



Xanthogranulomatous Pyelonephritis Complicated by Gram-Negative Sepsis – Case Report

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Abstract

This clinical report presents a 45-year-old female patient with acute kidney injury, hepatic dysfunction and urosepsis without past history of any illness related to urinary system. Laboratory investigations showed Hb 15.6 gm/dl, neutrophilic leukocytosis, thrombocytes 9000/mm³, ESR 90 mm/h, plasma thromboplastin time (PTT) 48 seconds (control 32 seconds), INR 2, and fibrinogen degradation product 7,500 ng/ml (normal < 250 ng/ml). Serum creatinine was 450 µmol/L (5.1 mg/dl), direct bilirubin 68.40 µmol/L, alanine aminotransferase 1.6 mmol/L and albumin 25 gm/L. Urinalysis revealed leucocyturia, hematuria without casts and proteinuria. Abdominal plain x-ray revealed left side radio-opaque shadow at the ureteric line, left uretero-hydronephrosis with thickened cortex and left ureteric stone, but normal right kidney. Computerized tomography of abdomen showed a large left kidney (13.5 cm) with totally distorted architecture, multiple enhancing and non-enhancing sectors, thickening of renal capsule, moderate dilatation of renal pelvis, dilated ureter and no passage of contrast in the ureter. The patient was treated with a 2-week course of Ceftriaxone and Aztreonam and pre-operative session of hemodialysis before the performance of a left-sided nephrectomy. Gross examination revealed adherent capsule, granular cortex covered with purulent exudates, no demarcation between cortico-medullary junction, and the cut surface showed minute abscess and necrosis with markedly dilated pelvis. Light microscopic examination showed fibrosis and inflammation of parenchyma with vacuolized histiocytes foam cells surrounding necrotic areas confirming the diagnosis of xanthogranulomatous pyelonephritis. The patient was discharged after 35 days of hospital stay with normal renal and hepatic function.

Keywords: Acute renal failure; Hematuria; Proteinuria; Hydronephrosis; Pyelonephritis; Xanthogranulomatous; Nephrectomy

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Case Presentation

A previously healthy 45-year-old housewife was admitted to hospital with left loin pain and increased frequency of micturition of 4 days' duration. The pain was colicky in nature with radiation to the lower abdomen. There was no associated vomiting, fever, alteration in the urine volume or color. There was no weight loss, anorexia or progressive fatigue. There was no history of recurrent urinary tract infections, renal stone disease, blood transfusion, diabetes mellitus, hypertension or analgesic abuse. Physical examination showed that she was jaundiced, dehydrated, but there was no pallor, leg edema, and skin rash of any type or lymphadenopathy. The lying blood pressure was 120/80 mmHg and sitting 105/78 mmHg with pulse rate of 100 beats per minute and oral temperature of 36.8°C. The abdominal examination showed a soft and lax abdomen with left lumbar ballotable tender mass of ill defined margins. There was no ascitis, hepatosplenomegaly or bruit. Physical examination of the head and neck, heart, lungs, and extremities did not show any abnormalities.

Laboratory tests showed hemoglobin 15.6 gm/dl, hematocrit 40%, with normal cell indices, neutrophilic leukocytosis with shift to the left, thrombocytes 9000/mm³, ESR 90 mm/h, plasma thromboplastin time (PTT) 48 seconds (control 32 seconds). INR 2, fibrinogen degradation product (FDP) 7,500 ng/ml (normal < 250 ng/ml). Serum creatinine was 450 µmol/L (5.1 mg/dl), direct bilirubin 68.40 µmol/L, alanine aminotransferase 1.6 mmol/L and albumin 25 gm/L. Arterial blood gases (ABG) showed PH 7.397, PCO₂ 25 mmHg, PO₂ 67 mmHg, HCO₃ 16 mmol/L, Saturation 93.6%. Urinalysis revealed leucocyturia, hematuria without casts and proteinuria. The differential diagnosis, with these results, included infections (pyelonephritis and sepsis), hepatorenal syndrome, malignancy (renal cell carcinoma).

Further evaluation by abdominal plain x-ray revealed left side radio-opaque shadow at the ureteric line, left uretero-hydronephrosis with thickened cortex and left ureteric stone, but normal

right kidney (abdominal ultrasound). Computerized tomography of abdomen showed a large left kidney (13.5 cm) with totally distorted architecture, multiple enhancing and non-enhancing sectors, thickening of renal capsule, moderate dilatation of renal pelvis, dilated ureter and no passage of contrast in the ureter. The urine and blood culture showed *Escherichia Coli* sensitive to Ceftriaxone and Aztreonam. Serum antinuclear antibodies, paraprotein and tuberculin skin test were all negative.

In view of the computerized tomography findings and other investigations a diagnosis of left xanthogranulomatous pyelonephritis with ureteric stone, urosepsis, nephrogenic hepatic dysfunction with dissemination intravascular coagulation (DIC) was made but renal cell carcinoma could not be excluded at this stage.

After a 2-week course of Ceftriaxone and Aztreonam and pre operative session of haemodialysis, a left- sided nephrectomy was performed. This resulted in normalization of hepatic function and right renal function within seven days. The antibiotics were continued for a total of 4 weeks. The blood and urine cultures repeated 48 hours after the cessation of antibiotics did not reveal any growth. The left kidney was 250 gm in weight, 14 x 5 x 6 cm in size, adherent capsule, granular cortex covered with purulent exudates, no demarcation between cortico-medullary junction and the cut surface showed minute abscess and necrosis with markedly dilated pelvis.

The light microscopic examination showed fibrosis and inflammation of parenchyma with vacuolized histiocytes foam cells surrounding necrotic areas confirming the diagnosis of xanthogranulomatous pyelonephritis. There was no evidence of malignancy.

Eighteen days after nephrectomy, she complained of dull pain in the left loin without any radiation or fever. The ultrasound of abdomen showed fluid collection at the left kidney space which was drained. It was serosanguinous in character and the culture yielded methicillin resistant staphylococcus aureus (MRSA) for which she received Rifampicin and Clindamycin.

She was discharged after 35 days of hospital stay with normal renal and hepatic function, coagulogram, ESR and blood cell count. Thus the final diagnosis was left xanthogranulomatous pyelonephritis with left ureteric stone, gram negative urosepsis complicated by acute kidney injury, nephrogenic hepatic dysfunction, DIC and MRSA infection of the retroperitoneal space.

Discussion

Xanthogranulomatous pyelonephritis is a rare form of chronic pyelonephritis which is usually caused by calculous obstructive uropathy [1,2], as in this reported case. The disease is characterized by renal destruction and replacement of renal parenchyma by foam cells (xanthoma cells) i.e. lipid laden macrophages that was first described by Schlagenhauer in 1916 [3,4]. It usually occurs in middle aged woman with a history of recurrent urinary tract infections [5], which was lacking in this patient leading to diagnostic difficulty. The typical presenting symptoms are flank pain, as in this case, and fever. However, the initial investigation at presentation of this patient showed that this case was complicated by acute kidney injury, hepatic dysfunction, DIC and urosepsis. The organism isolated was *Escherichia Coli*, a gram negative organism, from the urine and blood, though in one-fourth of cases urine culture may be sterile [6]. The initial findings of left ureterohydronephrosis on a renal ultrasound and the totally distorted architecture with multiple non-

enhancing sectors on computed tomography raised the possibilities of isolated xanthogranulomatous pyelonephritis, renal cell carcinoma or their coexistence [7-10]. The treatment plan was to cover the patient with a course of antibiotics and to manage conservatively the renal failure (including a preoperative sessions of hemodialysis). This was followed by left total nephrectomy as recommended [11-13]. The rapid recovery after the nephrectomy showed that the xanthogranulomatous pyelonephritis was responsible for most of the uncommon features encountered in this case. Xanthogranulomatous pyelonephritis has a good prognosis if it is unilateral, as in this case, or with a localized form (usually children) against bilateral disease [14].

Conclusion

In conclusion, this case of xanthogranulomatous pyelonephritis, without prior history of any urinary symptoms, was associated with multiple organ involvement that led to diagnostic and treatment challenges. Thus, the diagnosis of xanthogranulomatous pyelonephritis should be kept in mind when a middle aged female patient presents with unilateral nonfunctioning hydronephrotic kidney which is totally distorted and has enhancing as well as non-enhancing regions on computed tomography.

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