

Xanthogranulomatous Pyelonephritis – Challenges in Management

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Abstract We present a case of a 50-year-old lady left gluteal collection with communication into the left retroperitoneal plane and continuous with left xanthogranulomatous pyelonephritis. She underwent wound debridement and left nephrectomy. It was subsequently complicated with delayed colonic perforation, recurrent episodes of colocolic and nephro-colic fistula. We discuss the complications associated with xanthogranulomatous pyelonephritis and the challenges in managing it.

Keywords: Xanthogranulomatous pyelonephritis, Nephro-colic, Fistula, Nephrectomy, inflammation, psoas abscess

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1. Introduction

Xanthogranulomatous pyelonephritis (XGP) is a histological subset of pyelonephritis which is described as being a chronic destructive granulomatous inflammation of the renal parenchyma. It is reported that until 1980, only about 400 cases were reported worldwide. [1] XGP can lead to complications with associated morbidity and mortality such as perinephric abscess, psoas abscess, nephro-cutaneous fistula and nephro-colic fistula. [2] Fistulas between the gastrointestinal tract and kidney are not common with about 130 cases reported. [2] In most scenarios, symptoms such as flank pain and urinary symptoms were present leading to early diagnosis and subsequent definitive management. Peculiar to our case, the patient presents with a long-standing left gluteal abscess which was later investigated to be originating from left XGP.

2. Case Presentation

A 50-year-old overweight female with long-standing left nephrolithiasis and diabetes mellitus type 2 presents with a 3-month history of a left gluteal abscess with features of fever, anorexia, pus, and discharge from the gluteal wound. She has a history of left percutaneous nephrolithotripsy (PCNL) performed for left renal stone 10 years prior. She subsequently defaulted further follow up.

Computed tomography (CT) with intravenous contrast abdomen and pelvis revealed contrast-enhanced collection

at the subcutaneous layer of the left gluteal region with the presence of air within the left lower calyx with moderate left hydronephrosis. This is consistent with left gluteal collection with left abdominal wall involvement and retroperitoneal extension with left xanthogranulomatous pyelonephritis (XGP). (Figure 1, Figure 2 and Figure 3)

She underwent wound debridement and exploration of the left gluteal abscess with left nephrectomy. Intraoperative findings include left gluteal sinus tract tracking superficially from left gluteal to left psoas muscle and into the left renal parenchyma (Figure 4 and Figure 5). Left nephrectomy was done in view of the left kidney involvement with the collection. Histopathological report of the specimen is consistent with left chronic pyelonephritis with *Actinomyces* infection and sinus tract formation.

Feculent discharge was noted from the surgical scar on postoperative day 8. Gastrografin small bowel follow-through contrast study showed no contrast leak from the small intestine. Follow up abdominal x-ray after 12 hours showed the presence of contrast leak from descending colon into the retroperitoneal plane.

Wound re-exploration was done and intra-operative findings are 2 extraperitoneal perforations in descending colon measuring less than 0.5cm. The perforations were primarily repaired using interrupted absorbable sutures followed by a diversion loop ileostomy. A passive subcutaneous drain was also placed. Postoperative recovery was uneventful and she was able to tolerate a regular diet and ambulating on postoperative day 3.

On postoperative day 6 however, pus mixed with feculent discharge was noted from the subcutaneous drain. Per rectal contrast study revealed contrast leakage from descending colon into the retroperitoneal plane and in

communication with the subcutaneous tissue. There was no evidence of a leak from the small bowel. A diagnosis of nephro-colic fistula with complex colo-cutaneous fistula was established (Figure 6). In view of functioning small bowel with diversion loop ileostomy she was allowed normal diet with oral nutritional support. The drain output progressively changed to serous in consistency with output volume decreasing in trend before it was removed on postoperative day 20. The surgical wound was initially managed with daily dressing using povidone-iodine, followed by normal saline. Suitable granulation of the subcutaneous tissue followed by epithelization of the overlying skin was observed.

The wound however, failed to heal entirely and an opening measuring less than 1cm along the surgical scar persisted with minimal amount of pus discharge noted. Ultrasonography assessment of the wound revealed localized subcutaneous collection measuring 5cm x 5cm with extension to the deep muscle. Pus culture showed mixed growth. She was advised for open drainage of the subcutaneous collection but she refused consent. It was managed with one week course of intravenous antibiotics and daily dressing of the wound.

A small opening less than 0.5cm however persisted and continued to discharge minimal amount of pus daily. CT scan showed an extension of the subcutaneous collection into the deep gluteal muscle. Contrast study showed persistent communication between the subcutaneous collection and a pin-point point of the leak at the

descending colon. She was advised for open drainage but persistently refused consent.

While she continued to tolerate orally with oral nutritional support regime, it was noted that her loop ileostomy output progressively increased in volume over time. Daily stoma charting consistently recorded production of between 1 to 2 liters per day. During the 4-month duration following her second surgery she required multiple admissions for management of the high output fistula. Imaging showed good length of functioning small bowel and her electrolyte parameters were within normal limits. While the pus discharge from the wound persisted she was never in sepsis during her repeated admissions.

In view of persistent discharge from the wound associated with the pinpoint point of leak from the splenic flexure and the high output ileostomy she was planned for endoscopic clipping of the luminal fistula followed by a reversal of the loop ileostomy.

In an unfortunate turn of events, she failed to turn up at her scheduled clinic follow up and further inquiry revealed that she was having fever at home associated with poor oral intake for one week and pus discharge from the surgical wound. She was brought to the hospital in septic shock and despite resuscitation, she succumbed.

3. Figures



Figure 1. Axial CT showing Left XGP with extension to left pararenal areas including psoas muscle

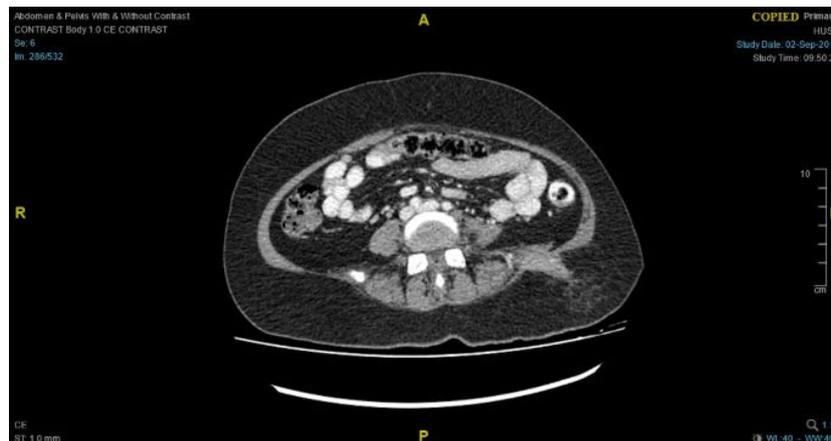


Figure 2. Axial CT showing psoas muscle involvement with extension into gluteal region forming an abscess



Figure 3. Axial CT showing the sinus tract originating from XGP

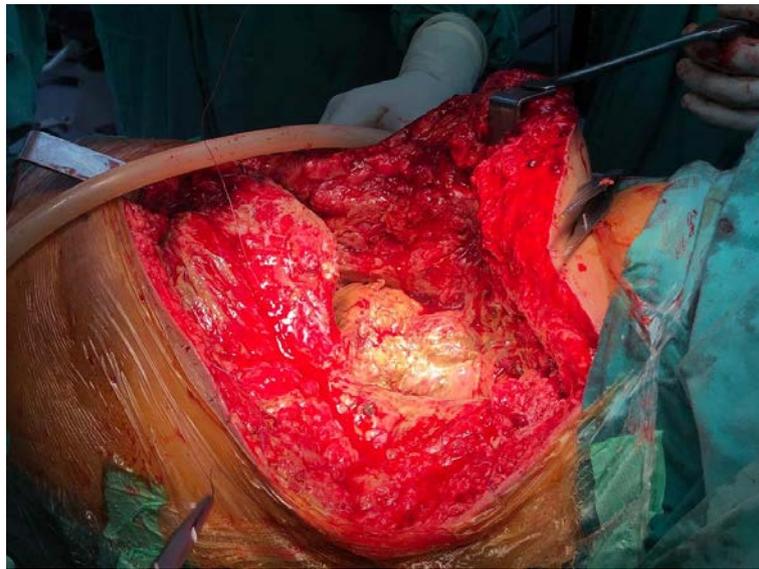


Figure 4. Intraoperative diagram showing the area of fistula of descending colon



Figure 5. Resected specimen of nephrectomy and gluteal sinus tract

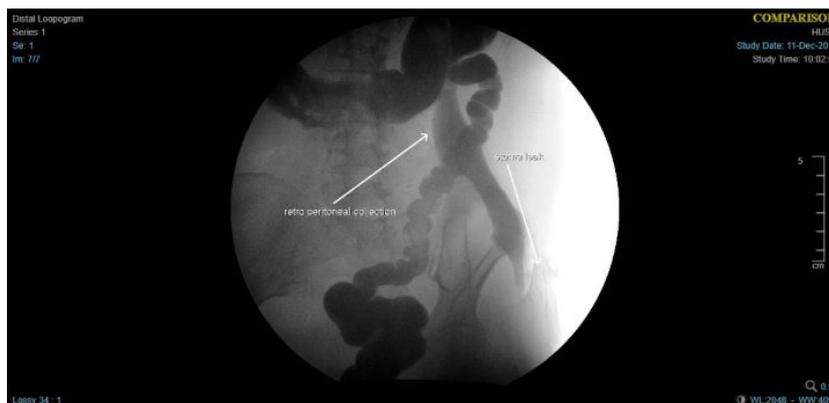


Figure 6. Lopogram showing established complex enterocutaneous fistula

4. Discussion

XGP is a chronic destructive granulomatous process of the renal parenchyma and was first described by Schlagenhauer in 1916 [3]. It is more common in females between 50 to 60 years of age. Symptoms typically include flank or abdominal pain along with pyrexia, gross hematuria and unintentional weight reduction. [4] Our patient presented with chronic left gluteal abscess and on CT there was the presence of left psoas muscle involvement. This coincides with the classification by Malek et al. in which XGP is divided into 3 stages. Stage 1 is a localized process that involves renal parenchyma; stage 2 can spread to the perinephric fat, stage 3 further spreads to involve the perinephric and paranephric areas resulting in perinephric or psoas abscesses. [5] However, more commonly, a radiologically based classification stratifies XGP into a focal(localized, segmental) form (15%) and a diffuse type (85%). [6] The presence of psoas abscess, like our case, is rare and the diagnosis and management is difficult. Goldman et al. discovered six psoas abscess in his case series with 16 cases of XGP. Moreover, like our case, all patients had a diffuse form of the disease. [7]

Complications include nephro-colic fistulas [8]. The ascending and descending parts of the large intestine are the most frequently affected. It is not seen often as a complication of XGP and as of now, there are no big case series published in the literature [9]. Even though the left kidney was removed during the primary surgery, the presence of persistent communication between the descending colon and the left renal tissue plane strongly suggests that the underlying chronic inflammatory tissue of the XGP as the most likely etiology for the fistula.

Nephro-colic fistulas commonly are either traumatic or spontaneous. Traumatic cases are less common and are usually iatrogenic, occurring after open or percutaneous surgical procedures, although they can be inflicted by severe renal trauma (penetrating and blunt). Spontaneous nephro-colic fistulas almost always arise as a consequence of primary renal pathology, which may include stone diseases and renal malignancy. Some of the other causes include diverticular disease and inflammatory bowel diseases. Tuberculosis also remains a significant cause of spontaneous nephro-colic fistulas. [10]

In this case, we postulate the nephro-colic fistula is a result of a longstanding XGP. Anatomically to the posterior wall of the descending colon lies in the

retroperitoneal plane in contact with the anterior surface of the left kidney. The chronic inflammatory process in the renal parenchyma extends to the adjacent wall of the descending colon and subsequent nephro-colic fistula [11].

Investigations are incessantly non-specific, and the diagnosis is likely made radiologically through a barium enema, retrograde pyelography, antegrade pyelography, CT scan and or if there is a cutaneous extension, fistulogram. The classical CT scan finding of XGP is the ‘Bear’s paws sign’ which refers to the cross-sectional appearance of the kidney which resembles the paw of a bear. The renal pelvis is contracted whereas the calyces are dilated, mimicking the toe-pads of the paw. [12] The mainstay of treatment for nephro-colic fistulas is open surgery, except for post-instrumentation fistulas, which, if very small, maybe treated conservatively. In the focal/segmental variants, if diagnosed early, preoperatively can be treated with antibiotics, followed by resection of the fistula tract to the kidney. In the majority of cases, however, nephrectomy is necessary, and the affected kidney is often completely destroyed and occasionally shows malignancy. The involved bowel is resected, and where conditions permit, a primary anastomosis is performed. [13]

In our case, the presence of extensive renal damage and associated changes due to XGP affected our decision to perform a left nephrectomy. During the primary surgery assessment of the adjacent descending colon upon removal of the left kidney showed normal bowel anatomy macroscopically. It was only during the re-exploration that the small perforations on the retroperitoneal part of descending colon wall with surrounding inflammatory tissue that we postulate a nephro-colic fistula as a likely etiology for the perforation.

The prognosis of the condition mainly depends on the underlying cause, the duration of the disease, the extent of renal insufficiency and the general condition of the patient.

5. Conclusion

Xanthogranulomatous pyelonephritis with nephro-colic fistula is an entity that needs to be recognized early for prompt intervention. It is not a common entity and will benefit from multi-disciplinary involvement to provide the best possible care with the best possible outcome for the patients.

Conflict of Interest

None.

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