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Case Report

Xanthogranulomatous pyelonephritis presenting as giant gluteal abscess ^{☆,☆☆}

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ABSTRACT

Xanthogranulomatous pyelonephritis (XGP) is a rare chronic suppurative granulomatous infection of the kidney associated with renal obstruction and progressive renal parenchymal destruction. We present an unusual clinically occult case of extensive right XGP which presented clinically with discharging right gluteal sinus and swollen right posterolateral chest and abdominal walls extending caudally to right gluteus. Contrast enhanced computed tomography of the abdomen and pelvis revealed obstructing right renal calculi, severe hydronephrosis, renal destruction and large (20.7 × 10.2 × 14.7 cm) abscess extending caudally to right gluteus. Histopathology of the specimen was concluded as right xanthogranulomatous pyelonephritis. This case demonstrates how distant an insidious XGP can infiltrate. It also emphasizes the need to have a broad range of differential diagnoses including XGP when presented with a case of gluteal abscess and sinus. Contrast enhanced computed tomography plays a key role in evaluating the cause, extent and complications of XGP and is also useful in pretreatment planning.

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Introduction

Xanthogranulomatous pyelonephritis (XGP) is a rare form of pyelonephritis [1]. In majority of the cases, it is characterized by renal obstruction which induces repeated renal infection and inflammation which if not relieved and treated progresses to chronic renal inflammation. Consequently, in some cases there is renal parenchymal necrosis and destruction with subsequent invasion of adjoining retroperitoneal spaces, structures, and organs [2]. Renal calculus is the most frequently reported obstructing lesion with staghorn calculus contributing half of the cases [3]. We discuss an unusual case of XGP

secondary to obstructing right renal pelvis causing severe hydronephrosis, extensive renal parenchymal breakdown, with extensive infiltration of right retroperitoneal spaces and posterior chest and abdominal wall muscles with formation of giant abscess extending from the right posterior chest wall to right gluteus.

Case presentation

The patient is a 62-year-old male with no significant prior medical history. He presented to the hospital with

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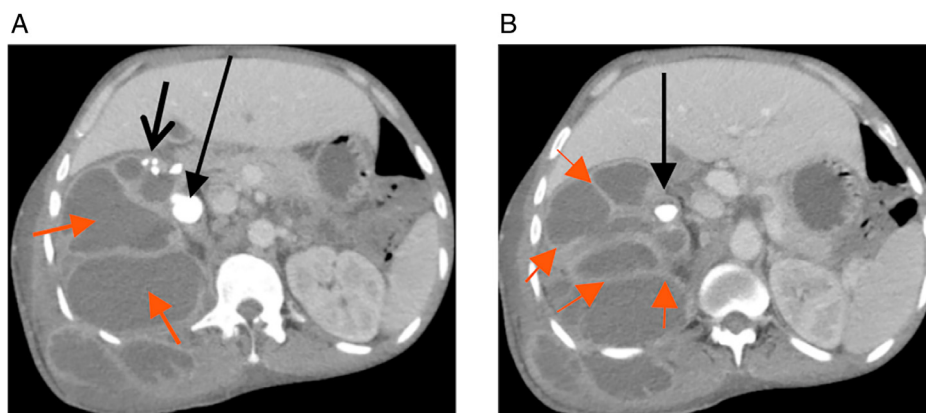


Fig. 1 – A) Contrast enhanced computed tomography of abdomen, axial view: Shows multiple right renal calculi (short black arrow) and 37 mm right renal pelvis calculus (long black arrow) causing severe hydronephrosis [small orange arrows]. (B) Contrast enhanced computed tomography of abdomen axial view: Shows contracted right renal pelvis (long black arrow) and enhancing walls of hydronephrosis (small orange arrows). (Color version of figure is available online.)

progressive swelling on his posterior right side of the chest and abdomen which was of gradual onset over the last 2 months and unintentional weight loss of 6.3%. He also reported some discomfort on the same side but denied pain. He did not have night sweats, fever, chills, or rigors. There was no report of dysuria, hematuria, urgency, or frequency of micturition. A month prior to hospital presentation, he had sought treatment at his local medical facility where he was treated with unspecified antibiotics with no significant resolution of his symptoms. No history of diabetes mellitus, hypertension, or kidney stones. No previous history of admissions or surgical procedures. On the day of admission, he was in fair general condition and not in distress. The blood pressure was 102/71 mm Hg, pulse was 81 beats per minute, temperature was 37.1° centigrade, and respiratory rate was 18 breaths per minute. Pulse oximetry was 95% in room air. Abdominal examination showed swelling and diffuse soft tissue thickening of right lateral and posterior chest and abdominal walls with fullness of right lumbar region. There was pitting edema in right posterolateral abdominal and chest walls and right gluteus. Deep palpation elicited tenderness and ill-defined mass in the right loin, lumbar, and iliac fossa. The margins could not be defined. No guarding, rigidity, and rebound tenderness were elicited. The bowel sounds were present. The hernia orifices were free. Respiratory examination revealed transmitted sounds at right lung base posteriorly. The cardiovascular examination and central nervous systems were normal.

Full blood count showed mild leukocytosis (white blood cell count of 14,100/mm³), anemia of 9.8 g/dL (13.5–17.5 g/dL), and elevated C-reactive protein of 23 mg/L (<3.0 mg/L). Urinalysis demonstrated pyuria (white blood cells of 10) but no red blood cells were detected. Urine culture was positive for *Proteus mirabilis*. Liver function tests were normal. Urea and creatinine were normal.

A chest radiograph done showed right basal atelectasis. Ultrasound of the abdomen demonstrated loss of right renal contour and a large collection which was extending from right subhepatic space to right iliac fossa. The right kidney had multiple renal stones and severe hydronephrosis. On



Fig. 2 – Contrast enhanced computed tomography of abdomen, coronal view: Shows right lower renal parenchymal destruction (small red arrows) with extension of abscess into perinephric and paranephric spaces (orange arrows). (Color version of figure is available online.)

the basis of the above findings, contrast enhanced computed tomography (CECT) of abdomen and pelvis was ordered. CECT demonstrated multiple right renal calculi with 37 mm calculus in right renal pelvis (Fig. 1A) causing contraction right renal pelvis and severe hydronephrosis (bear paw sign) (Fig. 1B). Lower pole renal parenchymal destruction was also seen with extension into perinephric and paranephric spaces (Fig. 2). There was large (21.1 × 12.5 × 13.6 cm) abscess which was involving right posterior chest wall, right psoas muscle, quadratus lumborum muscle, and erector spinae muscle (Fig. 3). The abscess was extending caudally to right gluteus where there was a small sinus (Fig. 4). Multiple reactive

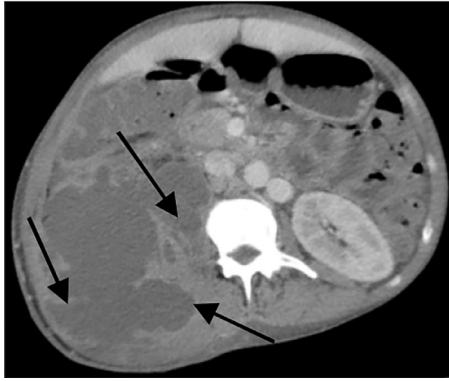


Fig. 3 – Contrast enhanced computed tomography of abdomen, axial view: shows large abscess involving right psoas, quadratus lumborum and erector spinae muscles (black arrows).

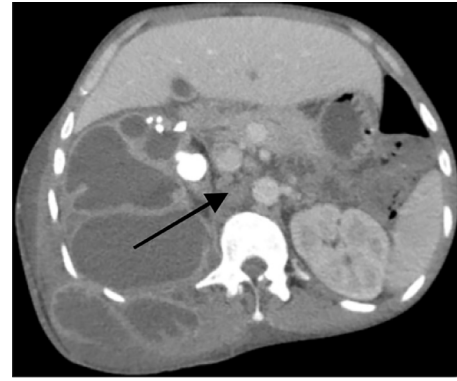


Fig. 5 – Contrast enhanced computed tomography of abdomen, axial view: Shows multiple reactive retroperitoneal adenopathy (black arrow).

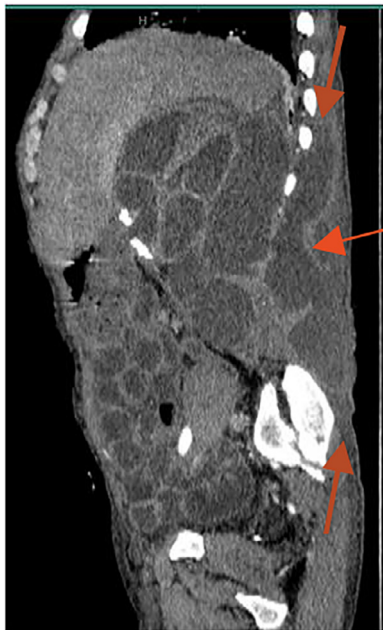


Fig. 4 – Contrast enhanced computed tomography of abdomen, sagittal view: Large right posterior chest wall, abdominal wall and right gluteal abscess extending from level of 9th right posterior rib to lower margin of right sacroiliac joint (orange arrows). (Color version of figure is available online.)

retroperitoneal adenopathy was also present (Fig. 5). The left kidney was normal.

The patient was started on gentamicin and ceftriaxone and later switched to piperacillin and tazobactam after urine culture results were obtained which grew proteus mirabilis. Ultrasound guided percutaneous drainage of the abscess was performed successfully and drained 5.7 L of frank pus. On day 8 of admission, the patient was taken to the operating room for total right nephrectomy and debridement of perinephric and paranephric inflammatory and necrotic material.

The postoperative period was uneventful and the patient was discharged home 5 days after the surgery and he has remained stable 3 months on follow-up.

Discussion

XGP is known for its local and regional spread and extension. Previously described case reports have highlighted how the disease progression and extension is varied. Our present case supports that notion. There have been case reports of XGP extension to the left pleural space, duodenum, colon, and skin [4,5]. Our current case is quite unique because of the magnitude of local, regional and distant invasion, and destruction of tissues with consequent formation of giant abscess. The extensive spread to involve the gluteus, abdominal wall, and chest wall all in one patient has not been reported previously. In our case, the obstructing lesion was a large renal pelvis calculus which caused severe hydronephrosis. While the pathogenesis of XGP is unclear, it is believed that chronic renal obstruction results in chronic infection and inflammation [6]. The disease progresses if the obstruction is not relieved and the infection is not treated. The infection/inflammation can be acute, subacute or insidious. Our case was most likely of insidious progression allowing for the extensive renal parenchymal destruction demonstrated on CECT. Pathologically, XGP has been demonstrated to contain xanthoma cells which are lipid rich macrophages which destroy and replace the renal parenchyma [7]. It is most likely these inflammatory cells with other mediators of chronic inflammation which cause breakdown and necrosis of the adjacent structures including fascial planes and muscles to cause abscess. The case we present is also unique in the sense that there was involvement of both the extensor and flexor muscles of the back. Most of the previously documented case reports involved mainly the psoas muscle [8]. In our case, there was involvement of psoas, quadratus lumborum, and erector spinae group of muscles with extension to gluteus. While it is mind boggling how such large abscess could form without concomitant serious clinical picture, we postulate that the isolated *Proteus mirabilis* may

have been less virulent to elicit systemic illness or there was no induction of systemic inflammatory response syndrome. It could also be explained by delay in seeking medical attention thus allowing the low grade infection to gradually progress. XGP is commonly reported in women and clinically presents with flank pain, fever, and weight loss [9].

Imaging, particularly CECT of abdomen and pelvis plays a critical role in the evaluation, diagnosis, and assessing the extent of XGP. On CECT, the obstructing lesion which is a calculus in about 70% of reported cases of XGP, is depicted. The calyces are dilated, the calyceal walls enhance and renal pelvis is contracted giving the “bear paw sign” which was well demonstrated in this case [10]. The renal involvement of XGP can be diffuse (90%) or focal (10%). CECT can also demonstrate renal cortical destruction, perinephric and paranephric involvement. As in our case the abscess formation in various muscles groups, fistula and sinus tracts can also be elucidated. Differential diagnoses for our case included tuberculous psoas abscess, infective (pyogenic or tuberculous) spondylodiscitis, septic arthritis, or osteomyelitis of spine or iliac bone with abscess formation. CECT was very useful in excluding these pathological entities.

The final diagnosis of XGP is confirmed histologically from biopsy tissue obtained under image guidance or from nephrectomy specimen. The treatment begins with infection and inflammation control with broad spectrum antibiotics and culture sensitive antibiotics. Definitive treatment is surgery which may be total nephrectomy for diffuse XGP or nephron sparing nephrectomy for some forms focal XGP.

The learning points of this unusual case is how extensively insidious XGP can spread to involve distant anatomic structures such as the gluteus with formation of giant abscess along its tract. Lastly, XGP can involve all groups of muscles in the chest and abdominal walls. CECT is very useful in the diagnosis and pre-treatment assessment of XGP.

Patient consent

The patient gave written and verbal consent for this publication and use of images and is available for review.

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