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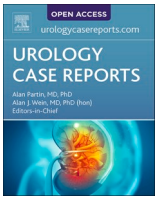

Inflammation and infection

Paediatric xanthogranulomatous pyelonephritis with reno-psoas fistula, psoas abscess and migration of renal calculi

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ABSTRACT

Xanthogranulomatous pyelonephritis (XGP) is a rare form of chronic pyelonephritis characterized by granulomatous tissue replacing renal parenchyma, primarily in adults. It's often linked to chronic obstruction, urolithiasis, and pyelonephritis, with rare associations with psoas abscess or fistula. A 15-year-old girl, initially treated conservatively for suspected pyelonephritis, underwent CT imaging due to non-response. This revealed XGP with a sizable psoas abscess caused by a kidney-psoas muscle fistula and renal calculus migration. Treatment involved percutaneous abscess drainage, open right nephrectomy, and a prolonged antibiotic regimen.

1. Introduction

Xanthogranulomatous pyelonephritis (XGP) is a rare subtype of chronic pyelonephritis characterized by the replacement of renal parenchyma with granulomatous tissue containing high concentrations of foam cells.[1] Histologically it is characterised by loss of renal parenchyma, granulomatous inflammation, fibrosis, diffuse inflammatory infiltrates and classically, lipid-laden foamy macrophages resulting in fatty deposits throughout the kidney.[1] XGP accounts for only 0.6 % of histologically confirmed chronic pyelonephritis cases.[2] This condition predominantly affects females and occurs most frequently between the ages of 40–60 years.[3] Chronic obstruction, often due to a large stone burden, leads to the inflammatory destruction of renal parenchyma, which can manifest as focal or diffuse damage.[4] Associated risk factors include diabetes mellitus, hypertension, chronic renal stone formation, recurrent urinary tract infections, immunosuppression, malnutrition or congenital kidney disease.[1] Complications may include emphysematous pyelonephritis, perinephric abscess, psoas abscess, nephrocutaneous fistula (rare), and ischemic colitis secondary to mass effect. The most common infectious agents associated with XGP are Escherichia coli and Proteus species.[1]

Computed tomography (CT) is the preferred imaging modality, revealing renal parenchymal loss, a contracted renal pelvis, and enlarged dilated renal calyces, often referred to as the "bear paw sign".[4] Contrast CT intravenous pyelogram will usually demonstrate poor uptake in the affected kidney with little to no excretion due to its reduced or non-functioning status. Stones are easily identifiable if present.[5] Ultrasound (US) can demonstrate some of these features with an enlarged kidney, loss of tissue architecture, stones and dilated calyces.

Nephrectomy with adjunctive antibiotic therapy is considered the gold standard for treating XGP.[2] For XGP complicated by abscesses, percutaneous drainage is recommended before nephrectomy, for source control and achieving a cleaner operative field.[2]

In the pediatric literature, XGP is exceedingly rare, with only 283 reported cases.[5] It presents atypically with a constellation of symptoms, including abdominal pain, a palpable mass, weight loss, fever, leukocytosis, anemia, pyuria, and thrombocytosis. XGP should be considered in any child presenting with a renal or peri-renal mass or abscess, especially in the context of fever, leukocytosis, or urolithiasis.[5]

To date, only one case report has documented XGP associated with a reno-psoas fistula and migration of a renal calculus into a psoas abscess in adults, with no such cases reported in the pediatric population.[4]

2. Case presentation

A 15-year-old fit and healthy female with right lumbar tenderness and a month of subacute lumbar pain, initially linked to recent COVID-19, presented in sepsis. She had no diagnosis of diabetes mellitus, immunological disorders, immunosuppression, hypertension, recurrent urinary tract infections, renal calculi, or genetic/congenital abnormality. Despite having no genitourinary symptoms, she exhibited reproducible flank and suprapubic pain, tachycardia (120 bpm), a systolic

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blood pressure of 90 mmHg, and a 38-degree fever. Urinalysis showed leukocytes, microscopic hematuria, and nitrates. Elevated inflammatory markers (19 × 109/L, CRP 249 mg/L) and microcytic anemia (Hb 78g/L) were observed, along with urine microscopy confirming pan-sensitive Escherichia coli. She was diagnosed with presumed acute pyelonephritis, receiving antibiotics under pediatric care without immediate imaging.

Despite treatment, the patient's fevers persisted, prompting a four-day follow-up ultrasound (US) revealing a mega-ureter with dilated right renal calyces but no renal pelvis distension. Subsequent CT IVP confirmed a large psoas abscess with intramural calcification. The right kidney appeared enlarged with dilated calyces, reduced parenchyma, poor enhancement, and no hydronephrosis, leading to a provisional diagnosis of xanthogranulomatous pyelonephritis (XGP) with a psoas abscess (Fig. 1.). Renal function remained intact.

Upon surgical consultation, an initial attempt at source control involved percutaneous drain placement into the psoas abscess. During this procedure, an effort was made to insert a ureteric stent for additional source control and kidney evaluation, but contrast failed to reach the renal pelvis from the proximal ureter, and wire placement proved unsuccessful (Fig. 2.).

Inflammatory markers continued to rise, with persistent tachycardia and recurrent fevers, necessitating ICU admission for monitoring and support. Her blood pressure remained stable. After three days of no improvement, a decision was made to proceed with nephrectomy, the delay due to a supra-therapeutic INR resulting from the infection.

The open right nephrectomy revealed a fistulous tract extending from the posterior inferior kidney pole into the psoas muscle and abscess cavity. Despite thorough exploration and washout, no kidney calculus was found. Examination of the specimen showed significant purulent contamination throughout the renal collecting system with large, pus-filled calyces (Fig. 3.).

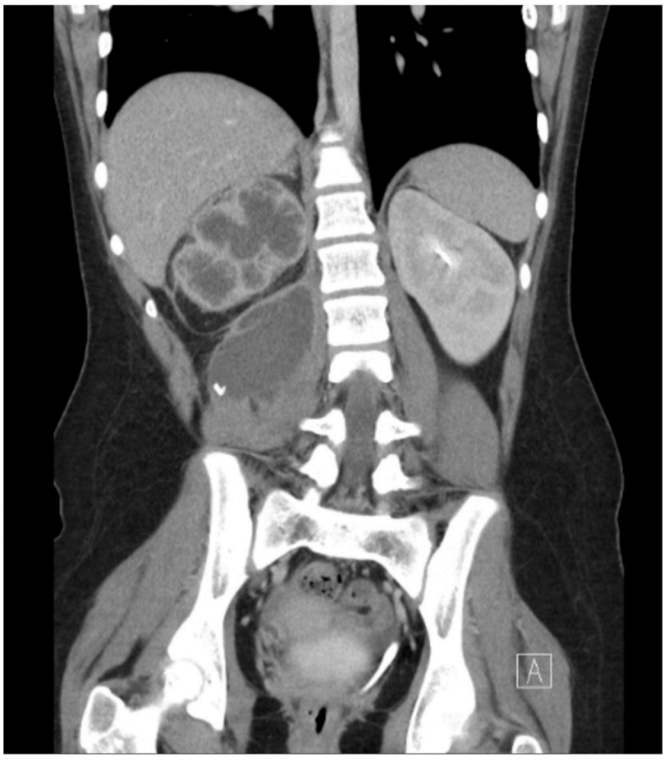

Fig. 1. CT abdomen pelvis multiphase with pre and post IV contrast imaging. Findings: Non-functional right kidney, thickened contracted renal pelvis with hypo attenuating distended calyces give a classical "bear paw" sign indicative of XGP. Calculus dependent within the psoas abscess indicating probable reno-psoas fistula.

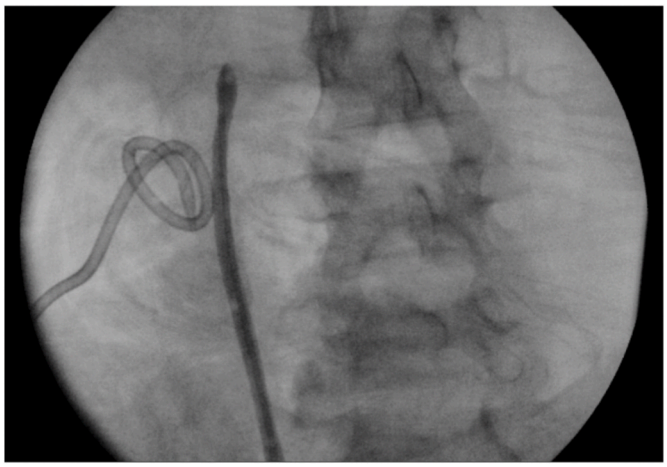

Fig. 2. Intra-operative retrograde pyelogram demonstrating absent contrast flow into right renal pelvis and blind ending proximal ureter unable to be bypassed with guide wire or ureteric catheter. Pigtail psoas drain also present.

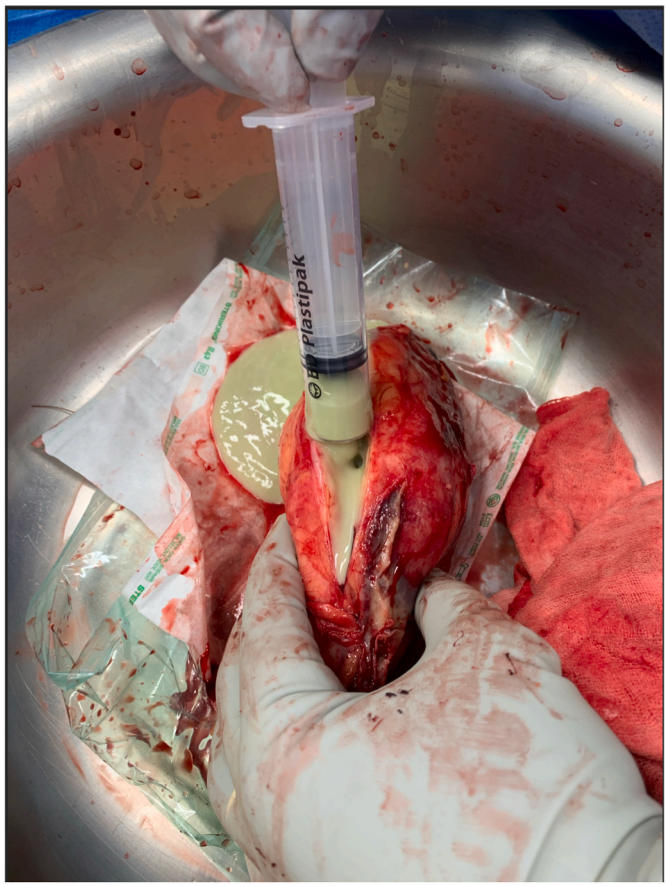

Fig. 3. Intra-operative imaging of specimen with longitudinal incision through lateral border of kidney. Large amounts of purulent exudate aspirated and sent for culture.

The patient had an extended hospital stay, with ongoing fevers and a prolonged antibiotic course. However, she ultimately made a full recovery, was discharged in good health, and experienced no impact on kidney function, with XGP confirmed through histopathology. The histological findings were of fibrous thickening of the renal capsule with loss of parenchyma and glomeruli. There were marked inflammatory infiltrates with granuloma formation, sheets of foamy histiocytes and

neutrophils identified throughout the specimen. There was no evidence of malignancy.

3. Discussion

Our case highlights the challenge of diagnosing XGP due to its nonspecific symptoms, further complicated by concurrent COVID-19 infection, initially misattributing her symptoms and biochemistry. Elevated inflammatory markers, inflammatory-induced anemia, and reactive thrombocytosis were observed. Based on CT imaging, intra-operative findings, and histopathology, our patient had diffuse XGP with complete kidney loss, caused by E. coli, the most common pathogen in XGP.[1]

This case emphasizes CT's crucial role as the gold standard for diagnosing suspected XGP. Initial ultrasound misinterpreted findings as renal obstruction with a hydro-ureter and dilated renal calyces but no hydronephrosis. The reported hydro-ureter was the psoas abscess. While ultrasound provided clues, CT intravenous pyelogram delineated the psoas abscess, calculi, diffuse kidney involvement, the XGP "bear claw sign," and non-functional kidney status. The lack of response to sensitive antibiotics during the initial admission underscores the limitations of medical management, necessitating surgical intervention. Histopathological diagnosis is essential to rule out malignancy, achievable only through resection.[3]

Our patient's condition likely resulted from a small renal calculus obstructing the system due to stricture or physiological narrowing, leading to inflammatory XGP and a fistulous tract to the psoas with subsequent abscess formation. This presentation is unique, as XGP typically arises from a chronic large stone burden like staghorn calculi with additional risk factors for developing XGP. Despite multidisciplinary input on this patient no attributable risk factors, aside from gender, were identified.

We have identified only one similar case in the literature where XGP was associated with renal calculus migration into a psoas abscess, as documented by Singh et al. in a 36-year-old female.[4] No such cases have been reported in pediatric patients.

4. Conclusion

XGP is a rare diagnosis in the paediatric population. This is the first case report of a paediatric patient having XGP with reno-psoas abscess and migration of renal calculus into the cavity. This case report highlights a rare pathology with even rarer complications that should be included in the paediatric and urology differentials.[5] CT is imaging modality of choice and percutaneous source control and nephrectomy should be considered standard management.

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