

Case Series

The Notorious Nephroenteric Fistulas and Nephrocutaneous Fistulas

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Abstract

Background: Nephroenteric and Nephrocutaneous fistulas occur rarely. The incidence of this condition has in recent times has declined due to advancement in treatment therapies. Currently, only a few cases have been reported in the literature.

Methods: We present largest single case series across the globe comprised of 6 patients treated at our hospital between 2016 and 2022, secondary to various pathologies like renal tuberculosis, XGPN etc.,

Results: The symptoms included loin pain with weight loss. *Escherichia coli* were the most common isolate on urine culture. CT with Fistulogram is diagnostic investigation of choice and management includes nephrectomy, excision of fistula and repair or resection of affected bowel.

Conclusion: Nephroenteric or Nephrocutaneous fistulas are an understudied entity. In most of the cases, affected kidney is usually non-functioning and hence, warrants Nephrectomy with surgical removal of fistula.

Keywords: Nephroenteric fistula; Nephrocutaneous fistula; Xanthogranulomatous pyelonephritis; XGPN; Tuberculosis; Retrograde pyelogram; CT; Nephrectomy

Abbreviations

XGPN: Xanthogranulomatous Pyelonephritis; CT: Computed Tomography; DJ: Double J; DTPA: Diethylenetriamine Pentaacetate; PCNL: Percutaneous Nephrolithotripsy; PCN: Percutaneous Nephrostomy; AFB: Acid Fast Bacilli; RFA: Radiofrequency Ablation

Introduction

Nephroenteric and Nephrocutaneous fistulas occur rarely and were first described 2400 years ago by Hippocrates [1,2]. The incidence of this condition in recent times has declined due to advancement in treatment therapies of renal pathologies. Out of Nephroenteric fistulas, nephrocolic fistulas are the most common. However, the vast majority of cases arise from a renal pathology rather than an enteral cause and of the former; the most common cause is Xanthogranulomatous Pyelonephritis (XGPN). XGPN is a distinct clinicopathological entity characterized by suppuration and histologically by the accumulation of lipid-laden macrophages. Approximately one third of XGPN cases have associated complications, such as abscess and fistulas. Here, we present a study of 6 cases of Nephroenteric and Nephrocutaneous fistulas treated in our hospital.

Methods

We would like to share our experience with Nephroenteric and Nephrocutaneous fistulas with a case series of 6 patients who presented

at our hospital between 2016 and 2022. The demographic data, clinical features, findings of Ultrasonography, Intravenous Urogram & Contrast Enhanced CT scan with fistulogram, preoperative diagnosis, Retrograde Pyelogram, operative findings and pathological features were recorded.

Cases Description

Case 1: XGPN with left nephrocolic fistula

A 37 years old diabetic male patient came with left flank pain since from 9 months. Patient had undergone DJ stenting previously for Left sided Emphysematous Pyelonephritis. Patient was anemic (Hb: 7.9 g/dL) and Serum Creatinine was normal (0.84 mg/dl). CT scan showed bulky Left kidney with DJ stent *in situ*. Multiple calculi were noted in shape of calyces, largest measuring 36 mm × 19 mm (mean HU=960) noted in mid pole extending into pelvis. Air foci were noted in pelvicalyceal system. Significant perinephric fat stranding was noted with fluid collections in perirenal space along psoas fascia (Figure 1).

Patient was taken up for Left sided PCNL 6 weeks following the DJ stenting. Intraoperatively, when Retrograde Pyelogram was done, contrast was found to be pouring into colon. Hence, procedure was



Figure 1: CT showing multiple calculi in left kidney with air foci.

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abandoned and Left Sided PCN & DJ stent was placed. Urine Culture from PCN was positive for *Escherichia coli* (Figure 2).

Following this event, DTPA Scan was done which was suggestive of non-functioning left kidney. So, Patient was further planned for Nephrectomy. The colon was dissected off from the kidney cautiously. The fistula was identified between the Interpolar region of the left kidney and descending colon, which was doubly ligated and excised (Figure 3). The colon was healthy overall with no signs of inflammation or necrosis. The part of colon with rent was excised and End-to-end anastomoses performed in two layers. The patient had a good postoperative recovery (Figure 4).

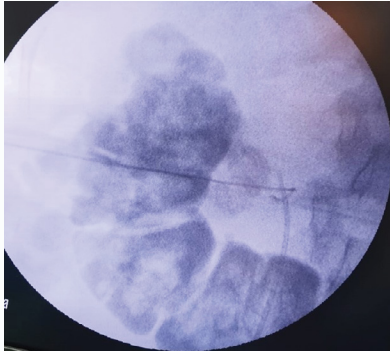


Figure 2: Retrograde Pyelogram (RGP) showing opacification of colon on instillation of contrast.

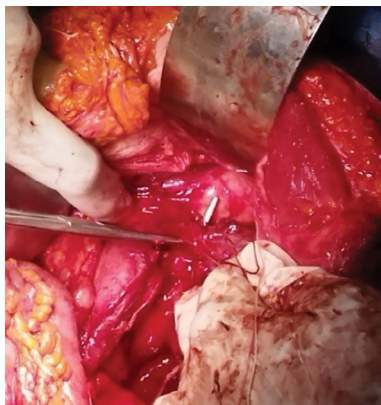


Figure 3: Intraoperative picture depicting right-angled dissector holding fistulous tract between left kidney and descending colon.

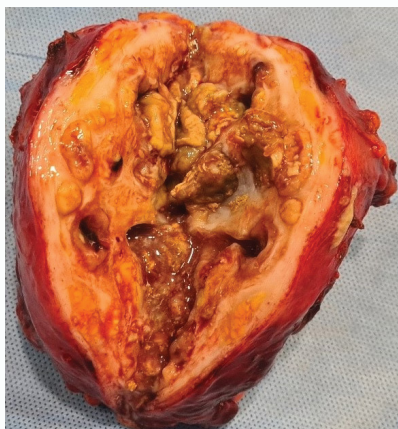


Figure 4: Specimen pictures of kidney showing presence of multiple cavities.

On Microscopy, diffuse inflammatory infiltrate predominantly composed of foamy histiocytes and multinucleated giant cells, admixed with lymphoid aggregates, plasma cells and few neutrophils were present. The fistula was lined with dense mixed inflammatory cells with XGPN tissue; there was no evidence of malignancy. Ziehl-Neelsen staining was negative (Figure 5).

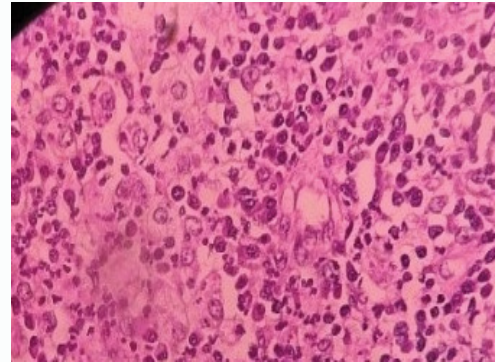


Figure 5: Microscopy picture of XGPN kidney showing lipid-laden macrophages.

Case 2: Chronic pyelonephritis with right nephrocolic, nephroduodenal and nephrocutaneous fistula

A 58-year-old female came to us with low grade fever and pus tracking from right flank region. No History of pain neither abdomen nor flank pain. On Examination, a fistula track was present at L2 level in post axillary line with active pus discharge. Laboratory investigations revealed normal creatinine with Total Leucocyte counts of $15600/\text{mm}^3$. Urine AFB Staining was negative. Urine Culture tested positive for *E. coli*. Pus culture showed Group A *Streptococci*. On Ultrasonography, right small kidney $5.8 \text{ cm} \times 3.8 \text{ cm}$ with Right Renal Pelvic Calculus 2 cm with Mild Hydronephrosis.

On Contrast CT, right kidney was small in size with irregular contour with features of pyelonephritis. A large pelvic calculus was present of about 23 mm . Two fistula tracts identified from right kidney communicating with skin and ascending colon. The ascending colon found adherent to posterior aspect of right kidney and fluid also seen tracking to undersurface of liver (Figure 6).

DTPA Scan was done which showed less than 8% relative uptake in right kidney indicating gross parenchymal dysfunction. Patient was planned for Right Open Nephrectomy (Figure 7).



Figure 6: CT image with fistulogram depicting communication between skin, kidney and ascending colon.



Figure 7: Fistulogram showing opacified duodenum and ascending colon.

Intraoperatively, Ascending Colon was adherent to right kidney forming fistula. Hence, Right Nephrectomy with Right Hemicolectomy with End-to-Side Ileocolic Anastomoses was performed. Another fistula was found between kidney and 2nd part of duodenum. Excision of fistulous tract was done with primary closure of duodenal rent. Postoperative period was uneventful. Histopathology showed features of chronic pyelonephritis (Figure 8).



Figure 8: Intraoperative picture showing the fistulous tract between right kidney and ascending colon.

Case 3: Left renal tuberculosis with psoas abscess with left nephrocutaneous fistula

A 47 years old female HIV positive patient known case of Pulmonary Tuberculosis presented with pus draining out of left flank region with flank pain for 2 months. Patient had completed Anti-Tubercular Treatment 5 years back. History of Left DJ Stenting in the past for Left Pyelonephritis 5 years back. Urine analysis revealed pyuria. Urine culture was sterile. Renal function tests were normal.

CT scan confirmed bulky Left kidney with thinned out renal parenchyma with Perinephric fat stranding. Left psoas muscle appeared bulky with collection. Fistulous tract was noted extending from skin in left posterolateral abdominal wall into the left psoas collection and left perinephric collection. Diuretic renogram showed non-functioning left kidney. Four weeks following her initial presentation, patient underwent Left Nephrectomy. Histopathology report revealed chronic granulomatous inflammation with central necrosis of the kidney. ZN Staining was positive (Figure 9).

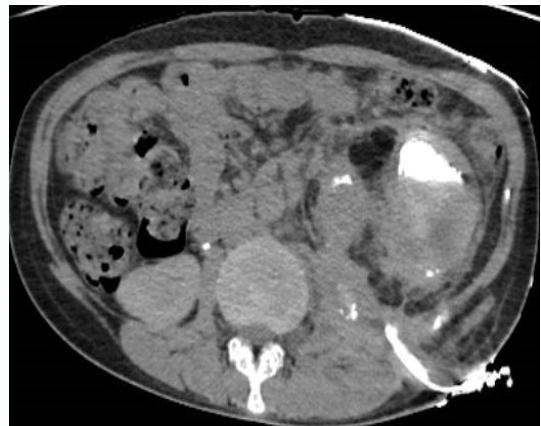


Figure 9: CT image depicting contrast flowing through the communication between skin, left kidney and left psoas muscle.

Case 4: XGPN with right nephrocutaneous fistula

A 58 years old diabetic female presented with complaint of Right sided flank pain and purulent discharge from right sided flank region. History of Pyonephrosis with Right Perinephric collection 2 months back for which Percutaneous Nephrostomy tube was inserted. On examination, there was tenderness in the Right renal angle but no palpable mass. A fistula tract opening seen just behind Posterior axillary line at L1 level. Patient was anemic.

Serum Creatinine and blood urea were within normal limits. Urine Culture grew klebsiella pneumoniae. Fistula was confirmed on CT scan combined with fistulogram along with presence of calculus measuring 2.6 cm noted in renal pelvis. No evidence of contrast excretion was noted with 60-minute delayed phase.

She was administered broad-spectrum antibiotics for the urinary infection. Patient underwent Right Open Nephrectomy with excision of fistula tract. Histopathology showed features consistent with XGPN (Figure 10).

Case 5: XGPN with right nephrocolic fistula

A 46 years old female patient came to us with right sided flank pain for the 6 months with low grade fever. No history of comorbidities. On examination, patient was having Right sided Palpable mass in lumbar region (Bimanually palpable). Upon Evaluation, renal functions were normal. Proteus mirabilis was cultured in the urine. CT showed right sided bulky kidney. Renal pelvi-ureteric junction calculus was noted measuring around 65 mm × 15 mm causing gross hydronephrosis with multiple dilated calyces. Few small air pockets were noted along the right renal cortex in the mid-pole region (Figure 11).

DTPA Scan showed non-functioning right kidney. Patient was taken up for Right Open Nephrectomy. Adhesions noted between right kidney and transverse colon. Adhesiolysis was done. Small

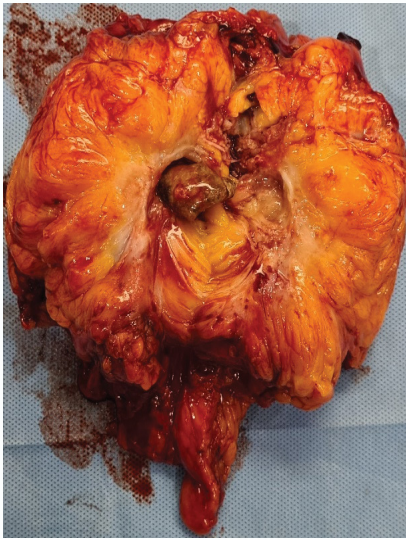


Figure 10: Picture showing specimen of XGPN Kidney with renal pelvic calculus.



Figure 11: CT showing Right sided Enlarged kidney with multiple dilated calyces (Classical-Bear Paw Appearance).

fistula tract was identified between right kidney upper pole and transverse colon and excised. Primary closure of rent in the transverse colon done. Surgery went on well. Pathology showed histiocytes and xanthoma cells which indicated xanthogranulomatous pyelonephritis (Figure 12).

Case 6: Left chronic pyelonephritis with left nephrocolic and nephrocutaneous fistula

A 37 years old lean, paraplegic male patient was admitted with purulent discharge from left lumbar region since from 2 months. History of fall from height 20 years back followed by spinal fixation. On evaluation, AFB staining was negative. *Pseudomonas aeruginosa* was detected in the urine culture. Creatinine was normal.

USG showed a sinus tract in left lumbar region, extending to paravertebral muscles. Left kidney could not be well visualized? Small in size with multiple renal calculi.

CT showed grossly shrunken left kidney in size with grossly thinned out renal parenchyma with few calculi in renal pelvis. No evidence of contrast uptake even after 1 hour of contrast administration- suggestive of nonfunctioning kidney. Multiple

irregulars conglomerated peripherally enhancing collections noted in the left psoas muscle and muscles of the posterior abdominal wall on the left side. Few pockets of air noted within the collection. CT Fistulogram confirmed communication between collection, left renal pelvis, left descending colon and fistula tract leading to skin.

Upon exploration, a renocolic fistula was found. Patient underwent Left open Nephrectomy with excision of fistula tract with large bowel resection and defunctioning colostomy. He was discharged home on post-operative day 7. Final Histopathology report confirmed presence of Chronic Pyelonephritis (Figure 13).



Figure 12: Gross specimen image of XGPN kidney with multiple dilated calyces.

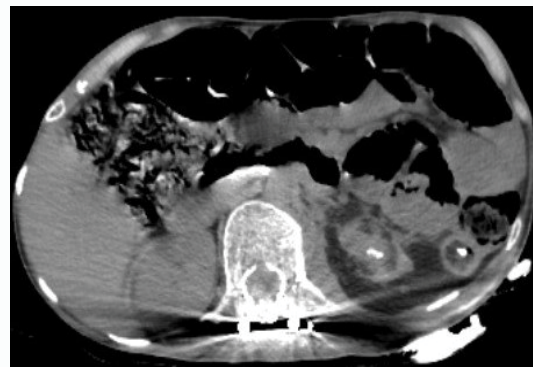


Figure 13: CT scan image showing presence of contrast and hence, communication between skin, left renal pelvis and left descending colon.

Discussion

Nephroenteric fistulas are a rare and unique entity. The ascending and descending colon are the most frequently affected. Whilst Tuberculosis was the leading cause of nephroenteric fistulas prior to 1950s, now it has been replaced by XGPN [3,4].

Aetiology includes Xanthogranulomatous Pyelonephritis, Calculous Pyonephrosis, non-calculous pyonephrosis, tuberculosis, perinephric abscess, and the rupture of hydronephrosis, malignancies, traumatic and sometimes iatrogenic causes like post-PCNL, RFA Ablation etc [5]. XGPN is a rare, severe inflammatory disease process

characterized by multiple cavities that replace the renal parenchyma. Microscopically, it is characterized by lipid-laden macrophages (xanthoma cells). Though rare but XGPN is notorious to make fistulas between upper urinary tract and surrounding organs [6,7]. There are reports of colonic, gastric, and jejunal fistulas [3,8,9]. There are also reports of fistulas to bronchial tissues and skin [7], psoas muscle, and the flank and gluteal regions.

Anatomically, the anterior surface of the left kidney is directly opposite the posterior wall of the descending colon, which lacks serosa and is the most typical location for nephroenteric fistulas. A chronic inflammatory process that first starts in the renal parenchyma and allows for the kidney's gradual adhesion to the colon, which clears the way for a nephrocolic fistula. Plus, however, may form and find its way to the skin's surface, leading to a cutaneous fistula [1].

The clinical picture resembles suppurative illness. Symptoms include fever, and weight loss associated with gastrointestinal symptoms like abdominal pain and diarrhea. Pneumaturia can be a presenting symptom in some cases. In some circumstances, pneumaturia may be a presenting symptom. There may be signs of anaemia, uremia, and septic shock in patients with persistent renal sepsis. There may occasionally be renal masses [5].

Investigations are non-specific, and the diagnosis is nearly always made radiologically through CT, RGP, or if there is cutaneous extension, fistulogram [6]. CT is usually the first line investigation in these patients and can reliably delineate anatomy and characterize the cause and extent of renal inflammation. Whilst CT can often suggest the presence of a Nephrocolic fistula, antegrade or retrograde pyelography is considered the gold standard in its diagnosis. A diuretic renogram can also be a useful adjunct in determining renal function prior to planning for nephrectomy. In the majority of cases, nephrectomy is necessary with excision of fistula tract and occasionally shows malignancy. The affected bowel is resected usually, and where conditions permit, a primary anastomosis may be performed.

Conclusion

Magnifying the awareness of Nephroenteric or Nephrocutaneous fistulas and identification of the correct etiology is crucial in our clinical practice. The most common etiology is XGPN. Recent years have shown a dramatic decline in the incidence of spontaneous nephrocolic fistula in TB patients due to timely multidrug tubercular treatment. However, renal tuberculosis being endemic in South Asian countries, we cannot afford to dismiss sneaky development of such fistulas in retained non-functioning kidneys. In our study, the most common organism cultured in urine was found to be *E. coli*. The preoperative CT can elicit the presence of these fistulas. The involved kidney is usually nonfunctioning and hence recommended treatment entails Nephrectomy with excision of the fistula tract with repair or resection of bowel segment. Usually, higher numbers of intra-operative challenges are encountered during these procedures and prolonged hospital stay can be expected after the surgery.

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