

Xanthogranulomatous Pyelonephritis Successfully Treated with Antibiotics Only

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A 73-year-old woman with leukemia presented with urinary tract infection, splenic abscess, and a renal mass. Both urine culture and pus culture of the splenic abscess yielded *Klebsiella pneumoniae*. Percutaneous biopsy of the renal mass confirmed the diagnosis of xanthogranulomatous pyelonephritis. Because of high risk for surgery, the patient received treatment with antibiotic therapy for 2 months. With antibiotic therapy, not only was the splenic abscess cured but follow-up ultrasonography also showed progressive resolution of the renal mass. Xanthogranulomatous pyelonephritis is frequently associated with urinary tract obstruction or nephrolithiasis. In this first report of xanthogranulomatous pyelonephritis in a patient with leukemia and splenic abscess, we provide a short review of xanthogranulomatous pyelonephritis successfully treated with antibiotics only. [*J Chin Med Assoc* 2008;71(12):643–645]

Key Words: antibiotics, renal tumors, xanthogranulomatous pyelonephritis

Introduction

Xanthogranulomatous pyelonephritis (XPN) is an unusual variant of chronic pyelonephritis that is frequently associated with urinary tract obstruction or nephrolithiasis. The nonspecific presentations may include fever, chills, flank pain, malaise, weight loss, dysuria, or a palpable flank mass. The most typical laboratory findings are anemia and leukocytosis. Azotemia and abnormal liver function test may be seen. Affected patients usually have massive destruction of the renal parenchyma, with granulomatous tissue-replacing infiltrates containing lipid-laden macrophages that impart a yellowish-tan appearance to the tissue. It has been believed that removal of the xanthogranulomatous inflammatory tissue is required to cure this disease. Therefore, the long-standing mainstay treatment for XPN has been *en bloc* or partial nephrectomy. However, there have been some reported cases of XPN successfully managed with medical treatment.^{1–5} Herein, we describe a case of XPN concurrent with splenic abscess in a leukemia patient, which was adequately treated with antibiotics only.

Case Report

A 73-year-old woman was admitted with a 1-week history of fever and chills. The patient had a history of left breast cancer and had received operation and adjuvant hormone therapy when she was 50 years old. Positron emission tomography 3 years before this present admission showed no abnormal findings except for thyroid nodular goiter. Chronic myeloid leukemia was diagnosed 1 year previously, and the patient had been maintained on hydroxyurea therapy. Other comorbidities included congestive heart failure and chronic hepatitis B. About 1 month prior to admission, the patient experienced an episode of urinary tract infection and was treated with 1 week of oral ciprofloxacin. Over the ensuing days, she continued to note frequent urination. One week prior to admission, she developed fever with chills and left flank pain, and did not respond to another 1 week of oral ciprofloxacin.

Physical examination on admission found abdominal tenderness in the left upper quadrant area. Complete blood count showed a white blood cell count of



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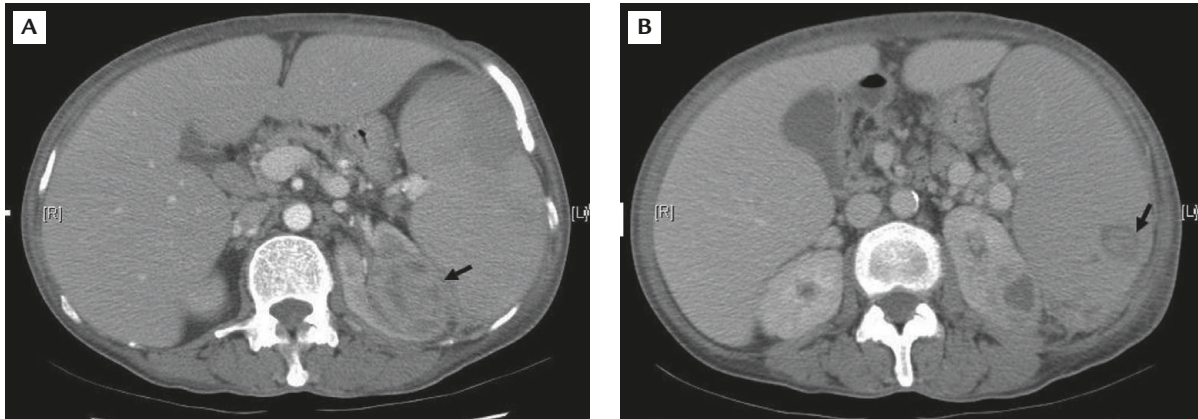


Figure 1. Enhanced computed tomography of the abdomen shows: (A) hypoattenuating mass in the left kidney (arrow); (B) hypodense area in the spleen (arrow).

32,100/ μL with 19.5% segmented neutrophils, 8% lymphocytes, 20% bands and 25.5% blast cells, hemoglobin of 7.6 g/dL, hematocrit of 22.2%, and platelet count of 46,000/ μL . Blood chemistry showed aspartate aminotransferase of 20 U/L (normal, 8–40 U/L), alanine aminotransferase of 11 U/L (normal, 4–44 U/L), blood urea nitrogen of 16.9 mg/dL (normal, 5–25 mg/dL), and creatinine of 1.0 mg/dL (normal, 0.7–1.4 mg/dL). Urinalysis revealed occult blood (++) , protein (+), and nitrite (+), with 20–25 red blood cells, numerous white blood cells, and many bacteria per high-power field in the urinary sediments. Urine culture grew *Klebsiella pneumoniae* with the presence of ciprofloxacin resistance.

Ultrasonography of the abdomen revealed a hypoechoic area in the spleen, with the features of splenic abscess, and a heterogeneously dense mass in the left kidney. Enhanced computed tomography of the abdomen demonstrated a hypodense area in the spleen and a hypoattenuating mass in the left kidney (Figure 1). The differential diagnosis of the renal mass included unliquefied renal abscess, XPN, and renal cell carcinoma. Ultrasound-guided aspiration from the splenic lesion showed thick yellowish pus; however, aspiration from the left renal mass was negative. Pus culture of the splenic abscess also yielded *K. pneumoniae*.

The patient was treated with the intravenous antibiotic cefmetazole after admission. After 2 weeks of antibiotic therapy, follow-up ultrasonography showed resolution of the splenic abscess with no change in the renal lesion. At this time, percutaneous biopsy of the renal mass was performed. The specimen showed many characteristic foamy lipid-laden macrophages of XPN. On periodic acid–Schiff (PAS) stain, no obvious intracytoplasmic PAS-positive Michaelis-Gutmann bodies could be found within the macrophages (Figure 2).

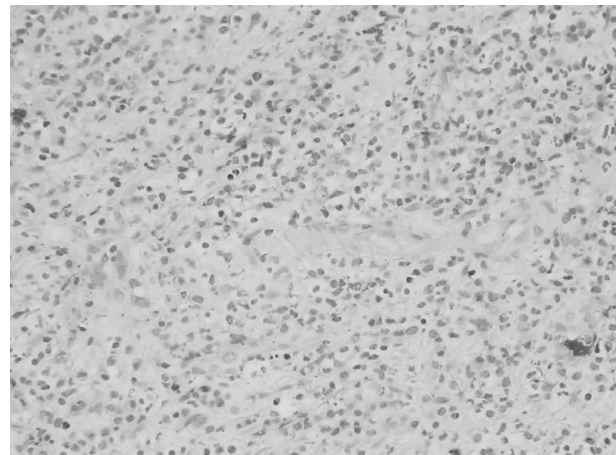


Figure 2. Percutaneous biopsy of the renal mass shows foamy lipid-laden macrophages, with no obvious Michaelis-Gutmann bodies within the macrophages (periodic acid–Schiff stain, 400 \times).

Surgical intervention for XPN was indicated. Because of the patient's underlying comorbidity, the risk for surgery was relatively high. Therefore, the patient was continued on medical treatment. With antibiotic therapy, follow-up ultrasonography 4 weeks later showed that the renal mass had decreased in size. The patient was maintained on the oral antibiotic cefixime for another 1 month, and follow-up ultrasonography showed complete resolution of the renal mass.

Discussion

XPN is a particular type of chronic renal infection with an unusual inflammatory response. Pyuria and bacteriuria are frequent but not uniformly present. One third of patients have sterile urine, and the infectious organism will be found only by tissue culture. The most common

offending organisms are *Proteus mirabilis*, *Escherichia coli*, *K. pneumoniae* and *Streptococcus faecalis*. Computed tomography provides the most useful information, especially by defining the extent of renal involvement and spread to adjacent organs. Computed tomography features that have been considered to be characteristic, though not pathognomonic, for XPN include renal enlargement, strands in the perinephric fat, thickening of Gerota's fascia, and thick enhancing septa separating hypodense areas in the renal parenchyma.⁶ However, focal XPN may be erroneously interpreted as renal cell carcinoma. The heterogeneous and solid aspect on computed tomography is indeed often indistinguishable from hypovascular renal cell carcinoma. Especially when, as in our case, there is no sign of urinary tract obstruction or nephrolithiasis, the diagnosis may be equivocal. It is reported that magnetic resonance imaging may be useful in the differentiation. The diagnosis of focal XPN is suggested in the absence of hyperintensity on fast T2-weighted sequences.⁵ The histologic hallmark xanthoma cells are lipid-laden macrophages or foamy macrophages, which on electron microscopy are initially found to contain bacteria and subsequently contain numerous phagolysosomes. It is hypothesized that XPN may be caused by a lysozymal defect of macrophages that prevents complete digestion of ingested bacteria. The cause of lipid accumulation in the lesion is not well understood.

Although the pathogenesis of XPN remains unknown, the most common factors predisposing to the development of XPN are urinary tract obstruction and nephrolithiasis. The occurrence of XPN has rarely been reported in the absence of clear predisposing uropathy.⁷ To the best of our knowledge, this is the first report of XPN occurring in leukemia patients and concurrent with splenic abscess. Whether the underlying immunocompromised state due to leukemia further predisposes to an abnormal inflammatory response, as seen in XPN, is speculative.

Surgical nephrectomy has been the treatment of choice for diffuse XPN. For focal XPN, most patients undergo surgical exploration because of the difficulty of preoperatively differentiating it from renal malignancy.⁸ Furthermore, the association of malignancy with XPN has prompted several authors to dispute the

wisdom of medical therapy for XPN. As a result, the response to medical therapy has rarely been reported. Focal XPN has been successfully treated with antibiotics only for 3–10 weeks without surgical intervention.^{1,2,4,5} XPN has also been seen in the transplanted kidney. A trial of antibiotic therapy prior to surgery was attempted to salvage the renal allograft and was successful in 2 cases.³

In conclusion, since focal XPN commonly presents with a renal mass and is frequently difficult to differentiate from renal cell carcinoma, maximal efforts, including renal biopsy, should be made to establish the diagnosis before a therapeutic decision is reached. This case and review of the literature suggest that successful treatment of focal XPN can be achieved with antibiotic therapy alone. Therefore, a trial of antibiotic therapy is warranted and unnecessary nephrectomy may be avoided.

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