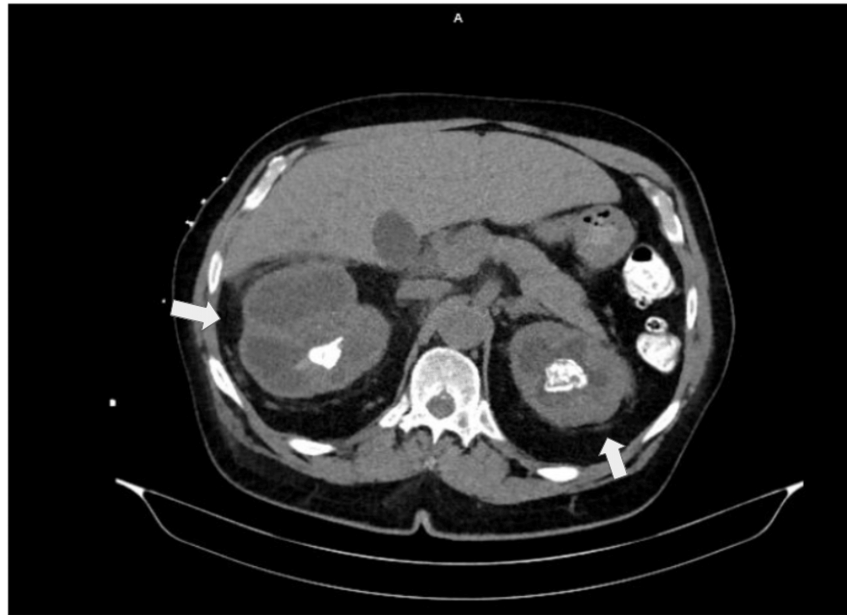


FIGURE 2: Preoperative CT Findings of Bilateral Xanthogranulomatous Pyelonephritis

Axial noncontrast computed tomography demonstrates bilateral staghorn calculi, renal enlargement, and destructive multiloculated renal changes characteristic of xanthogranulomatous pyelonephritis. The right kidney was more extensively involved. White arrows indicate the affected renal regions.

Taken together, the bilateral staghorn calculi, severe parenchymal destruction, bear-paw morphology, pyuria, *P. mirabilis* infection, and renal dysfunction strongly supported bilateral XGP. Bilateral percutaneous nephrostomy tubes were therefore placed for decompression and drainage, with purulent material obtained during the procedure. Serum creatinine subsequently improved slowly to approximately 3 mg/dL by the time of his transfer.

Repeat CT approximately one week later showed improvement in collecting-system dilation following nephrostomy placement but persistent destructive changes involving both kidneys. Multiple small fluid- and gas-containing collections were evident in the right perirenal region. Importantly, the radiology report described a fistula between an abscess collection anterior to the right kidney and the proximal transverse colon. The fistula was identified on CT; no retrograde pyelography was performed.

The patient was transferred to a long-term acute care facility while nephrectomy was being arranged. He subsequently underwent right nephrectomy. At surgery, a duodenal fistula was also identified.

Initial histopathologic review did not identify diagnostic features of XGP. On further review, an additional slide demonstrated sheets of lipid-laden macrophages with abundant foamy cytoplasm and bland nuclei, together with necrotic tissue and prominent neutrophilic inflammation consistent with abscess formation (Figure 3). No malignancy was identified. No immunohistochemical staining was performed; the diagnosis was rendered on the morphologic findings. These findings confirmed XGP in the right kidney.

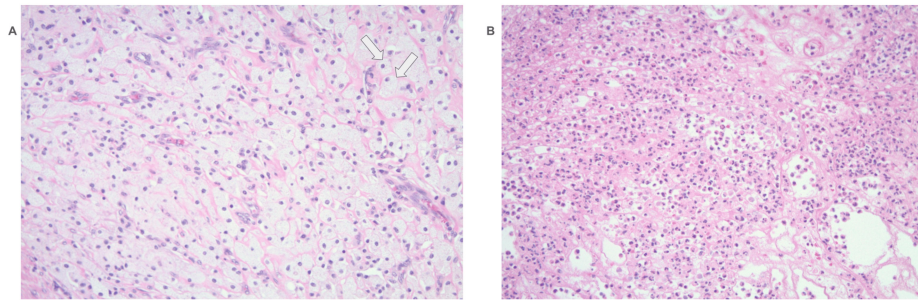


FIGURE 3: Histopathologic Findings of Xanthogranulomatous Pyelonephritis (H&E, x200)

(A) Right nephrectomy specimen demonstrating sheets of lipid-laden macrophages with abundant foamy cytoplasm and bland nuclei; white arrows indicate representative foamy histiocytes. (B) Right nephrectomy specimen demonstrating extensive suppurative inflammation with numerous neutrophils, necrotic debris, and abscess formation. Together, these findings supported the histopathologic diagnosis of xanthogranulomatous pyelonephritis.

Postoperative imaging continued to demonstrate a large left staghorn calculus and marked destructive left renal abnormalities consistent with XGP (Figure 4). Left nephrectomy was discussed but was deferred at the patient's request. No left renal tissue was obtained. After transfer, the patient was subsequently lost to long-term follow-up; later renal function, dialysis status, and the subsequent course of the retained left kidney were unavailable.

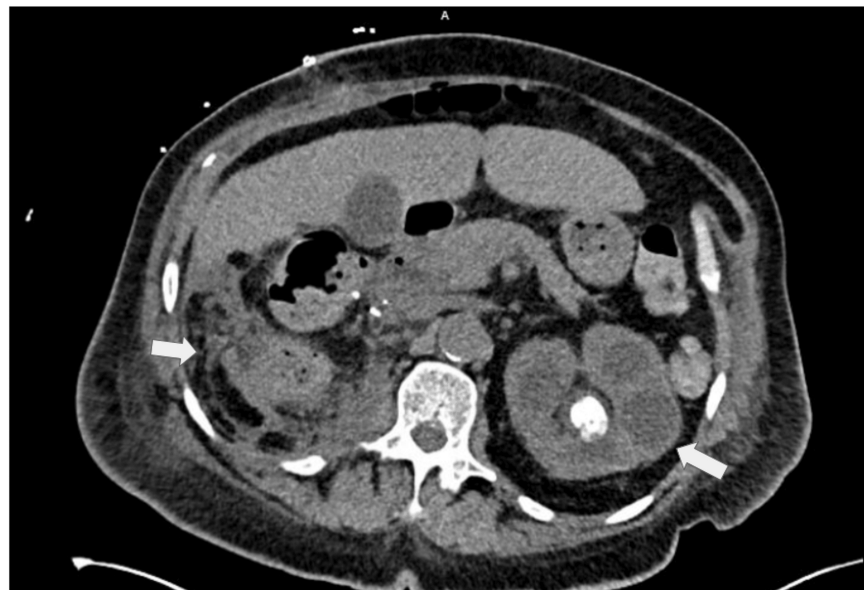


FIGURE 4: Postoperative CT Findings Following Right Nephrectomy

Axial computed tomography following right nephrectomy. The white arrow on the patient's right indicates the right nephrectomy bed, while the arrow on the patient's left identifies the retained left kidney, which demonstrates a persistent staghorn calculus and destructive renal changes.

Discussion

This case is notable for the combination of bilateral disease, an atypical presenting complaint, extensive right-sided inflammatory extension with enteric fistulization, and the therapeutic problem created when a disease commonly treated with nephrectomy affects both kidneys.

XGP is strongly associated with urinary obstruction, calculi, and chronic infection [1,2,4,5]. Our patient had

several characteristic features simultaneously: bilateral staghorn calculi, pyuria, *P. mirabilis* infection, marked renal dysfunction, and extensive destructive renal abnormalities. Despite this disease burden, he did not present with a typical urinary syndrome. Instead, severe anemia and shortness of breath prompted his evaluation.

Anemia and other nonspecific systemic abnormalities have long been described in XGP [4,5]. In this case, however, the available clinical information did not establish a specific cause of the anemia.

CT was particularly important because it demonstrated not only bilateral calculi and parenchymal destruction but also the characteristic bear-paw morphology and the extent of extrarenal disease [2]. The distinction between imaging and tissue confirmation remains important. Both kidneys had highly characteristic radiographic abnormalities in the same infectious and obstructive setting, but histopathologic confirmation was obtained only from the right nephrectomy specimen. The left kidney therefore remained a radiographic diagnosis.

The pathology sequence also illustrates the value of clinicoradiologic-pathologic correlation. XGP may mimic renal malignancy radiographically, and limited tissue sampling can be nondiagnostic [2,6]. In this patient, the initial histopathologic review was nondiagnostic despite highly suggestive imaging. Re-review of the specimen on an additional slide subsequently demonstrated the characteristic foamy macrophages and suppurative inflammation of XGP [6]. Continued diagnostic concern therefore led to the definitive pathologic diagnosis.

The Malek and Elder classification describes stage I disease as confined to the renal parenchyma, stage II as extending into the perinephric fat, and stage III as extending into the pararenal or retroperitoneal tissues and adjacent structures [7]. Given the right perirenal abscesses and fistulous involvement of adjacent bowel, the right-sided disease was consistent with stage III XGP.

XGP can extend beyond the renal parenchyma into the perinephric and surrounding tissues and may produce abscesses or fistulous communication with adjacent organs [2,7]. Renocolic fistulas are uncommon but well described. Numan et al. reported XGP with renocolic fistula in a patient who also presented with severe symptomatic anemia, demonstrating that an anemia-predominant presentation has precedent [8]. CT and direct pyelography have both been used to define renocolic fistulas [9].

In our patient, repeat CT directly demonstrated a fistula between a right anterior perirenal abscess and the proximal transverse colon. A duodenal fistula was also identified during nephrectomy. Detailed operative information was unavailable, so the relationship between these fistulous communications could not be further characterized. What is clear is that the right-sided XGP had extended beyond the kidney and involved adjacent bowel.

Gas was present in the right-sided inflammatory process on the initial examination. Although this raised concern for emphysematous infection at the time, the subsequently demonstrated enteric fistulization provides another potential explanation for intrarenal and perirenal gas. We therefore describe the imaging finding without assigning a separate diagnosis of emphysematous pyelonephritis.

Nephrectomy remains a principal definitive treatment for diffuse or extensively destructive XGP, while antimicrobial therapy, percutaneous decompression, and drainage may be used for stabilization and source control [10,11]. Nephron-sparing strategies, including partial nephrectomy in selected focal disease and staged drainage or stone-directed management, have been reported but are uncommon in diffuse XGP [1-3,10,11]. Contemporary bilateral reports likewise support an individualized or staged approach when renal preservation must be balanced against definitive source control [12]. Formal differential renal function was not available in this patient, so complete nonviability of the right renal unit cannot be established. Bilateral nephrostomy decompression improved serum creatinine, but the right kidney remained more extensively destroyed and was complicated by perirenal abscess formation and enteric fistulization, prompting right nephrectomy for definitive source control. The radiographically affected left kidney was retained after left nephrectomy was deferred at the patient's request.

The principal limitations are the absence of complete operative details and long-term follow-up. Detailed information regarding the surgical approach and management of the enteric fistulas was unavailable, and formal differential renal function was not documented. Consequently, the long-term renal function of the retained left kidney and subsequent dialysis requirements remain unknown.

Conclusions

XGP should be considered when urinary obstruction, infection, renal calculi, and destructive renal abnormalities occur together, even when the presenting complaint is systemic rather than urinary. In this patient, evaluation of severe anemia and shortness of breath revealed characteristic bilateral radiographic disease, while right nephrectomy ultimately provided histopathologic confirmation after repeat pathology review.

Bilateral XGP creates a therapeutic problem distinct from routine unilateral disease because removal of both kidneys eliminates native renal function. This patient underwent bilateral decompression followed by right nephrectomy for the more extensively involved kidney, which had developed perirenal abscesses and enteric fistulization. Recognition of disease extent, adjacent-organ involvement, residual renal function, patient preference, and nephron-sparing options when feasible is essential when planning treatment for bilateral XGP.

Additional Information

Author Contributions

All authors have reviewed the final version to be published and agreed to be accountable for all aspects of the work.

Concept and design: Noren Din

Acquisition, analysis, or interpretation of data: Noren Din

Drafting of the manuscript: Noren Din

Critical review of the manuscript for important intellectual content: Noren Din

Disclosures

Human subjects: Informed consent for treatment and open access publication was obtained or waived by all participants in this study. **Conflicts of interest:** In compliance with the ICMJE uniform disclosure form, all authors declare the following: **Payment/services info:** All authors have declared that no financial support was received from any organization for the submitted work. **Financial relationships:** All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. **Other relationships:** All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

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