Clinical Images and Case Reports Journal

Case Report | Vol 6 Iss 2

Retroperitoneal Abscess with Reno-colic Fistula from Focal Xanthogranulomatous Pyelonephritis (XGPN)

Joanne Ehemann*

Department of Urology, Medical Officer, Princess Alexandra Hospital, Australia

***Corresponding author:** Joanne Ehemann, Department of Urology, Medical Officer, Princess Alexandra Hospital, Australia. E-mail: joanneehemann@gmail.com

Received: January 18, 2024; **Accepted:** February 02, 2024; **Published:** February 15, 2024

Abstract

XGPN is a rare chronic granulomatous inflammation of the renal parenchyma that if left untreated can lead to chronic renal injury. The mainstay treatment for XGPN is simple nephrectomy with curative intent. It represents just 1% of histologically diagnosed pyelonephritis with a 2:1 predilection for females within the 5th-6th decade of life. Nearly all cases are associated with staghorn calculi and often present with complications inclusive of peri-renal abscess, psoas abscess and acute kidney injury. We present a case of a rare complication: reno-colic fistula formation in an elderly woman with XGPN confirmed on histopathology at nephrectomy. Successful simple wedge resection of large bowel was undertaken with the patient having excellent post operative outcomes.

Keywords: Reno-colic Fistula; Xanthogranulomatous pyelonephritis; XGPN

Background

XGPN is an uncommon urological entity akin to chronic pyelonephritis, typically presenting with flank pain, haematuria, fevers in the presence of irritative urinary symptoms [1]. It is commonly miss diagnosed with a broad differential diagnosis. The mainstay diagnosis for XGPN is via computed tomography (CT) with contrast, urine microscopy culture and sensitivities (MCS) and standard laboratory investigations [2]. Often CT will demonstrate a bear paw sign (dilated collecting system surrounded by enhancing rim giving a loculated appearance) with associated renal calculi. The recommendation for treatment is considered to be case specific with intravenous antibiotics and percutaneous drainage suitable for unilateral or focal disease without complication. Definitive treatment is a partial or simple nephrectomy.

Case Presentation

A female in her 70's with a recent diagnosis of Child-Pugh A NASH (non-alcoholic Steatohepatitis) cirrhosis, presented to the emergency department (ED) with severe left flank, lethargy and nausea. She had elevated inflammatory markers and reduced renal function with a CRP of 243 and eGFR 33, Cr 136 (baseline renal function eGFR >90 and Cr 86).

Her Hemoglobin (Hb) was reduced at 111 and White cell count (WCC) elevated at 25 despite these significantly elevated makers of inflammation she remained afebrile within the ED. Computed tomography (CT) with contrast of the abdomen demonstrated a large complex retroperitoneal collection in the left lumbar region, extending medially towards the psoas, measuring roughly 330cc. Incidental findings of multiple left inferior pole renal calculi were also noted, with background renal changes consistent with left lower pole XGPN. The patient was transferred to a tertiary facility for further work up and percutaneous drainage of the psoas Abscess.

Percutaneous drainage of the lumbar abscess was performed successfully and lumbar drain left in situ with good resolution of collection (Figure 1). Cultures of the collection revealed a growth of proteas mirabilis and pseudomonas, Pan sensitive. The patient completed 6 weeks of intravenous antibiotics with follow up plan for outpatient Ureteroscopy (URS) and laser. On admission for URS, no lower pole stone was able to be visualised and as such the procedure was abandoned. The patient was booked for a semi urgent left nephrectomy in discussion with Urological consultants.

Intraoperatively, an active abscess within the kidney was discovered eroding into the left psoas muscle with an associated fistula tract to the splenic flexure of the colon. Two small defects were noted at the posterior wall of the splenic flexure of the colon and diagnosis of fistulous disease secondary to XGPN was made. Colorectal specialist input was sought intraoperatively and aided in surgical management of the patient. Review of images demonstrated close proximity of colon to left renal lower pole and suspicion of fistula tract was retrospectively acknowledged (Figure 2 and 3). Intraoperative discussion was comprehensive with a decision to attempt a wedge resection, a novel management strategy as documented cases of reno-colic fistula have either been managed conservatively or with segmental resection and end to end anastomosis [3].

Investigations

Haematology and biochemistry results on admission were as follows: FBC; Hb 111g/L, WCC 25 (neutrophils 21) LFTs; total serum Bilirubin (conjugated) 6umol/L, ALP 150U/L, 104 U/L, estimated glomerular filtration rate 33ml/min, Cr 136 umol/L.

CT abdomen + Pelvis: Large complex collection at the left lumbar posterolateral abdominal wall with extension to left psoas muscle superficially (Figure 2). Hyperdense left lower pole renal parenchyma with a moderate calculus in the inferior pole (Figure 4).

Urine MCS: 300 leucocytes/<10 erythrocytes/40 epithelial Bacteria +3 pseudomonas species

Interventional drain culture: Proteus Mirabilis

Histopathology of Nephrectomy: sections of kidney show large destructive necrotising suppurative granulomas which involve the full parenchymal thickness. Only focally few foam cells present. Imparting a xanthogranulomatous character. Specimen stains for microorganisms were negative. Tract in the perinephric tissue extending from the serosal surface to mucosa showing suppurative and inflammatory changes, suppurative granulomatous reaction pattern usually indicated an infectious aetiology.

Differential Diagnosis/DDx

There are a multitude of differential diagnosis for XGPN however, the most common diagnostic dilemma is its similarities to a neoplastic process. Renal cell carcinoma is the most common DDx due to both clinical and radiological features being similar to XGPN. Thus, early identification and treatment are paramount.

Treatment

The patient underwent a left simple nephrectomy with intraoperative colorectal consult and intervention. Decision made intraoperatively to perform a wedge resection in splenic flexure to excise fistula tract and primary closure with 3/0 PDS.

Outcome and Follow Up

The patient had an uneventful post operative recovery. At 6 weeks post procedure the patient was clinically well with nil evidence of complication. The patient presented 6 months post operatively with a single urinary tract infection (*E-coli*) managed with simple oral antibiotics. There have been no further complications or follow up required.

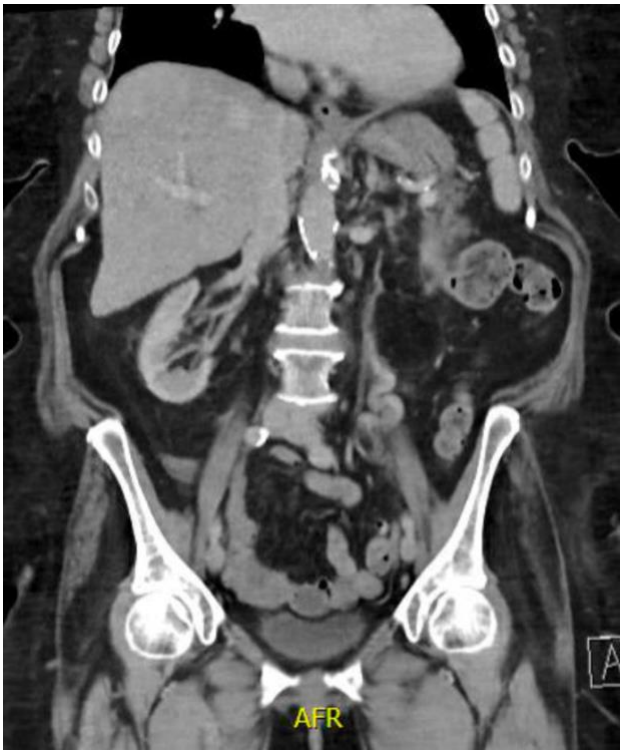

Figure 1: CT abdomen and pelvis with contrast on initial presentation demonstrating complex psoas collection.

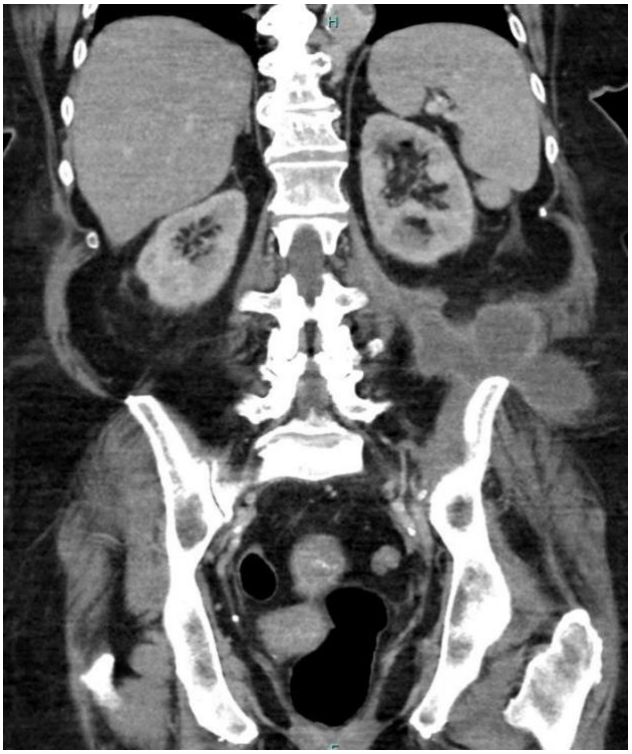

Figure 2: CT abdomen and pelvis with contrast on initial presentation demonstrating " Bear paw sign" unfortunately this was not identified at the time of reporting.

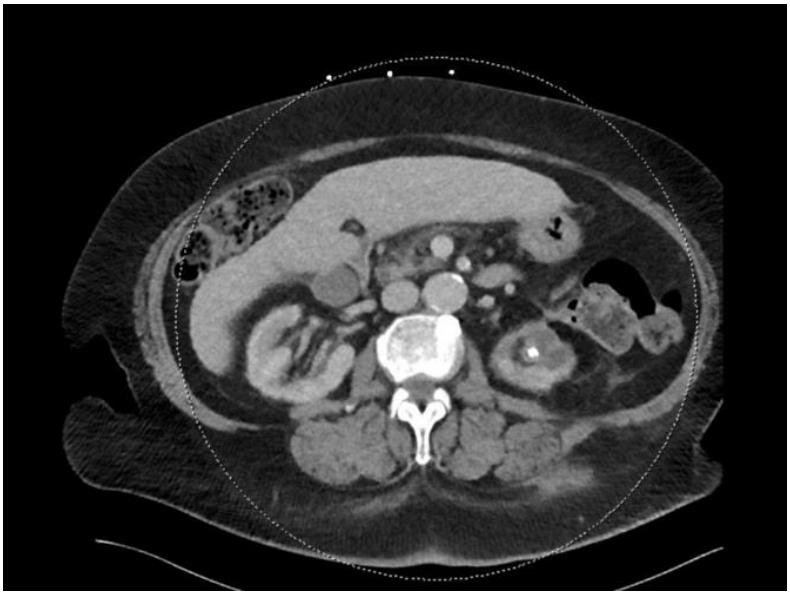

Figure 3: CT abdomen and pelvis with contrast on initial presentation demonstrating proximity of colon to lower pole XGPN.

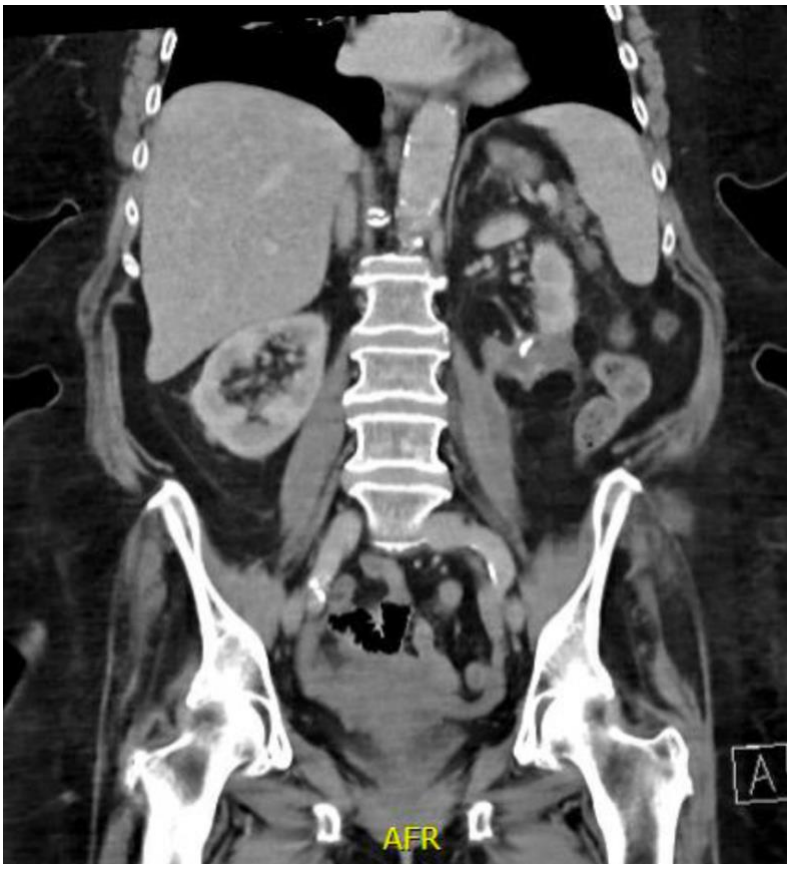

Figure 4: CT abdomen and pelvis with contrast post drainage of collection once again demonstrating proximity of colon to lower pole XGPN.

Discussion

Xanthogranulomatous pyelonephritis (XGPN) derives its name from "xantho" meaning yellow in reference to its gross appearance and granulomatous in regard to the foam laden granulomatous reaction seen on histology [1]. XGPN is a rare condition of severe chronic renal parenchymal inflammation leading to parenchymal replacement and destruction by foam laden macrophages. It usually presents in the context of chronic pyelonephritis, rental tract obstruction and/ or recurrent urinary tract infections (UTIs) [2]. Its incidence has been declining since the era of antibiotics and now accounts for roughly 0.6-1% of histologically classified pyelonephritis and causation of 19.2% of nephrectomies, performed due to chronic pyelonephritis [4]. XGPN can occur in all age groups however, has a predilection for middle aged to elderly patients with usual presentation in the 5th or 6th decade with a preponderance for females with a 2:1 ratio. This is thought to be due to the increased incidence of UTI in females due to anatomical factors [4]. Other notable risk factors include type two diabetes, advanced age, immunocompromised states and perhaps the most common association, is the presence of nephrolithiasis, predominantly struvite or staghorn calculi. These calculi are strongly associated with bacterial infection and colonisation, with the most commonly isolated bacterial species being *E-coli* and proteus mirabilis, as seen in our presented case. XGPN most commonly presents unilaterally yet there are case reports of bilateral disease which is traditionally associated with more severe disease and significant renal impairment [5].

Historically XGPN is classified into multiple subtypes;

1. Diffuse: diffuse renal involvement
2. Segmental: segmental involvement
3. Focal: cortical involvement only

Diffuse being the most common presentation, is further staged into three groups;

1. Nephric – limited to kidney
2. Perinephric – disease involving renal pelvis or perinephric fat within Gerotas fascia
3. Disease involving wider area including adjacent organs or retroperitoneum

As discussed, XGPN is associated with nephrolithiasis however, the exact pathophysiology and aetiology remains elusive. Modern theories centre around chronic urinary tract obstruction acting as a bacterial nidus and subsequent infection, this theory is backed by the presence of staghorn calculi in almost 80% of documented cases [6]. The associated inflammation and infection within the renal system leads to macrophage chemotaxis and granulomatous formation with lipid laden macrophages. XGPN can affect varying parts of the kidney however, it also due to its inflammatory nature, has the ability to affect surrounding organs inclusive of the liver, spleen, small bowel, pancreas, great vessels and in our case the colon [7].

Complications of XGPN are broad due to the inflammatory nature of the disease and close proximity of the kidney to multiple surrounding organs. Common complications include but not limited to; psoas abscess, perinephric abscess, acute kidney injury +/- nephrotic / nephritic syndrome and fistula formation as was the case with our patient. Whilst there are reports of fistulisation secondary to XGPN there is only one documented case of reno-colic fistula the authors are aware of [8]. Fistula diagnosis is considered a clinical diagnosis not histological, adding to potential error and missed diagnosis. The most common cause of reno-

colonic fistula is iatrogenic secondary to percutaneous nephrostomy and malignancy [9]. There are some reports of pyelo-duodenal fistula secondary to XGPN yet reno-colic appear to be extremely rare [10].

It is the broad nature of XGPN presentation that contributes to the diagnostic dilemma making the differential diagnosis pool vast. XGPN most commonly presents with fever, flank pain, lower urinary symptoms including dysuria and gross haematuria [11]. The vagueness of these presenting symptoms often mean XGPN is misdiagnosed as other inflammatory or neoplastic disorders and thus leads to higher rates of complications with delayed diagnosis [1]. Al Ghazo found patients with shorter duration of symptoms and early diagnosis had fewer complications at diagnosis and thus reinforces the need for early intervention and diagnosis within this pathological group [1]. Radiologically XGPN has similar appearance as RCC and further complicates clinical diagnosis. However, the most well-recognised pathognomic feature of XGPN is the bear paw sign, with pockets of abscess representing the bears paw, which can clarify diagnostic uncertainty [6].

The gold standard treatment for XGPN is nephrectomy with a reported 80% cure rate. Consideration as to partial or complete nephrectomy is based off disease classification i.e.focal vs diffuse and patient factors [1,6]. There is debate about best surgical approach laparoscopic vs open due to the inflammatory nature of the disease, often there is a high conversion to open (50%) and thus, it is an individual approach based on patient presentation, co-morbidities and surgeon expertise [12]. There are very few documented cases of conservative management of XGPN with these cases usually requiring prolonged course of intravenous antibiotics and close monitoring [1,11]. The general consensus is that it is imperative to removal all granulomatous tissue to prevent future complications and any sinus or fistula found should be primarily repaired and excised upon discovery. Reno-alimentary fistulas are an unusual and rare entity and best management is yet to be determined. Most cases of reno-alimentary fistulas are associated with urological procedures however, there is a small subgroup associated with inflammatory bowel disease. They may present with loose watery stools with or without blood, due to urine entering the lumen of the bowel. Concurrent symptoms may include faecal matter entering the renal tract presenting itself as gas within the collecting system, or propagation of infections.

Management of reno-alimentary fistulas are classically treated either conservatively with percutaneous nephrostomy placement and management of stone disease or nephrectomy with full bowel segment resection utilising either an end to end or side to side anastomosis [9,10]. This highlights the utility of our case report, not only does it add weight to current literature to highlight reno-colic fistula as a complication but it also demonstrates a novel technique in management. This is the first case to the authors knowledge whereby the fistula was managed with a simple wedge resection and primary closure with good long-term outcomes.

Learning Points

- Reno-colic fistula may be managed with simple wedge resection and primary bowel closure with good post operative outcomes.
- Delayed diagnosis of XGPN can lead to complex pathology and complications.
- Conservative management can be considered in select cases however, if patient failing to improve radiologically, clinically or biochemically then the decision to proceed to operative intervention should be made.
- Suspect reno-colic fistula in patients failing conservative measures with surrounding renal inflammation.
- Specialist Colorectal input should be sought in management of reno-colic fistulas.

REFERENCES

1. Al-Ghazo MA, Ghalayini IF, Matalka II, et al. Xanthogranulomatous pyelonephritis: Analysis of 18 cases. Asian J Surg. 2006; 29: 257-261.
2. Artiles-Medina, Laso-García, Lorca-Álvaro, et al. Xanthogranulomatous pyelonephritis: A focus on microbiological and antibiotic resistance profiles. BMC Urol. 2021; 21: 56.
3. Lee SD, Kim TN, Ha HK. Delayed presentation of renocolic fistula at 4 months after blunt abdominal trauma. Case Rep Med. 2011; 103497.
4. Kundu R, Baliyan A, Dhingra H, et al. Clinicopathological Spectrum of Xanthogranulomatous Pyelonephritis. Indian J Nephrol. 2019; 29: 111-115.
5. Fan CM, Whitman GJ, Chew FS. Xanthogranulomatous pyelonephritis. AJR. American journal of roentgenology. 1995; 165: 1008.
6. Patrick Jones, Darko Lazic, Bhaskar K Somani, et al. Xanthogranulomatous pyelonephritis: an overview and management guide for clinicians. British Journal of Hospital Medicine. 2021; 82: 1-8.
7. Jha SK, Aeddula NR. Pyelonephritis Xanthogranulomatous. Treasure Island (FL): StatPearls Publishing. 2023.
8. Sforza D, Siragusa L, Ciancio Manuelli, et al. A Rare Case of Xanthogranulomatous Pyelonephritis with Spontaneous Renocolic Fistula and IVC Thrombosis. Case Rep Nephrol. 2021: 3604017.
9. Bissada NK, Cole AT, Fried FA. Reno-alimentary fistula: an unusual urological problem. J Urol. 1973; 110: 273-276.
10. Dawoud S, Solomon RJ, Eyerly-Webb SA, et al. Pyeloduodenal Fistula in Xanthogranulomatous Pyelonephritis: A Series of Two Cases. Perm J. 2018; 22: 17-150.
11. Korkes Fernando. Xanthogranulomatous pyelonephritis: clinical experience with 41 cases. Urology. 71: 178-180.
12. Shah KJ, Ganpule AP, Kurien A, et al. Laparoscopic versus open nephrectomy for xanthogranulomatous pyelonephritis: An outcome analysis. Indian J Urol. 2011; 27: 470-474.