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CASE REPORT

Spontaneous Pyeloduodenal Fistula Complicating a Xanthogranulomatous Pyelonephritis

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Abstract
A 57-year-old female presented with recurrent episodes of right flank pain and fever. No pathogens were detected in urine cultures. Intravenous pyelography and computerized tomography revealed a poor-functioning right kidney with an upper pole cystic tumor. Barium meal showed a communication between the renal pelvis and the duodenum. A right nephrectomy was performed with repair of the fistula. Histological examination revealed chronic pyelonephritis with xanthogranulomatous reaction and cystic renal tumor. The patient remains well at seven year follow-up.

Key-Words: Duodenum, renal pelvis, fistula, nephrectomy, xanthogranulomatous pyelonephritis.

Introduction

Xanthogranulomatous pyelonephritis (XPG) is a severe chronic infection of renal parenchyma (1). Evolution towards diffuse renal destruction is usually observed. Formation of fistulas between kidney and adjacent organs or structures is another characteristic feature (1). We report, herein, a case of spontaneous pyeloduodenal fistula complicating a renal cystic nephroma secondary to XGP. To the best of our knowledge, this is the second report of this type of pyeloduodenal fistula.

Case Report

A 57 year-old female presented with history of epigastric and right flank pain of one year duration. She also had a history of hematuria and fever for five days prior to exam. There was no history of lumbar or abdominal trauma. The patient felt no nausea, vomiting, or lower urinary symptoms (especially pneumaturia). Abdominal examination was unremarkable, except for mild tenderness in the right lumbar area. No pathogens were detected in the urine culture but there was hyperleucocyturia. Serum creatinine level was normal. Plain chest and abdominal X-ray were normal.

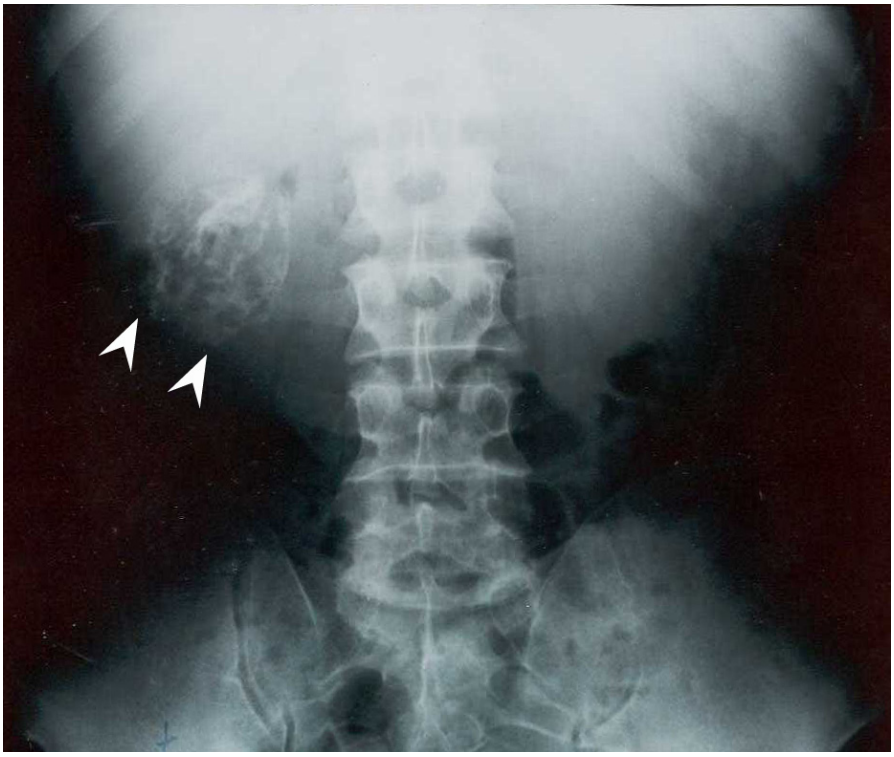
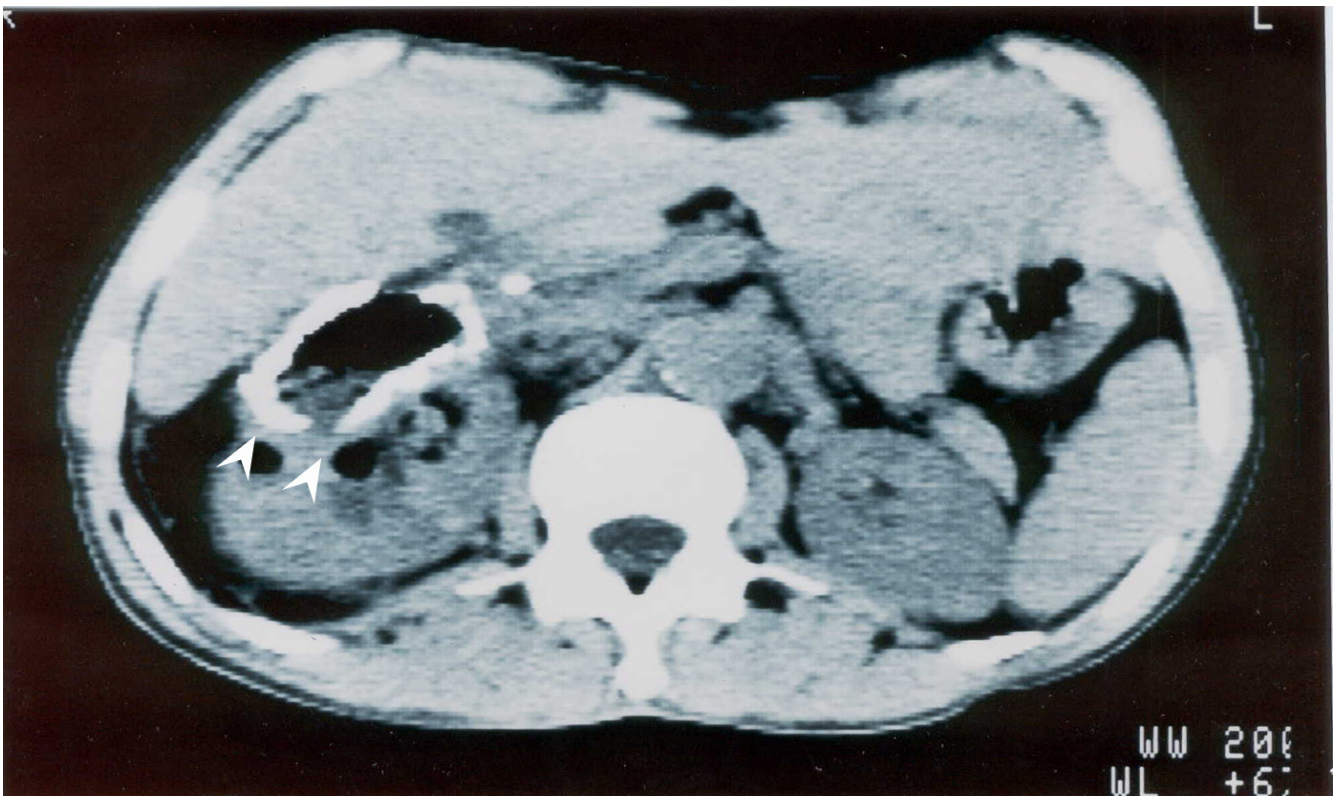
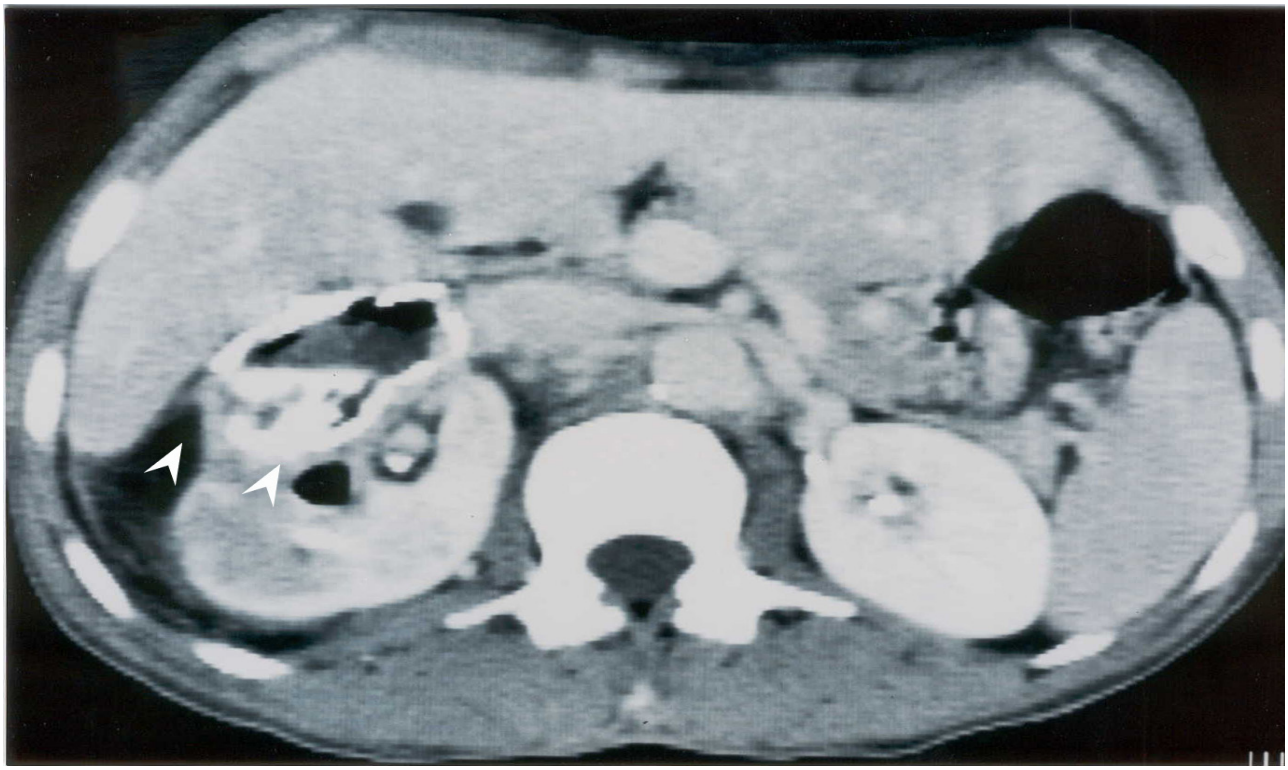


Figure 1: KUB film: Calcified mass of the right kidne





Figures. 2, 3: Enhanced CT scan demonstrating a fluid filled calcified cavity in the upper pole of the right kidney.

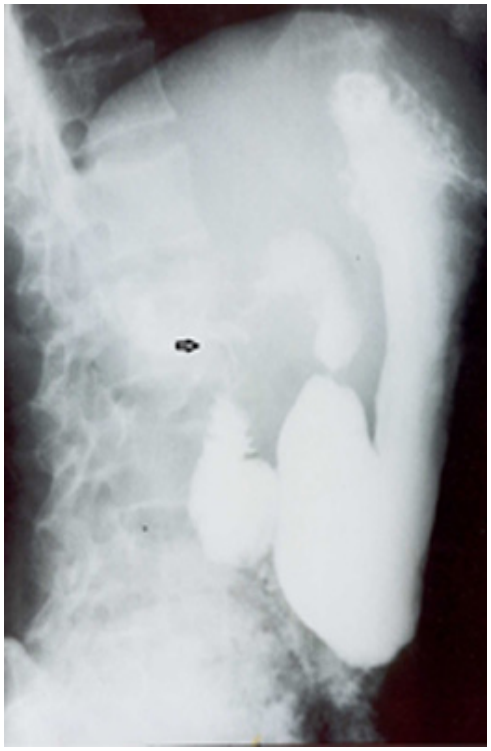


Fig. 4: Barium meal showed a passage of the contrast medium within the renal pelvis: pyeloduodenal fistula (arrow).

KUB film revealed a calcified mass in the right kidney area (Fig.1). Intravenous pyelography (IVP) confirmed the diagnosis and showed a poor-functioning right kidney. The left kidney was normal. Esophagogastroduodenoscopy revealed a gastric ulceration and a stricture of the second part of the duodenum. Ultrasonography revealed a heterogeneous renal mass. Abdominal computed tomography showed a calcified and heterogeneous renal mass adherent to the duodenum, containing air and mimicking an emphysematous pyelonephritis (Fig. 2,3). Barium meal revealed passage of the contrast medium into the renal pelvis confirming the presence of a pyeloduodenal fistula (Fig. 4). Three diagnostic hypotheses were suspected preoperatively: a calcified renal hydatid cyst, pseudo-tumoral renal tuberculosis (especially since our geographical region remains an endemic zone), and an atypical renal cancer. The pyeloduodenal fistula could be a complication of the lesion due to local extension. An exploration performed through a right lumbar incision confirmed the pyeloduodenal fistula between the renal pelvis and the 2nd part of the duodenum. An invasive inflammatory process spreading from the left kidney to adjacent structures was found. Right nephrectomy was performed and the duodenal defect was repaired. The procedure was laborious, taking 190 min. Perioperative bleeding was about 300 ml but no blood transfusion was required. Histological examination revealed chronic pyelonephritis with xanthogranulomatous reaction and cystic renal nephroma. Histological study of the fistulous tract and duodenum didn't reveal any evidence of malignancy. The enteral tube was maintained for seven days, after which a progressive oral diet was reintroduced. Postoperative course was uneventful. A CT scan at two months post-op showed no complications. The patient remains well at seven-year follow-up.

Discussion

XGP is an uncommon inflammatory disease process characterized by multiple fluid-filled cavities that replace the renal parenchyma (1). It evolves with localized infection, renal destruction, and systemic impairment (2). The exact etiology of the condition is unknown, but a combination of chronic urinary obstruction, chronic presence of stones in the excretory system, and infection all play a significant role and is present in most cases (4). As in this case, XGP most commonly affects women in their fifth and sixth decades and is usually unilateral. Clinical presentation includes flank pain, fever, palpable mass, and less commonly, weight loss, abdominal pain, urinary symptoms, anorexia, and malaise. Flank tenderness is present in less than a quarter of patients (1). On laboratory examination, anemia is a common finding. Leukocytosis is present in 50–75% of cases of XGP. An elevation in alkaline phosphatase and liver enzymes is seen in less than half of patients. Urinalysis is significant for pyuria in 90% of cases. Urine cultures are positive in 50–80% of patients, with Gram-negative organisms (P. mirabilis and E. coli) being the most common pathogens (1). CT scan has been advocated as the imaging modality of choice, not only for its ability to diagnose, but also for determining the extent of disease and for planning treatment. CT scan findings include a large calculus in the renal collecting system, present in 75% of all cases. Half of the stones are staghorn calculi (3). 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 (3). Extra-renal involvement (perinephric, psoas, bowel, diaphragm, posterior abdominal wall) is common (1). Fistula formation between the upper urinary tract and surrounding organs is a very uncommon complication of PXG (4). There are few reports of colonic, gastric, jejunal fistulas (2,5,6) and also fistulas to bronchial (4), to skin (4), to psoas muscle, flank, and gluteal regions (2). Pyeloduodenal fistulae are very rare and usually secondary to trauma or duodenal perforation by ingestion of a foreign body (7). Spontaneous pyeloduodenal fistula is extremely rare. Most of the etiologies belong to chronic renal inflammatory disease such as XGP. Only one case of pyeloduodenal fistula resulting from XGP has been previously reported (7). This case is the second reported case. The pathogenesis of this fistula can be explained by the inflammation surrounding the kidney that is the origin of a pyeloduodenal adhesive contact. The infectious phenomenon plays a great role in the erosion and perforation of the urinary wall and the digestive tract. Rodney, et al, through a series of 28 cases of pyeloduodenal fistula, suggest that the best management is nephrectomy and primary closure of the duodenum (8). Most authors maintain these rules and some reported treatment by laparoscopy (9) with excellent follow-up. Recently, Atalla, et al (10), reported a successful conservative treatment in a 50 year-old man with spontaneous pyeloduodenal fistula. Treatment consists of duodenum and renal pelvis repair and an interposition of an omental flap. Treatment in such entity should be considered on an individual basis as it depends on the underlying condition and the degree of renal damage.

In conclusion, the development of fistulae to surrounding organs is a very uncommon complication of renal XGP. It

should be suspected when air is present in the upper urinary tract. The modality of the treatment should be case by case, and almost always is surgical.

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