Case Report

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Nephrocutaneous Fistula: An Unusual Presentation of Xanthogranulomatous Pyelonephritis

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ABSTRACT

Spontaneous nephrocutaneous fistula is a rare manifestation of renal diseases. The majority of such fistulas are associated with xanthogranulomatous pyelonephritis, chronic renal tuberculosis, complicated renal stones, and post-traumatic or surgical injuries. We report the case of a 58-year-old male who presented with a history of massive lumbar cutaneous purulent discharge for 3 years. More investigations with ultrasonographic study and magnetic resonance imaging demonstrated the presence of a fistula between the kidney and skin. Detailed laboratory workup was negative for the diagnosis of renal tuberculosis. This patient was considered as a rare variant of xanthogranulomatous pyelonephritis and was managed effectively with a simple nephrectomy.

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Introduction

Spontaneous nephrocutaneous fistulas (NCFs) which usually present as spontaneous drainage through lumbar region are uncommon complications of nephrolithiasis. Presentation of NCF as the primary appearance of kidney stones is extremely scarce. The majority of such fistulas are secondary to xanthogranulomatous pyelonephritis, chronic renal tuberculosis, complicated renal stones, and post-traumatic or surgical injuries. A few cases of NCFs are described in the literature and most of them are associated with chronic urinary tract infection and nephrolithiasis (1-11).

We describe another case of NCF following xanthogranulomatous pyelonephritis in the setting of renal stones.

Case Report

A 58yearold diabetic male was admitted to the urology service with the complaint of

chronic purulent discharge from his lumbar region for the past 3 years. He had no history of pulmonary or extrapulmonary tuberculosis, renal surgeries, or flank trauma in the past. Physical examination revealed a fistulous orifice in the skin of his left flank region (Figure 1).

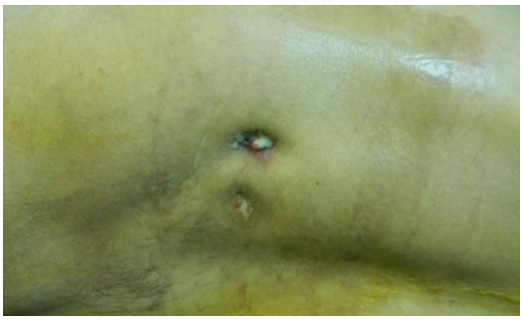

Figure 1. Nephrocutaneous fistula

In laboratory tests, the patient had mild leukocytosis, anemia, reactive thrombocytosis, pyuria, and normal renal function parameters. Urine culture showed mixed bacterial growth.

The kidney, ureter, and bladder X-ray imaging of the patient showed multiple renal stones in the left kidney (Figure 2).

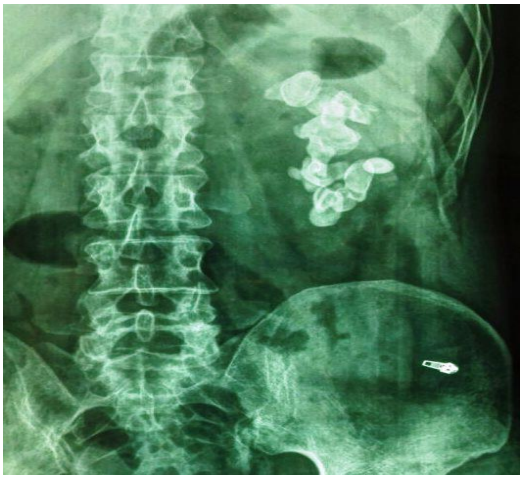

Figure 2. Multiple renal stones

Computed tomography scan with contrast injection was not performed because the renal function was slightly impaired. Magnetic resonance imaging demonstrated severe atrophy of the left renal parenchyma and the NCF (Figure 3).

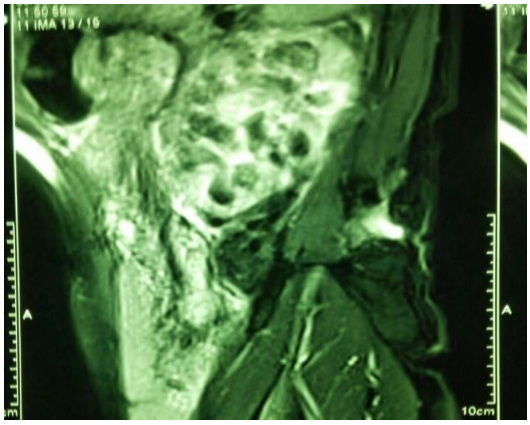

Figure 3. Magnetic resonance imaging showing nephrocutaneous fistulas

Dimercaptosuccinic acid renal scan showed poor left-side function with a differential function of 5% (Figure 4).

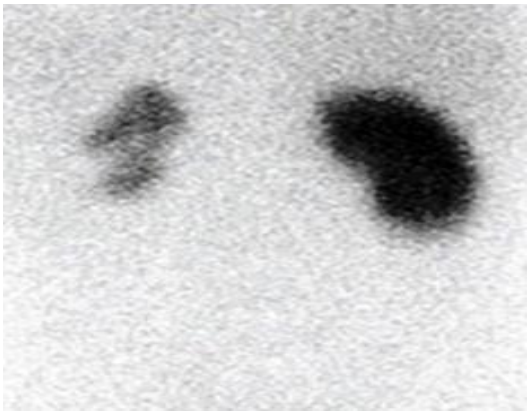

Figure 4. Dimercaptosuccinic acid renal scan

The preoperative Lowenstein culture was negative for *Mycobacterium tuberculosis*.

Based on these findings, the patient was planned for nephrectomy and excision of the sinus tract. Broad-spectrum antibiotics were administered for 4 days before surgery. In the operating room, the patient was placed in the flank position and an intercostal incision was carried out between the 11th and 12th ribs. After releasing of many dense adhesions, simple nephrectomy was done and the sinus tract was excised too. We left a corrugate drain in the renal fossa and the flank muscular layers were repaired.

The patient was discharged from the hospital on the third postoperative day. On subsequent follow-up visits in the 1st and 6th months, the

patient had no complaints of cutaneous discharge or any other related problems.

The pathological examination of the surgical specimen showed lipid-laden histiocytes with lymphocytes, giant cells, and plasma cells, suggestive of xanthogranulomatous pyelonephritis.

Renal stone analysis demonstrated approximately 75% struvite (magnesium-ammonium-phosphate) and 25% calcium-phosphate composition.

Discussion

Our literature review revealed that the spontaneous NCF is a rare condition. Likewise, NCF in the setting of renal stones is also very rare, and the most common underlying pathology of NCF is xanthogranulomatous pyelonephritis. The role of renal stones in the causation of a NCF is central, while other causes such as chronic renal tuberculosis, complicated renal stones, and post-traumatic or surgical injuries are also listed in the literature (2).

In xanthogranulomatous pyelonephritis, the vicious cycle of stasis, infection, and stone growth leads to a non-functioning kidney. The ongoing chronic inflammatory process may have central role in the formation of the NCF. NCF formation represents the late stage of the chronic inflammation when the renal tissue has been rendered non-functional by the underlying pathology. The fistulas most often drain through the lumbar region from points of the lowest resistance, such as lumbar triangle and lumbar quadrilateral (3).

Long-term complications of NCF are secondary amyloidosis, progressive destruction of the posterior abdominal wall, and invasion to the adjacent organs such as colon, stomach, major blood vessels, spleen, liver, and the pleural cavity (11).

The management of NCF needs considerations about the renal function as well as the ability of patients to tolerate the surgical procedure. Surgical removal with a timely nephrectomy results in satisfactory healing of most NCFs and prevents further morbidity.

Conflict of Interests

Authors have no conflict of interests.

Acknowledgments

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