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Renocolic Fistula As A Complication To Xanthogranulomatous Pyelonephritis

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ABSTRACT

Four patients with xanthogranulomatous pyelonephritis were found to have renocolic fistulae. Coincidentally, the left kidney was involved in all four cases. All patients presented with renal mass. Two cases have had coexistent renal stones, one of them presented with massive upper gastrointestinal bleeding as a result of portal hypertension. Another patient had a history of Schistosomiasis.

In none of the patients was the renal condition confidently diagnosed preoperatively, nor was the colonic fistula suspected. In all four patients, nephrectomy was performed together with resection of the involved colon followed by a satisfactory recovery.

The possibility of a colonic fistula should be kept in mind as a complication to this rare renal condition in spite of the absence of colonic symptoms and normal finding in barium enema studies.

Keywords: xanthogranulomatous pyelonephritis, renocolic fistula, urinary fistula, colonic fistula

INTRODUCTION

Xanthogranulomatous pyelonephritis (XGP) was first described in 1916 by Schlagenhaufer[1]. The disease is an uncommon and atypical form of chronic pyelonephritis with segmental or diffuse changes of the renal parenchyma[2]. These changes may involve the perinephric fat and may extend to other retroperitoneal organs[3]. The disease, so called the great imitator, often mimics other inflammatory or neoplastic renal disorders and is frequently misdiagnosed clinically[4].

We present here our experience in four cases with XGP complicated by renocolic fistulae. One of them initially presented with massive upper gastrointestinal bleeding from gastric fundal varices; the renal condition was discovered during the investigation.

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Case 1

A 55-year-old male patient was admitted with massive upper gastrointestinal bleeding. He had no previous similar attacks, nor was he aware of other illnesses. He was conscious, well built and not jaundiced. Abdominal examination revealed the presence of hepatosplenomegaly, and no ascites. A large firm mass was felt at the left loin. Although the mass was ill-defined, it roughly measured 15 x 8 cm. A discharging sinus near the tip of the last rib was present for many years. Emergency upper gastrointestinal endoscopy showed normal oesophagus and large congested gastric fundal varices, one of which was bleeding. The rest of the stomach and the duodenum were normal.

The patient was resuscitated and responded well to conservative measures. Laboratory investigations showed normal renal and liver profiles, negative hepatitis serology and sterile urine. Plain abdominal film revealed the presence of a radio-opaque gallstone, and osteomyelitis of the left last rib. IVU showed enlarged but normal right kidney and non-functioning left kidney with multiple stones. Through the discharging sinus, a sinogram showed a communication with the collecting system of the left kidney. Ultrasound examination revealed hepatosplenomegaly with homogenous echogenicity, a large gallbladder stone and a dilated portal system. The right kidney was normal, while the left kidney was enlarged with different areas of varied echogenicity.

Liver needle biopsy showed mild degree of non-specific fatty change, mild acute and chronic inflammatory cells infiltration in the portal tracts and between the lobules, and areas of focal necrosis. No features of cirrhosis were seen. Needle aspiration cytology was done twice from the left kidney mass reported to be suspicious of malignant non-Hodgkin's lymphoma. Coeliac angiography showed normal hepatic and splenic vasculature and no malignant blush of the left renal vessels. Splenoportogram revealed gastric varices supplied by the coronary vein and the short gastric veins. The portal and splenic veins were dilated and patent. Pressure recordings in cm saline were as follows: Intersplenic 45, hepatic wedge 22.5, left renal 13 and inferior vena cava 12.

The abdomen was explored through a continuous bilateral subcostal incision. The liver looked and felt normal. The gallbladder had a thick wall and contained a single large stone with adhesions around it. The spleen and the left kidney were enlarged, firm and badly adherent to the surrounding. Large varices were found only at the fundal region of the stomach and there was no ascites. The gallbladder was first dissected and removed. Preoperative cholangiogram was done and found to be normal. Bile culture and biopsies from the right and left hepatic lobes were taken. Dissection and removal of the spleen was difficult due to much adhesions all around the organ, particularly posteriorly and inferiorly, near the upper part of the enlarged left kidney. Huge varices were

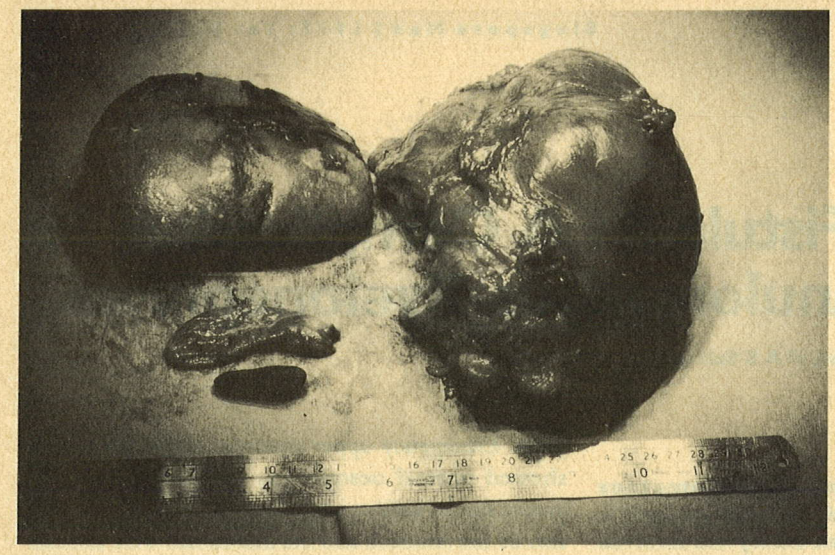

Fig 1 - The removed gallbladder, spleen and the left kidney.

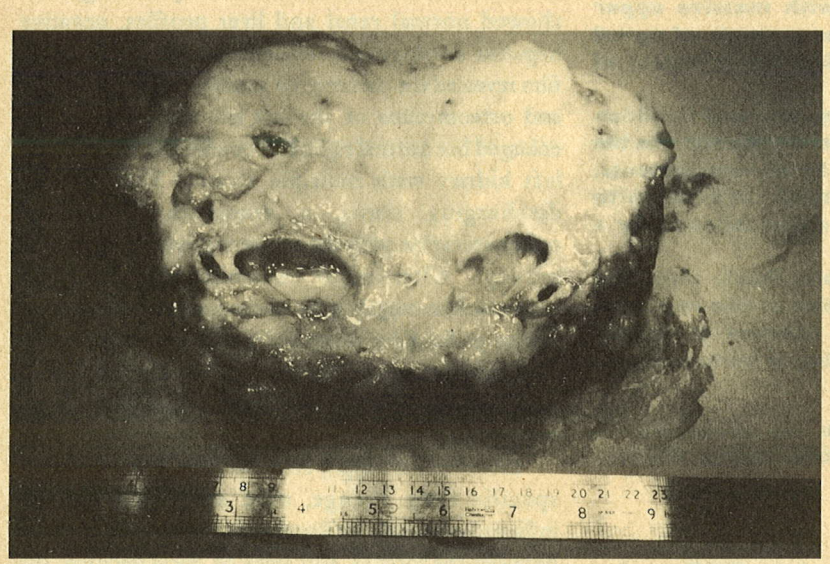

Fig 2 - The split kidney shows loss of normal features and multiple stones in the calyces.

found communicating the gastric fundus to the hilum of the spleen. After ligation, these were plicated meticulously within the wall of the stomach. The dilated coronary vein was then ligated and the abdominal part of the oesophagus and upper two-thirds of the stomach were completely devascularised. Lastly, dissection and removal of the left kidney was accomplished. Once the mass started to clear, it was found that the features are those of XGP. As expected, its dissection was difficult. Part of the descending colon was closely adherent to the mass. After dissection, a fistula was found and the 3cm defect in the colon was closed in two layers. Very bad adhesions were encountered at the region of the 12th rib and the sinus. During dissection, the pleura was opened but immediately closed. Post-operative recovery was uneventful. The bile culture grew *S typhi*. Fifteen days after operation, the patient was discharged home and continued to do well over the one-year follow-up.

The liver biopsies showed similar features to the previous preoperative report. The gallbladder had acute on chronic non-specific cholecystitis. The spleen had capsular thickening and fibrosis, red pulp congestion, white pulp atrophy, large areas of dense fibrosis and multiple ischaemic and haemorrhagic infarcts within the splenic pulp with occasional foci of non-specific calcification. Fig 1 shows the removed gallbladder, spleen and left kidney. The split kidney (Fig 2) had lost its architecture and contains multiple small stones. Microscopically, the capsule showed fibrosis and thickening. The changes extended to the perinephric fat which contained large areas of fat necrosis and abscess formation. Some tubules were atrophied while the others were dilated and contained colloid-like material. The majority of the glomeruli were sclerosed. The interstitium contained areas of chronic inflammatory cells infiltration with foci of oedema and fibrosis. There were large, multiple areas which contained central necrosis surrounded by an admixture of large finely granular foam cells, multinucleate giant cells, mononuclear cells, plasma cells, fibroblasts, polymorphonuclear leucocytes and lymphocytes. The granules were positive to periodic acid schiff stain. Also present were areas of calcification, dense collagenous fibrosis and foreign body type granulomata containing cholesterol clefts. The features were compatible with XGP with no evidence of malignancy. The reactions extend to the perirenal tissue and infiltrate the wall of the colon to the mucosa at the site of the fistula.

Case 2

A 41-year-old male patient presented with left loin pain and pyrexia of 10 days duration. Clinical examination was unremarkable, except for left loin mass which measured 8 x 4 cm. Laboratory investigations revealed the presence of leucocytosis and raised ESR. IVU examination showed normal right kidney and amputated lower calyx of the left kidney. Ultrasound examination and CT-scan showed a lower pole left renal mass. Angiogram study revealed the mass to be minimally vascular.

At exploration, dense adhesions were encountered around the lower half of the left kidney. It was stuck to the descending colon. The adherent part was therefore resected in continuity with the kidney and an end-to-end anastomosis was performed. The patient had an uneventful recovery.

On opening the colon, a small nodule was seen and felt at the site of the fistula through which a fine catheter was able to pass to the kidney. Macroscopically, the lower part of the kidney appeared to be replaced by fat and fibrosis and microscopically, the features were those of XGP.

Case 3

A 46-year-old male patient presented with left loin mass of 3 weeks' duration and fever over 2 weeks. He gave a previous history of urinary Schistosomiasis for which he received treatment. He looked toxic,

dehydrated and had a temperature of 39°C. On examination, a large tender left loin mass measuring 12 x 7 cm was felt. Laboratory investigations revealed leucocytosis and anaemia. The urine was sterile. Ultrasound examination revealed a left renal mass with mixed echogenicity. An IVU showed a normal right kidney and a mass involving the lower half of the left kidney and amputated calyces. At exploration, dense adhesions were encountered and after dissection, a fistula within the left colon was identified. The involved colon was resected and an end-to-end anastomosis was established. The patient had a satisfactory recovery. The final histology was consistent with XGP.

Case 4

A 55-year-old female patient presented with a 2-year history of recurrent left loin dull-aching pain and frequent attacks of urinary tract infection. She was known to be diabetic, hypertensive and had hemiplegia as a result of previous cerebrovascular haemorrhage. The patient was ill looking , had a temperature of 38.5°C and an ill-defined tender mass was felt in the left loin. Laboratory investigations revealed leucocytosis, anaemia, a raised ESR and blood sugar. *E coli* was cultured from the urine. Ultrasound examination revealed a left renal mass with mixed echogenicity.

IVU showed normal functioning right kidney and a large left renal mass with poor function and distorted calyces. Urine cytology reported to have high suspicion of malignant cells. A barium enema examination was carried out and the findings were normal.

At exploration, the large left renal mass measuring 10 x 6 cm was dissected out with difficulty. A perinephric abscess was found connected with the splenic flexure. The kidney and the involved colon were removed. The colonic continuity was established by end-to-end anastomosis and a proximal transverse colostomy was performed. Postoperatively, the patient had a slow recovery, and her colostomy was eventually closed six weeks later.

The split renal mass showed a large calculus lodged in the pelvis. The final histology was that of XGP. The resected colon segment showed mucosal ulceration and fissuring through the wall. The calculus consisted of xanthine crystals.

DISCUSSION

The clinical presentation of XGP, the possible association with other conditions, like diabetes, hypertension, chronic urinary tract infection, obstructive uropathy and renal calculi, had already been thoroughly investigated and discussed elsewhere[2-5]. The radiological changes and the pathological description with the possible aetiology of the disease had already been elucidated[6-9]. The disease had been reported to be associated with systemic amyloidosis[10], developed in a renal allograft[11] and occurred in patients with Schistosomiasis.

The disease usually affects adults in the fifth decade of life and women are affected more than men[4,5] . Occasionally, the disease was reported to have had a bilateral affection[12-14], however it is always unilateral with no predilection to either side[4,5].

According to Malek et al[4], the extent of the XGP inflammatory process can be divided into three stages: The first or nephric stage, when it is confined to the kidney. Stage two or perinephric, when Gerota's fat was affected in addition to the kidney. Stage three or the paranephric, when the process affected the kidney and its surrounding fat with widespread retroperitoneal involvement. According to this staging, all our patients had stage three disease. The descending colon being in the closest vicinity to the kidney was liable to be involved directly in the disease process. We think this involvement could be due to either the widespread extrarenal inflammatory process with the formation of dense adhesions, as it happened in cases 2 and 3; or the formation of perinephric abscess as seen in Case 4. The abscess thereafter eroded the colonic wall at its splenic flexure. In case 1 apparently, a chronic abscess had developed which eroded the tissue at the region of the 12th rib, thus creating a chronic sinus and osteomyelitis of the rib.

In all four cases, the hole which was found in the colon after dissection was not simply an iatrogenic one, but was actually a true fistula. Gross and microscopical examination revealed the colonic wall down to the mucosa to be abnormal and had the same inflammatory features.

All four patients have had no preoperative colonic disturbances. This might be due to the sufficiently patent bowel lumen and fine communication with the poorly functioning renal tissue. This could also explain why the barium enema examination which was carried out in Case 4 did not demonstrate the fistula.

Due to the benign nature of the disease, only the involved part of the colon needed to be resected. Colonic continuity can be established immediately, with or without colostomy, depending on the colonic contents at the time of resection and the condition of the patient.

In Case 1, the patient was discovered to be a typhoid carrier from the positive gallbladder bile culture. We think his gallbladder disease was a coincidental finding. The findings of normal liver histology and the negative hepatitis serology excluded hepatic origin. It is known that a patient with XGP might develop the syndrome of reversible hepatic dysfunction characterised by an increase in the serum alkaline phosphatase, glutamic oxalacetic transaminase 2 - globulin (occasionally 1 globulin) and sulfobromophthalein retention with hyperbilirubinaemia, hypothrombinaemia and hepatomegaly (non-specific focal hepatitis). The incidence of these changes varies from zero to 50% - 100%[4,15-17] and there is no evidence that these changes will lead to permanent pathology in the liver to cause portal hypertension. Furthermore, none of these features was present in our patients. On the other hand too many adhesions were encountered

around the spleen, particularly posteriorly and near the kidney. The spleen showed abnormal features microscopically. These adhesions most probably resulted in impedance of blood flow through the splenic vein and its tributaries near the hilum with secondary changes and congestion in the spleen, leading to the characteristic fundal varices usually associated with this disorder.

In conclusion, XGP frequently causes extensive extrarenal inflammation. The possibility of a colonic involvement should be kept in mind. Preoperative barium enema examination might not be useful in detecting the presence of the renocolic fistula. Resection of the involved part of the colon with immediate end-to-end anastomosis can be performed provided the colon was preoperatively prepared and the procedure was undertaken under antibiotics cover.

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