

## CASE REPORTS

### CUTANEOUS NEPHRO-BRONCHIAL FISTULA: A CASE REPORT AND REVIEW OF THE LITERATURE

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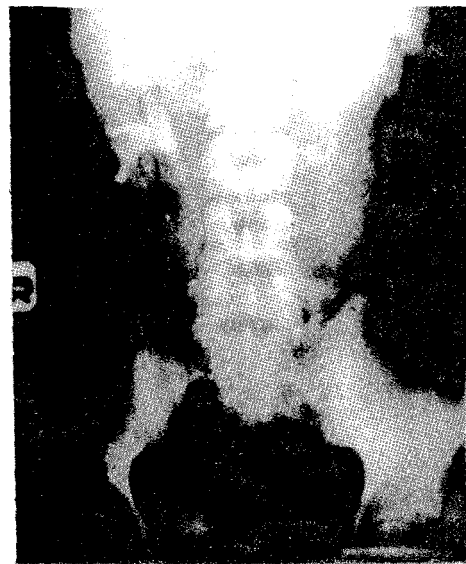
#### INTRODUCTION

Renal fistula can occur between the kidney and other adjacent organs, most commonly between the kidney and the colon. Nephro-colonic fistula is either secondary to xantho-granulomatous pyelonephritis<sup>1-2</sup> or a complicating diverticular disease communicating with a kidney harboring stones<sup>3-4</sup>. Nephro-bronchial fistula, although rare, is the second common renal fistula. So far, 73 cases have been reported in the literature. Most of these cases were reported before the antibiotics era. The majority of them were secondary to either staghorn stone or primary perinephric abscess. Renal tuberculosis was reported as a cause of fistulas between the urinary tract and the lung in 12 cases<sup>5-6</sup>. The main presenting renal symptom of nephro-bronchial fistula is renal pain. However, pulmonary symptoms including productive cough and a urinefrous taste in the mouth in non-uremic patients may mask the renal symptoms<sup>6</sup>. An abnormal chest X-ray was commonly seen in the form of pleural effusion, lower lobe infiltrate, hydropneumothorax, empyema or lung abscess<sup>7</sup>.

Herein we report on a cutaneous nephro-bronchial fistula in a 28-year-old man with non-functioning tuberculous pyelonephritis. He was successfully managed by nephrectomy. A review of literature on fistulas between the urinary tract and the lung is presented.

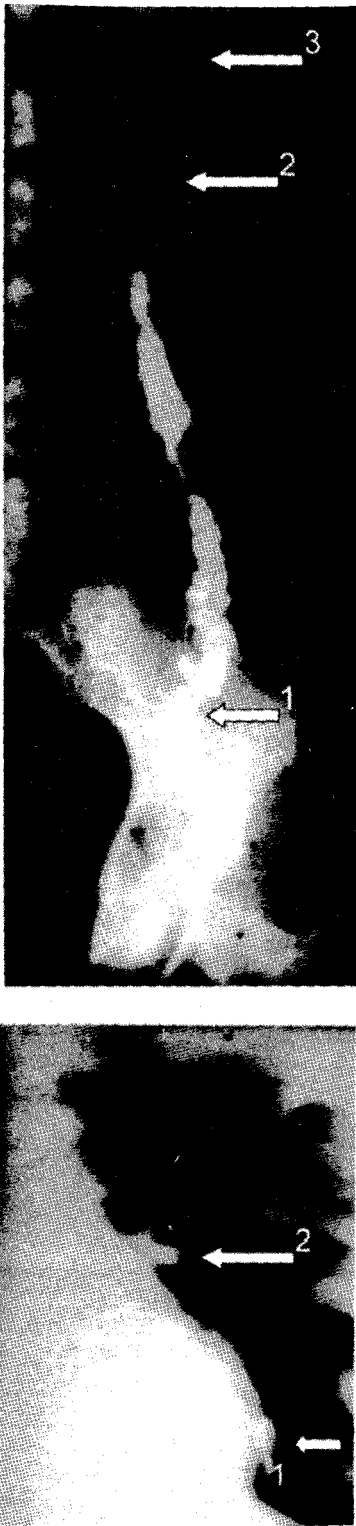
#### CASE REPORT

A 28-year-old man presented with recurrent left dull aching loin pain. Plain X-ray (KUB) showed punctuated calcifications at the region



**Fig. 1:** IVU revealing non-excretion of the contrast by the left kidney and a normal right renal unit

of the left kidney. Intravenous urography (IVU) revealed non-excretion of contrast by the left kidney and a normal right renal unit (Fig. 1). His past medical history was unremarkable. Physical examination revealed a healthy appearing man in no acute distress. Vital signs were stable. Chest and abdominal examinations were unremarkable. Urinalysis revealed no organism. Complete blood picture (CBC) showed normochromic microcytic anemia (Hb 10.4%) and mild leucocytosis. Serum creatinine was 1.3 mg/dl (normal 0.9 to 1.1). The patient was hospitalized for further evaluation



**Fig. 2: A:** Sinogram showing a communicating fistula (arrow 1) up to the posterior lower calyx of the left kidney (arrow 2) which is connected to the bronchial tree of the lower left lobe of the lung (arrow 3). **B:** Left lung showing the contrast in the bronchial tree of the lower left lobe of the lung (arrow 1) draining in the main bronchus (arrow 2)

and management. Two days after hospitalization, he developed cystic swelling at the left iliac fossa. Cyst aspiration drained a yellow slightly turbid fluid. The culture from this fluid was negative. The cyst recollected again, and a test for acid-fast bacilli was positive. Computed tomography did not identify the origin of the cyst. Secondary infection and abscess developed in the cyst which was incised and drained. The patient was subjected to antituberculous treatment in the form of isonicotinic acid hydrazide (INH), rifampicin, streptomycin, and ethambutol. The abscess continued to drain in spite of the local irrigation with rifampicin for six weeks. The sinus tract and the wall of the abscess were excised, but recurrence of the discharge started to occur after three weeks.

At that stage, a sinogram was done under fluoroscopy which showed a communicating fistula up to the lower calyx of the left kidney and connected to the bronchial tree of the lower left lobe of the lung (Fig. 2). The patient developed irritating productive cough and expectoration two days after the study. Chest X-ray showed retained contrast in the lower left lobe of the lung (Fig. 3).

Under cover of the antituberculous management, left nephrectomy was performed through an 11<sup>th</sup> rib left flank incision. There was no perinephric abscess or collection, but dense adhesions around the kidney and a thick fibrous band extending between the kidney and the diaphragm were seen. The band was ligated and cut. The retroperitoneum was irrigated and drained with J-vac drain. The kidney was small and the renal parenchyma showed multiple small abscesses full of caseous material and small stones.

Convalescence was uncomplicated. The retroperitoneal drain was removed on the 6<sup>th</sup> postoperative day. The fistula in the left iliac fossa stopped draining. The patient's renal function remained normal. He was discharged home on the 12<sup>th</sup> postoperative day. Histopathological examination revealed an 8.5 x 6.6 x 5 cm kidney with tuberculous glomerulonephritis and grade-3 nephropathy.

## DISCUSSION

Nephro-pulmonary fistulas are classified into three major types, including nephro-pleural fistulas causing empyema, nephro-



Fig. 3: Chest X-ray showing retained contrast in the lower left lobe of the lung

parenchymal fistulas causing lung abscess and nephro-bronchial fistulas causing passage of purulent urine into the bronchus<sup>8</sup>. In the 73 cases of nephro-pulmonary fistula which have been reported so far, nephro-bronchial fistula was seen in 58 cases (79%), 8 were nephro-pleural (11%), and 7 were nephro-parenchymal (10%)<sup>5,6,8</sup>. A review of literature revealed only 10 cases after 1949, some of them were discovered during postmortem autopsy<sup>5,6,8-16</sup>. Nephro-pulmonary fistula was associated with nephro-colonic fistula in 7 of these 10 cases<sup>5,11,14</sup>. Only one case had a cutaneous nephro-bronchial fistula opening in the renal angle after open drainage of the perinephric abscess<sup>15</sup>. The current case is the first reported cutaneous nephro-bronchial fistula that opens into the iliac fossa.

The chief complaint of most of the patients with nephro-pulmonary fistula was renal pain. However, some patients had pulmonary complaints like cough and fever<sup>5,6</sup>. Gordonson reported on a patient with nephro-pulmonary fistula suffering from expectoration of stones<sup>9</sup>. In the present case, the main complaint was dull aching left loin pain. The presence of a recurrent discharging cutaneous sinus gave the key for the definitive diagnosis by doing sinogram. The fistulous tract and the irritating pulmonary

symptoms after the procedure threw light on the diagnosis three months after the initial presentation. In agreement with Doughney et al.<sup>14</sup>, perinephric abscess and complicating fistula may remain occult. Symptoms, physical examination and laboratory investigations often are non-specific. A sterile urine culture does not exclude the presence of perinephric abscess<sup>17</sup>. Radiological studies may not be diagnostic. Caberwal et al. reported on a case of nephro-pulmonary fistula associated with nephro-colonic fistula in a patient that had been treated for a chronic lung abscess for four years before the final diagnosis could be made<sup>15</sup>. It is unusual for the nephro-pulmonary fistula to be radiologically demonstrated unless cutaneous fistula is present and a sinogram can be performed<sup>15</sup>. Although some cases of closed nephro-pulmonary fistula were detected by doing retrograde pyelography<sup>6,14</sup>, renal angiography was able to detect other cases by demonstrating a draining vein extending upward from the upper pole of the kidney through the diaphragm and emptying into the pulmonary venous system<sup>6</sup>.

The tuberculous kidney usually is adherent to the adjacent structures due to the severe inflammatory reaction. When it becomes adherent to the diaphragm, a sinus or fistulous tract may pass through a small area of areolar tissue between the costal part of the diaphragm and the fibers coming from the lateral arcuate ligament which separates the postero-superior surface of the kidney from the pleura. This could give a hint to the pathogenesis of the formation of nephro-pulmonary fistulas.

Treatment options include preoperative percutaneous drainage of the lung and perinephric abscess that could reduce the risk of intraoperative contamination of the lungs, retroperitoneum and abdominal cavity<sup>8,14,15</sup>. Rao et al. suggested the use of a double lumen endotracheal tube intra-operatively for sealing the main stem bronchus to prevent or minimize the contamination of the contralateral lung with purulent material in the dependent lateral decubitus position. They suggested that the fistulous tracts be transected early via a thoracoabdominal surgical approach to reduce the spillage of purulent material<sup>12</sup>. The present case was managed successfully by nephrectomy. The presence of a draining fistulous tract may explain the absence of intraoperative perinephric collection. The preoperative antituberculous treatment provided a clean operative field and a smooth convalescence.

In conclusion, this report is the first case of cutaneous nephro-bronchial fistula opening into the iliac fossa and the 13<sup>th</sup> reported case of tuberculous nephro-pulmonary fistula. It was successfully managed by nephrectomy under cover of the preoperative antituberculous therapy.

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