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GENERAL UROLOLOGY
Case Report

A case of Xanthogranulomatous pyelonephritis with spontaneous renocolic fistula

Spontan renokolik fistül ile birlikte bir Ksantogranulomatöz piyelonefrit vakası

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ABSTRACT

Xanthogranulomatous pyelonephritis (XGPN) is a rare clinicopathological syndrome that is unique among the various inflammatory conditions of the kidney, and it closely mimics renal cell carcinoma, both clinically and radiologically. Approximately one third of XGPN cases have associated complications, such as abscess and fistulas, although the latter is much less common. Spontaneous renocolic fistulas of non-tubercular origin are also rare, especially in Asia. Fistula or deep sinus formation as a complication of XGPN is a rare clinical condition. Currently, only approximately 10 such cases (including our case) have been reported in the literature. We present one such case of spontaneous nephrocolic fistula complicating XGPN. Ultrasonography, an intravenous urogram, retrograde pyelogram, and computerized tomography aided in diagnosing the presence of a renocolic fistula. The treatment regimen of total nephrectomy with primary closure of the rent in the colon was adequate.

Key words: Nephrectomy; nephroenteric fistula; nephrocolic fistula; renocolic fistula; xanthogranulomatous pyelonephritis.

ÖZET

Ksantogranulomatöz piyelonefrit (KGPN) böbreğin çeşitli inflamatuar durumları arasında benzersiz olan nadir bir klinikopatolojik sendromdur ve renal hücre karsinomunu, hem klinik hem de radyolojik olarak yakından taklit eder. KGPN vakalarının yaklaşık üçte biri, apse ve fistül gibi (ikincisi daha az yaygın olmakla beraber) komplikasyonlarla ilişkilidir. Spontan, tuberkül-dışı kökenli Renokolik fistüller de; özellikle Asya'da çok nadirdir. KGPN komplikasyonu olarak fistül veya derin sinüs oluşumu çok nadir bir klinik durumdur. Dünya literatüründe şimdiye kadar bu tür sadece 10 civarı vaka (bu vaka dahil) bildirilmiştir. Biz böyle bir KGPN'yi komplike eden spontan nefrokolik fistül vakası sunmaktayız. Ultrasonografi, İntravenöz ürogram, Retrograd piyelogram ve bilgisayarlı tomografi renokolik fistülün varlığını teşhis etmede yardımcı oldu. Total Nefrektomi ile kolondaki yırtığın primer kapatılmasını içeren tedavi rejimi yeterli idi.

Anahtar sözcükler: Ksantogranulomatöz piyelonefrit; nefrektomi; nefroenterik fistül; nefrokolik fistül; renokolik fistül.

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Introduction

Fistulas between the gastrointestinal tract and kidney are rare. Among nephroenteric fistulas, nephrocolic fistulas are most common and over 130 cases have been reported. [1] Xanthogranulomatous pyelonephritis (XGPN) is regarded as a rare condition; indeed, until 1980, only approximately 400 cases were reported in the literature.[2] XGPN is a distinct clinicopathological entity histologically characterized by suppuration and the accumulation of lipid-laden macrophages.[2] Approximately one third of XGPN cases have associated complications, such as abscess and fistulas. After taking informed consent from the patient. Here, we present a case of extensive XGPN complicated by spontaneous renocolic fistula.

Case report

A 25-year-old female was admitted with a 6-month history of abdominal pain, dyspepsia and fever. On examination, there was ten-

derness in the left lumbar region but no mass. Investigations revealed anemia. The leukocyte count and erythrocyte sedimentation rate were normal. *Escherichia coli* was cultured from the urine. She was administered antibiotics for the urinary infection. Ultrasonography (USG) suggested left renal calculi with hydronephrosis. Intravenous urography (IVU) showed a nonvisualized left kidney with calculus and a normal right kidney. A left retrograde pyelogram (RGP) showed gross upper pole caliectasis with pelvic staghorn calculus. Communication between the lower pole calyx and descending colon was demonstrated. Contrast-enhanced computerized tomography (CT) showed a left renal mass with irregular borders and an inhomogeneous density and multiple hypodense egg-shaped areas. A pelvic staghorn calculus and the fistula were also observed (Figures 1-3).

The kidney was exposed through a left flank incision. The colon was dissected from the kidney with difficulty. The fistula was identified between the lower pole of the kidney and descending colon, and it was disconnected from the colon and excised. The rent in the colon was oversewn in two layers. Nephrectomy was performed after dividing the dense adhesions present between the kidney and the surrounding structures. The large bowel mucosa showed no evidence of diverticula.

The patient had a good postoperative recovery, and on follow-up at 1, 3 and 6 months later, the patient was asymptomatic, and follow up USG/urinalysis/renal function tests were normal.

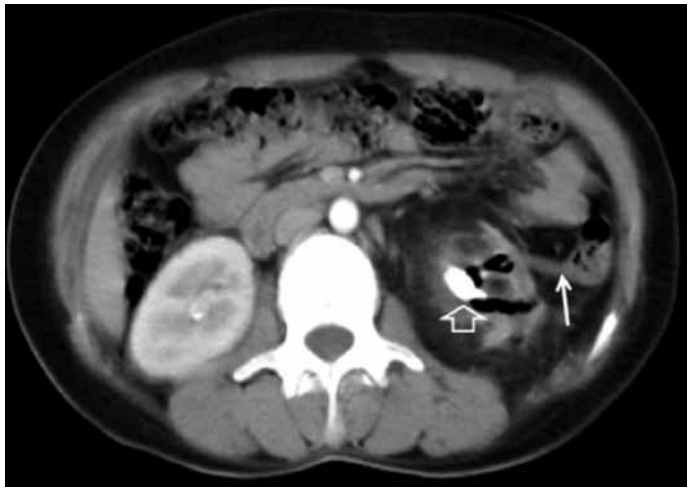

Figure 1. CT scan: Axial contrast-enhanced CT of the abdomen: The left kidney is replaced by a heterogeneously enhancing mass lesion with multiple foci of air in the pelvicalyceal system. The adjacent descending colon is mildly thickened with an abnormal tract connecting the renal pelvis and bowel. The hollow arrow shows staghorn calculi, and the thin arrow shows the communicating tract

The gross specimen showed a shrunken kidney and a distended pelvicalyceal system containing a calculus with thick pus. Yellow-orange tissue was present around many calices. The inflammatory process had extended beyond the renal capsule, and xanthogranulomatous tissue was present at the margin of surgical resection. Numerous dilated blood vessels were noted in the pericapsular area.

The fistula was lined with XGPN tissue; there was no evidence of epidermoid carcinoma. Histopathological examination of the nephrectomy specimen slides showed xanthogranulomatous tissue with lipid-laden macrophages, multinucleated giant cells and acute inflammatory cells as well as loose "xanthoma" cells; these features were suggestive of XGPN. Periodic acid-Schiff and Ziehl-Nielsen stains were used to exclude acid-alcohol fast bacilli.

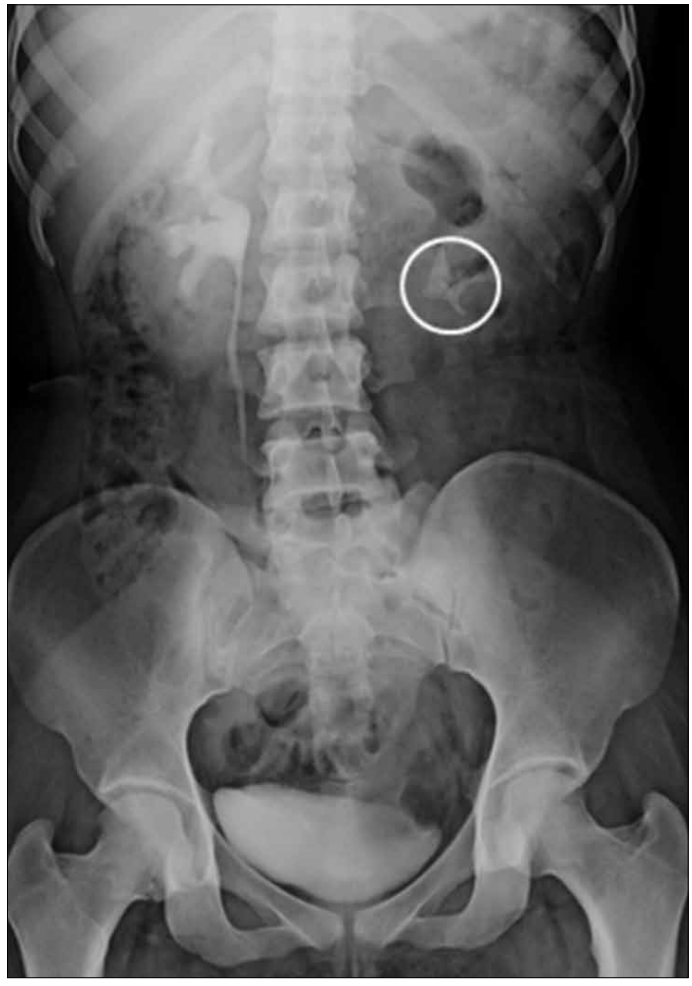

Figure 2. IVU-The circle in the figure indicates the staghorn calculus in the left kidney. There was no evidence of contrast excretion from the left kidney, and there was gas in the left renal region

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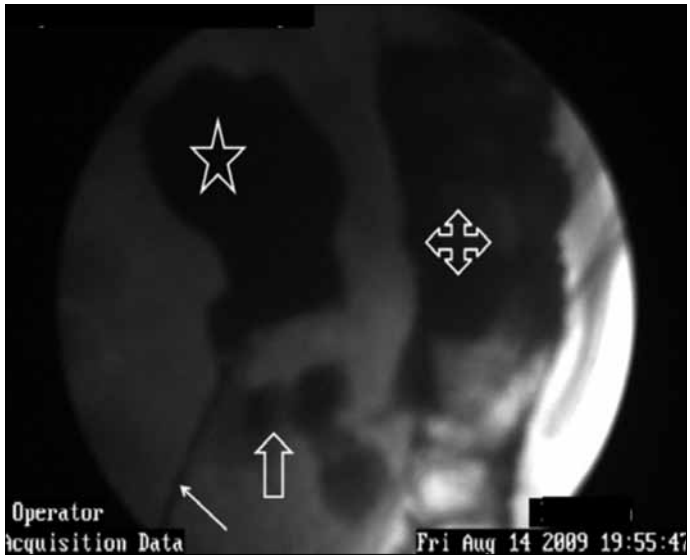

Figure 3. RGP: The dilated left pelvicalyceal system and a tract communicating with the left pelvicalyceal system and descending colon. The star shows gas contrast in the upper calyx. The plus mark shows contrast in the descending colon. The hollow arrow shows the tract communicating with the lower calyx and colon. The thin arrow shows the ureter catheter

Discussion

Renocolic fistulas were first described by Hippocrates in 460 B.C. The ascending and descending colon are the most frequently affected.[1] These fistulas are most often found in younger and middle-aged adults at equal frequency in both sexes.[2]

Parsons et al.[3] reported a case of XGPN with a renocolic fistula. Since then, there have been few individual reports in the literature of fistula formation in XGPN.

The etiology of renocolic fistulas can be traumatic or spontaneous. Traumatic cases are the minority and are usually iatrogenic, following open or percutaneous surgical procedures, although they can follow severe renal trauma (penetrating and blunt). Spontaneous renocolic fistulas almost always arise as a consequence of primary renal pathology (chronic renal diseases). In the majority of early reports, this pathology was tuberculosis, but more recently, calculous pyonephrosis has been the predominant underlying cause. Other known causes are non-calculous pyonephrosis, perinephric abscess, the rupture of hydronephrosis, hydatid cysts, renal malignancy and papillary necrosis.[1,4-6] There are cases of epidermoid carcinoma arising in a chronic renocolic fistula.[3] A colonic origin has only been reported three times, and in two cases, there was pre-existing renal disease.[1,3]

Anatomically, the posterior wall of the left colon, which is devoid of serosa, is directly opposite the anterior surface of the left kidney. The basic pathology is a chronic inflammatory process (XGPN in our case), which first begins in the renal parenchyma.[4] This permits the slow adherence of the kidney to the colon, which provides for subsequent perforation and the drainage of infected urine and necrotic material, resulting in a chronic renocolic fistula. However, in some cases, pus may form and tract down to open exteriorly, resulting in a cutaneous renocolic fistula.[4] The clinical features are classically those of an acute suppurative illness, most likely associated with the formation of a perinephric abscess. Alternatively, the course may be more insidious, with progressive weight loss and malaise. Pneumaturia can be a presenting symptom in some cases. The clinical signs are rarely pathognomonic. In long-standing cases with persistent renal sepsis, there may be evidence of wasting, dehydration, anemia and uremia. Loin tenderness and a renal mass are occasionally present, and in 10% of cases, there is a cutaneous opening to the fistula.

Investigations are invariably non-specific, and the diagnosis is nearly always made radiologically through a barium enema, RGP, antegrade pyelography, CT scan or if there is cutaneous extension, fistulogram.[2,5] The mainstay of treatment for renocolic fistulas is open surgery, with the exception of post-instrumentation fistulas, which if very small, may be treated conservatively. In the majority of cases, nephrectomy is necessary, and the affected kidney is often completely destroyed and occasionally shows malignancy. The affected bowel is resected, and where conditions permit, a primary anastomosis is performed.[4] In our case, the presence of extensive renal damage and associated changes due to XGPN encouraged us to remove the affected kidney. However, as the involved colon was macroscopically normal and the fistulous opening was very small, primary closure was performed.

The prognosis of the condition largely depends on the underlying etiology, the duration of the disease, the degree of renal insufficiency and the general condition of the patient.

The kidney in XGPN is usually adherent to adjacent structures, such as the abdominal and psoas muscles and adrenal gland, with numerous new blood vessels extending from the inflamed kidney. If the kidney is adherent to structures such as the large or small bowel or the diaphragm, then the presence of a sinus or fistula should be assumed for purposes of surgical management.[3]

The designation of XGPN as a separate entity is important because it is not only often recognizable as a clinicopathological syndrome but also unique among the inflammatory conditions

of the kidney in closely mimicking the clinical, radiological and even histological features of renal cell carcinoma.

Conflict of Interest / *Çıkar Çatışması*
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