

Xanthogranulomatous Pyelonephritis Complicated by Spontaneous Pyeloduodenal Fistula: Role of Computed Tomography Scan

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ABSTRACT

Xanthogranulomatous pyelonephritis (XGP) is a rare chronic inflammatory disease of the kidney. It is uncommon in children, and formation of spontaneous pyeloduodenal fistula is exceedingly rare. Computed tomography (CT) scan is useful in comprehensive preoperative evaluation and detection of associated congenital anomalies. It is also accurate in detection and delineation of fistulous tracts. We present a case of XGP in a young girl with malrotation of gut and horseshoe kidney complicated as spontaneous pyeloduodenal fistula and psoas abscess. Here, we demonstrate the role of imaging, especially CT scan, in the preoperative diagnosis of such fistula.

Key words: Computed tomography scan; pyeloduodenal fistula; xanthogranulomatous pyelonephritis

Introduction

Xanthogranulomatous pyelonephritis (XGP) is a rare entity in children, and formation of spontaneous pyeloduodenal fistula even more so. XGP in children is commonly associated with an obstructed urinary tract secondary to nephrolithiasis or congenital anomalies such as horseshoe kidney. Computed tomography (CT) scan is instrumental in a comprehensive preoperative evaluation of XGP. It is also accurate in detection and delineation of fistulous tracts. We present a case of XGP in a young girl with malrotation of gut and horseshoe kidney complicated as spontaneous pyeloduodenal fistula and psoas abscess.

Case Report

A 15-year-old female presented with pain in the right loin for 6 months along with vomiting and intermittent low-grade fever. Two months back, she had difficulty in extending the

right thigh. An ultrasound (USG) study was done outside that suggested the diagnosis of horseshoe kidney with a right perinephric abscess. She was managed conservatively there with antibiotics and analgesics with improvement in her symptoms. One month back, her symptoms reappeared and were associated with fever, chills, rigor, vomiting, and difficulty in walking. Then, she was referred to our institute.

On examination, she was malnourished, pale, and febrile. Her serum creatinine was 1.1 mg%. There was tenderness in her right lumbar region with a vague palpable lump on the right side of the abdomen. There was no rigidity or guarding. She also had difficulty in extending the right thigh. The USG showed horseshoe kidney with hydronephrosis of the right moiety of kidney associated with floating echoes within the dilated pelvicalyceal system (PCS). The right psoas muscle was

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bulky and hypodense suggesting psoas abscess. A diagnosis of right pyonephrosis with psoas abscess was made, and percutaneous nephrostomy (PCN) and abscess drainage were planned.

During the PCN procedure, nonionic iodinated contrast was injected in the right PCS. Contrast leakage into duodenum was noted via a thin tract [Figure 1]. 12 F Malecot catheter was placed within the right PCS, and contrast enhanced CT scan (CECT) was performed for proper delineation of the tract. On CECT, fistulous communication was seen connecting the medial aspect of the right PCS and the second part of duodenum [Figure 2a-e]. The right perinephric fistula also noted with tracking of contrast along the right psoas muscle [Figure 2e]. The right psoas muscle abscess along with multiple small pockets of collections in the

retroperitoneum were noted [Figure 3a and b]. Marked ascites and mild left pleural effusion were also present. This is the first time when we suspected associated malrotation of the gut [Figure 3c and d]. Thus, after CECT, she was diagnosed to have malrotation of gut and horseshoe kidney with right upper moiety pyonephrosis complicating as pyeloduodenal fistula with right perinephric and psoas abscess.

The patient was optimized and operated under general anesthesia. All preoperative diagnosis was confirmed after exploration. Complete excision of the right moiety of horseshoe kidney, placement of double J stent on left moiety with correction of malrotation of the gut (Ladd's procedure with appendectomy) was performed, and duodenal fistula was repaired. Psoas abscess was drained, and a wide bore drain was placed in abscess cavity. A feeding jejunostomy was also done. No significant event occurred in the postoperative period, and feeding was started through jejunostomy tube from the 3rd day of surgery, which was well tolerated by the patient. Subsequently, water soluble contrast meal and follow through the study was done which showed patent bowel with no evidence of leak. The patient was allowed oral feeds and discharged.

Histopathological analysis of nephrectomy specimen showed markedly attenuated renal parenchyma which was infiltrated by sheets of foamy, lipid-laden histiocytes along with dense mixed inflammatory cells infiltrate. There was no evidence of malignancy. Pathologically, a diagnosis of XGP was made.

Discussion

XGP is a rare and aggressive form of chronic bacterial infection of the kidney and surrounding tissues are characterized by destruction and replacement of renal parenchyma by

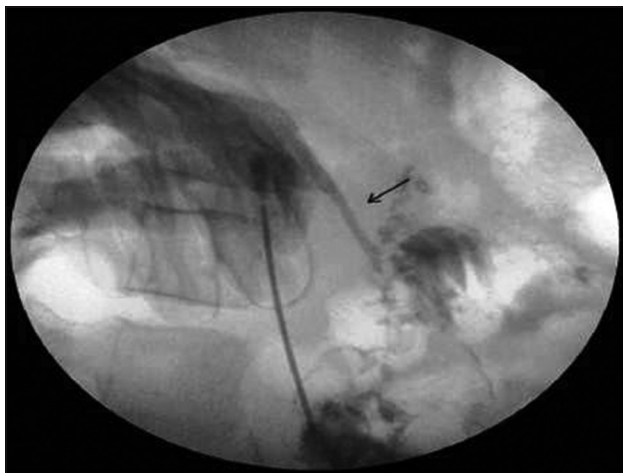


Figure 1: Fluoroscopic image showing Vygon needle in the pelvicalyceal system of the right kidney. Contrast agent is seen tracking down from the pelvicalyceal system of the right kidney to the bowel loops (arrow)

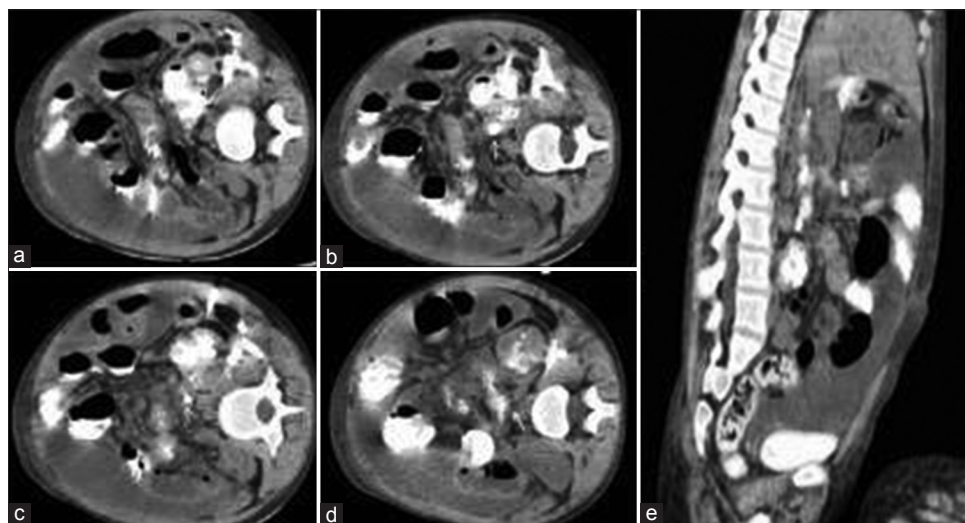


Figure 2: Left lateral decubitus multiple detector computed tomography images from above downward. (a-d) The passage of contrast agent from the right pelvicalyceal system to the duodenum (arrow) and sagittal multiplanar reformation of left lateral decubitus view. (e) The fistulous tract from the right pelvicalyceal system to the duodenum (black arrow)

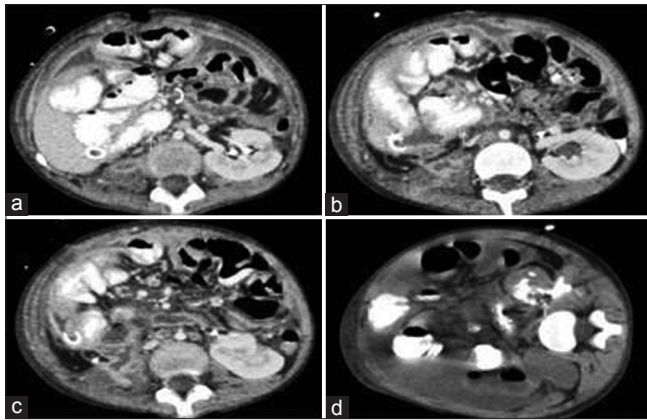


Figure 3: (a) Right-sided psoas abscess along with multiple pockets of retroperitoneal collection. (b) Left lateral decubitus image showing the leakage of contrast agent from the right pelvicalyceal system to the right perinephric space (arrow). (c and d) The malrotation of gut as seen by left-sided placement of duodenal jejunal junction (straight arrow) and inverse relation of superior mesenteric arteries superior mesenteric vein (curved arrow)

lipid-laden macrophages. It is often associated with calculi and urinary tract obstruction.^[1] XGP usually occurs in the middle-aged, most commonly in females who are diabetic or immunocompromised. XGP is rare in children. In children, it is most commonly associated with nephrolithiasis.^[2] Congenital anomalies such as horseshoe kidney causing urinary tract obstruction are also described in XGP.^[2] In our case, possibly the presence of horseshoe kidney was the cause of obstruction of the right PCS.

XGP is associated with fistula formation between the genitourinary tract and other hollow viscera such as the colon.^[3] Nephrobronchial^[4] and nephrocutaneous fistulas^[1] are also described. To our knowledge, however, there is only one case of spontaneous pyeloduodenal fistula associated with XGP reported in literature.^[5] Spontaneous pyeloduodenal fistulas are usually associated with chronic inflammatory processes of the kidney.^[6] Retrograde ureterography/pyelography is traditionally described as the investigation of choice to detect pyeloduodenal fistula. Other procedures such as antegrade pyelography may also demonstrate the fistulous tract as in our case.

CECT scan is the preferred imaging modality for the evaluation of pyelonephritis. XGP typically shows unilateral renal enlargement or focal mass, parenchymal, and perirenal inflammation associated with multiple areas of low attenuation representing dilated renal calyces and pus filled cavities. The presence of obstructing calculus is a common finding.^[7] In addition, CT accurately delineates any fistulous tract as it did in our case. Furthermore, the leakage of contrast agent seen in the perinephric space was demonstrated, which was not observed on antegrade pyelography.

Nephrectomy with primary duodenal repair has been described as the treatment of choice in cases of spontaneous pyeloduodenal fistula associated with chronic inflammatory disease of kidney as the affected kidney is often nonfunctioning.^[6] About 75% of cases occur spontaneously either due to chronic infectious disease, calculus or because of malignancy. Rest is associated with trauma or surgery.^[1-7] Furthermore, the presence of horseshoe kidney and malrotation of the gut were two different congenital conditions complicated by XGP as pyeloduodenal fistula. All these conditions and complications were accurately diagnosed preoperatively by CECT.

Conclusion

XGP is a rare disease in children; however, its possibility should be considered in the presence of a focal or diffuse renal enlargement associated with perinephric involvement and the presence of obstruction such as nephrolithiasis and congenital anomalies. Spontaneous fistulization to hollow organs and skin is possible in XGP. CT scan is effective not only in evaluating the extent of inflammation but also delineates any fistulous communications accurately along with other associated congenital anomalies.

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Conflicts of interest

There are no conflicts of interest.

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