Bilateral Xanthogranulomatous Pyelonephritis

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Xanthogranulomatous pyelonephritis is an uncommon form of chronic bacterial pyelonephritis characterized by the destruction of renal parenchyma and the presence of granulomas, abscesses, and collections of lipid-laden macrophages (foam cells) replacing the renal parenchyma. This case report illustrates the clinical course of bilateral diffuse xanthogranulomatous pyelonephritis with a subtle manifestation in contrast to those typically presenting with fever, flank pain or urinary tract infection. The patient therefore received supportive treatment for 18 months without hemodialysis, instead of the curative treatment, bilateral nephrectomy, which would have caused immediate loss of residual renal function and dependence on hemodialysis. [*J Chin Med Assoc* 2008;71(6):310–314]

Key Words: nephrectomy, pyrexia, urinary tract infection, xanthogranulomatous pyelonephritis

Introduction

Xanthogranulomatous pyelonephritis (XGP) is an uncommon form of chronic bacterial pyelonephritis characterized by the destruction of renal parenchyma and its replacement with granulomas, abscesses, and collections of lipid-laden macrophages (foam cells).[1] There are 1.4 cases per 100,000 population per year.[2] Most of them are unilaterally involved. Bilateral XGP is extremely rare. Poorly functioning kidney is always the consequence, and the outcome is grave for patients with bilateral XGP. Nephrectomy is deemed as curative treatment. With regard to the immediate loss of residual renal function and the relevant surgical risks, whether or not to undergo bilateral nephrectomy is a dilemma for these patients. Therefore, looking for a parameter by which to make the decision of nephrectomy becomes important. This case and previous reports show that nephrectomy might not be necessary for patients with bilateral XGP without pyrexia.

Case Report

A 47-year-old woman presented with malaise of 1 month's duration. She denied any history of dysuria, urinary frequency, fever, chills, weight loss, abdominal or flank pain, urinary calculi and urinary tract infection, except hypertension in the past 10 years. There was no significant family history. On examination, she was afebrile and mildly obese (body height, 154 cm; body weight, 76 kg), with a blood pressure of 134/82 mmHg. Pale conjunctivae were revealed. No palpable mass or tenderness was noted in the abdomen and flank. Complete blood count of peripheral blood showed normocytic anemia (hemoglobin, 8.7 g/dL; hematocrit, 26.2%) with normal white blood cell (WBC) count (8,000/mm^3) and platelet count (342,000/mm^3). Blood biochemistry revealed elevated blood urea nitrogen (102 mg/dL), elevated serum creatinine (11.1 mg/dL) and normal C-reactive protein level (0.5 mg/dL). Urinalysis indicated WBC 11–20/high-powered field (normal, 0–5), and urine culture yielded *Proteus mirabilis* (6,000 colonies/mL). Urine acid-fast stain, culture and polymerase chain reaction for tuberculosis were negative. Urine cytology was negative for 3 studies. Plain film of the abdomen revealed irregularly shaped faint calcification at bilateral upper abdomen. Renal ultrasound showed right kidney 13.4 cm and left kidney 10.9 cm in size, with bilateral renal pelvic stones and multiple cystic areas in renal parenchyma (Figure 1A). Post-gadolinium enhanced T1-weighted

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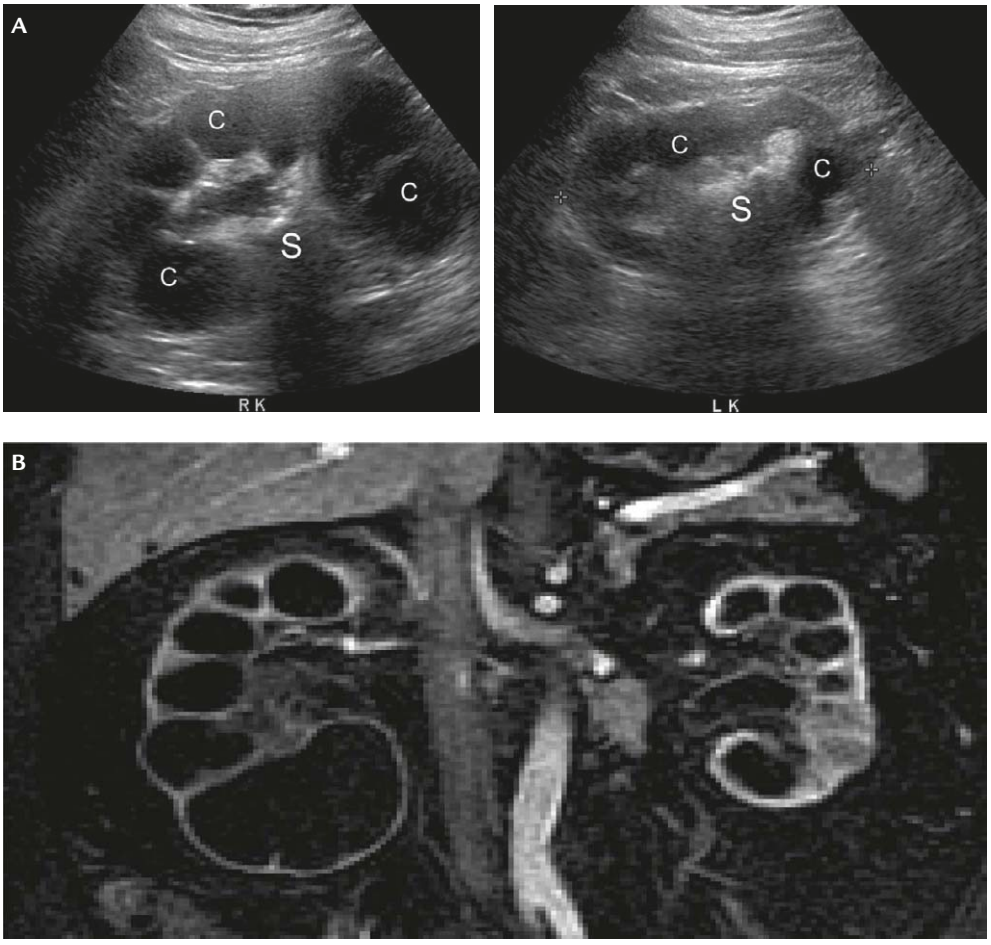

Figure 1. (A) Sonography shows bilateral renal pelvic stones (S) and multiple cystic areas (C) in the renal parenchyma. (B) Post-gadolinium-enhanced T1-weighted coronal magnetic resonance imaging also demonstrated the cystic areas in the renal parenchyma.

coronal magnetic resonance imaging (MRI) also demonstrated cystic areas in the renal parenchyma (Figure 1B) without malignant or metastatic lesions. The diffuse infiltration of mononuclear cells, histiocytes, plasma cells and focal aggregation of foamy macrophages in the renal biopsy tissue (Figure 2) supported the diagnosis of bilateral XGP.

The patient was discharged after arteriovenous fistula creation. The symptom of malaise improved after correction of anemia (hematocrit, from 26.2% to 28.2%) using erythropoietin therapy. The patient has sustained relatively stable renal function (serum creatinine, 10.6 mg/dL currently) and uneventful life without renal replacement therapy in the past 18 months of follow-up.

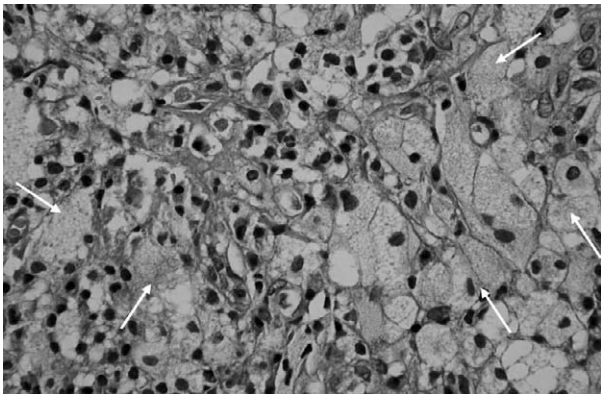

Figure 2. Histopathology of right kidney biopsy shows diffuse interstitial infiltration of mononuclear cells, histiocytes and plasma cells and focal aggregation of foam cells (arrows) (hematoxylin & eosin, 400×).

Discussion

Bilateral XGP is extremely rare. Middle-aged women are the most frequently affected.[3] Reviewing the literature, only 9 cases of bilateral diffuse XGP and 5 cases of bilateral focal XGP have been reported in the last 40 years (Table 1).[4–17] Several etiologic factors have been proposed, and genitourinary tract obstruction due to