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Spontaneous Pyeloduodenal Fistula Complicating a Xanthogranulomatous Pyelonephritis: A Case Report

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ABSTRACT

A 57-year-old female presented with recurrent episodes of right flank pain and fever for the past several years. No pathogens were detected in the urine culture. Intravenous pyelography and computed tomography revealed a poorly functioning right kidney with an upper pole cystic tumor. The right retrograde pyelography showed communication between the renal pelvis and the duodenum. A nephrectomy and fistula repair were performed. Histological examination revealed chronic pyelonephritis with xanthogranulomatous reaction and a cystic renal tumor. The patient remains well at 7-year follow-up.

INTRODUCTION

Xanthogranulomatous pyelonephritis (XPG) is a severe chronic infection of the renal parenchyma [1]. Evolution toward diffuse renal destruction is usually observed. Formation of fistulas between the kidney and adjacent organs or other structures is another characteristic feature [1]. The present report is a case of spontaneous pyeloduodenal fistula complicating a renal cystic nephroma secondary to XGP.

CASE REPORT

A 57-year-old female was admitted with a 1-year history of epigastric and right flank pain, hematuria, and 5 days of fever. There was no history of trauma to this area and the patient expressed no nausea, vomiting, or lower urinary symptoms (especially pneumaturia).

Evaluation

Abdominal examinations were unremarkable except for mild pain in the right lumbar area. There was no palpable mass. No pathogens were detected in the urine culture, with the exception of hyperleucocyturia. The creatinine level was normal.

An abdominal X-ray was normal. A kidney, ureter, bladder (KUB) film revealed a calcified mass in the right kidney area (Figure 1). Intravenous pyelography (IVP) confirmed the diagnosis and showed a poorly functioning right kidney. The left kidney was normal.

Gastroscopy was performed up to the second portion of the duodenum. It revealed only a gastric ulceration.

Ultrasonography showed a heterogenous renal mass. Abdominal computed tomography (CT) revealed a calcified and heterogeneous renal mass with adherent duodenum and localized air, mimicking emphysematous pyelonephritis (Figure 2). Barium meal showed a passage of the contrast medium within the renal pelvis, which confirmed the presence of a pyeloduodenal fistula.

Surgical Management

Surgical exploration through a right lumbar incision confirmed the presence of a pyeloduodenal fistula that was located between the renal pelvis and the second portion of the duodenum. An invasive inflammatory process spreading from the left kidney to adjacent structures was found. The patient underwent a simple nephrectomy with reparation of the duodenal defect.

KEYWORDS: Urodigestive fistulas; Urinary tract infection; Nephrectomy; xanthogranulomatous pyelonephritis

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Abbreviations and Acronyms

CT = computed tomography
XPG = xanthogranulomatous pyelonephritis

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Figure 1. Kidney, Ureter, Bladder X-Ray Showing Calcified Mass of the Right Kidney (arrows).

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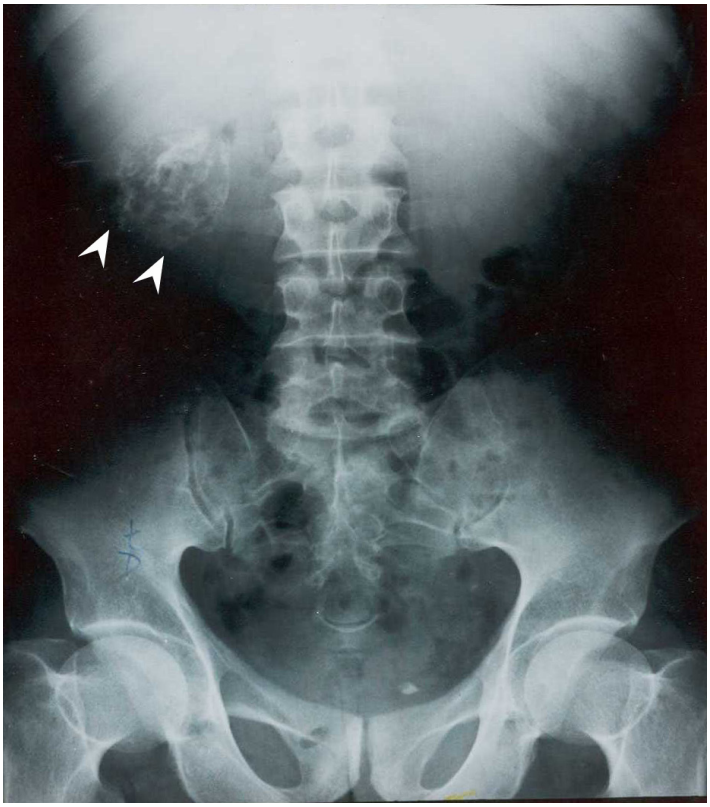

Figure 2. Enhanced Computed Tomography Scan Demonstrating a Fluid-Filled Cavity in the Upper Pole of the Right Kidney (arrows).

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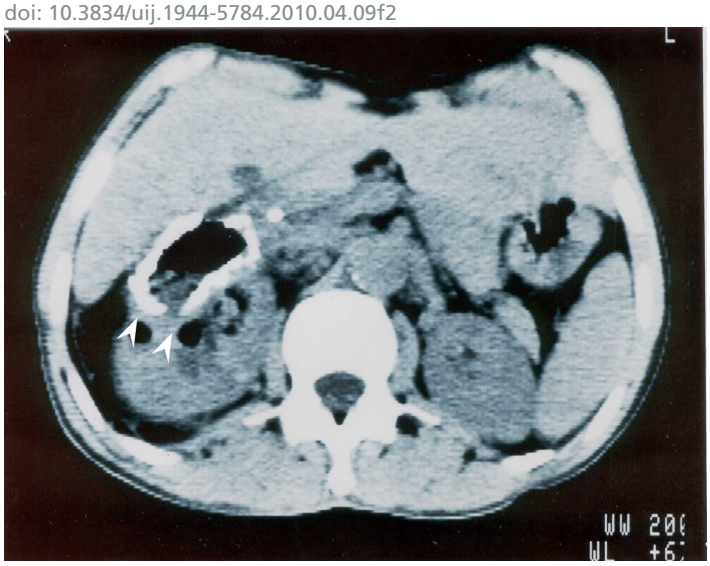

Initially, the Kocher maneuver was performed by dividing the lateral peritoneal attachment of the duodenum and mobilizing both the second and third parts. The surgeons performed a debridement of the margins of the duodenal defect and the margins were sutured together without tension. The suture was oriented transversely to avoid luminal compromise.

Postoperative Follow-up

Histological examination revealed chronic pyelonephritis with xanthogranulomatous reaction and cystic renal nephroma. The postoperative course was uneventful. Fluoroquinolone (400 mg/day) was given for a period of 6 weeks.

A CT scan taken 2 months later was normal. The patient remains well at 7-year follow-up.

DISCUSSION

XGP is an uncommon inflammatory disease process characterized by multiple fluid-filled cavities that replace the renal parenchyma. The cavities are lined with lipid-laden macrophages (xanthoma cells) that act as a barrier to the pus and debris that make up its contents [2]. It evolves with localized infection, renal destruction, and systemic impairment [3].

The exact etiology of XGP is not known, but a combination of chronic urinary obstruction, chronic presence of stones in the excretory system, and infection plays a significant role and is present in most cases [4]. As in the present case, XGP most commonly affects women in their fifth and sixth decades and is usually unilateral.

Symptoms of XGP are present for a period of up to 6 months in 70% of the patients [2]. Findings include flank pain, palpable mass, fever, and less commonly, weight loss, abdominal pain, urinary symptoms, anorexia, and malaise. Flank tenderness is present in less than 25% of the patients [2].

On laboratory examination, anemia is a common finding. Leukocytosis is present in 50-75% of the cases. An elevation in alkaline phosphatase and liver enzymes is seen in less than half of the patients [5]. Urinalysis is significant for pyuria in 90% of the cases. Urine cultures are positive in 50-80% of the patients, with Gram-negative organisms (*P. mirabilis and E. coli*) being the most common pathogens.

The CT scan has been advocated as the modality of choice for evaluation. It can accurately diagnose XGP in 90% of patients before surgery [5]. It also helps to determine the extent of

the disease and guides treatment planning. CT scan findings include a large calculus in the renal collecting system, which is present in 75% of the cases. Half of the stones are staghorn calculi [4-6]. Other typical CT scan features include an enlarged kidney that fails to excrete contrast and an enhancing rim of tissue surrounding low-density fluid-filled spaces [4]. Extra-renal involvement (eg, perinephric, psoas, bowel, diaphragm, posterior abdominal wall) is common. Perinephric fat, termed *replacement lipomatosis*, is less common [6].

Fistula formation between the upper urinary tract and surrounding organs is a very uncommon complication of PXG [7]. There are reports of colonic, gastric, and jejunal fistulas [3,8,9]. There are also reports of fistulas to bronchial tissues and skin [7], psoas muscle, and the flank and gluteal regions [3]. Pyeloduodenal fistulae are rare and usually secondary to trauma or duodenal perforation by ingestion of a foreign body [10]. A spontaneous pyeloduodenal fistula is extremely rare. Most of the etiologies belong to chronic renal inflammatory disease, such as XGP.

CONCLUSION

Fistulization to surrounding organs is a very uncommon complication of renal XGP. It should be suspected when air is present in the upper urinary tract. Treatment is determined on a case-by-case basis but always includes surgery.

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