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Postpartum xanthogranulomatous pyelonephritis: A case report

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ABSTRACT

Xanthogranulomatous pyelonephritis (XGP) is seldom seen nowadays due to the aggressive treatment of upper urinary tract infections as well as recent advances in the management of urolithiasis. It has been rarely reported in the peri-partum period. We present a case of XGP without any evidence of renal calculi, manifesting in a 26-year-old previously healthy woman immediately post-partum.

1. Introduction

Xanthogranulomatous pyelonephritis (XGP), a term coined by Schlagenhaufer in the early 1900s, is a rare but serious chronic inflammation of the kidney parenchyma. It often develops secondary to a chronic infection, from a long-lasting obstruction, resulting in an unusual suppurative and granulomatous reaction which can lead to parenchymal obliteration and thus a non-functioning kidney [1]. Calculi, stricture, and tumor all represent a nidus for obstruction. It accounts for less than 1% of all related inflammatory and infectious kidney processes [1]. This process is more likely to occur in females, during the 5th to 7th decade of life [2]. We herein present a case of XGP without any evidence of renal calculi, manifesting in a 26-year-old previously healthy woman immediately post-partum.

2. Case Report

A 26-year-old pregnant G1P1 woman, with no known prior history of febrile urinary tract infection, flank pain, or urolithiasis, presented at 39 weeks of gestation and underwent a normal vaginal delivery without any complications. Her prenatal care was uneventful and two screening urine analyses and cultures done at 10 and 29 weeks of gestation showed no evidence of infection. Throughout all of her prenatal appointments the patient denied any signs of fever or lower urinary tract symptoms. Nevertheless, on the same night of delivery, she developed sinus tachycardia with a maximal heart rate of 142 beats per minute as well as tachypnea associated with dyspnea with a respiratory rate ranging between 20 and 24 breaths per minute. Following that, she developed hypotension with a maximal drop in systolic blood pressure by 30 mmHg that was refractory to fluid boluses, but no fever. Apart from a normally contracted uterus, there were no abnormalities noted in her physical exam.

Laboratory values were all within normal range except for an elevated white count of 32,500/ml. Computed Tomography (CT) angiography of the chest was performed because of suspected pulmonary embolism. The lower cuts of the chest CT showed a huge lesion occupying the right kidney topography (Fig. 1); so a dedicated abdominal and pelvic MRI with Gadolinium was performed revealing a large multiloculated multi-septated cystic mass arising from the upper pole of the right kidney, measuring 19.0 × 15.5 × 16.5 cm. The mass was abutting the right hepatic lobe with no definite infiltration. Mass effect was caused on the aorta, celiac axis, portal confluence, gallbladder and pancreas which were all deviated to the left side (Fig. 2). There was no evidence of any filling defect or obstructing stone within the renal pelvis or right ureter. The differential diagnosis at the time included multilocular cystic nephroma, cystic renal carcinoma or hydatid disease.

She was initially transferred to the Intensive Care Unit for stabilization, where broad-spectrum antibiotic coverage with meropenem and vancomycin was initiated. A few hours later, fever subsequently developed reaching a Tmax of 39.1 °C with slight worsening of her

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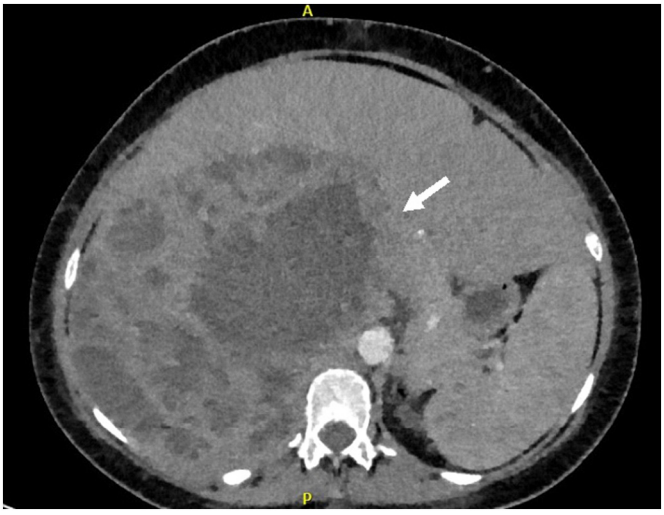

Fig. 1. CT Angiography of the Chest. Lower cuts of the chest revealing a large 19 × 14 cm heterogeneous and enhancing septated and multi-loculated mass (arrow), arising from the upper pole of the right kidney. There is secondary mass effect on the adjacent organs with elevation of the right hemidiaphragm and underlying atelectasis. As initial expression, this could represent a hydatid cyst versus a septated multicystic nephroma.

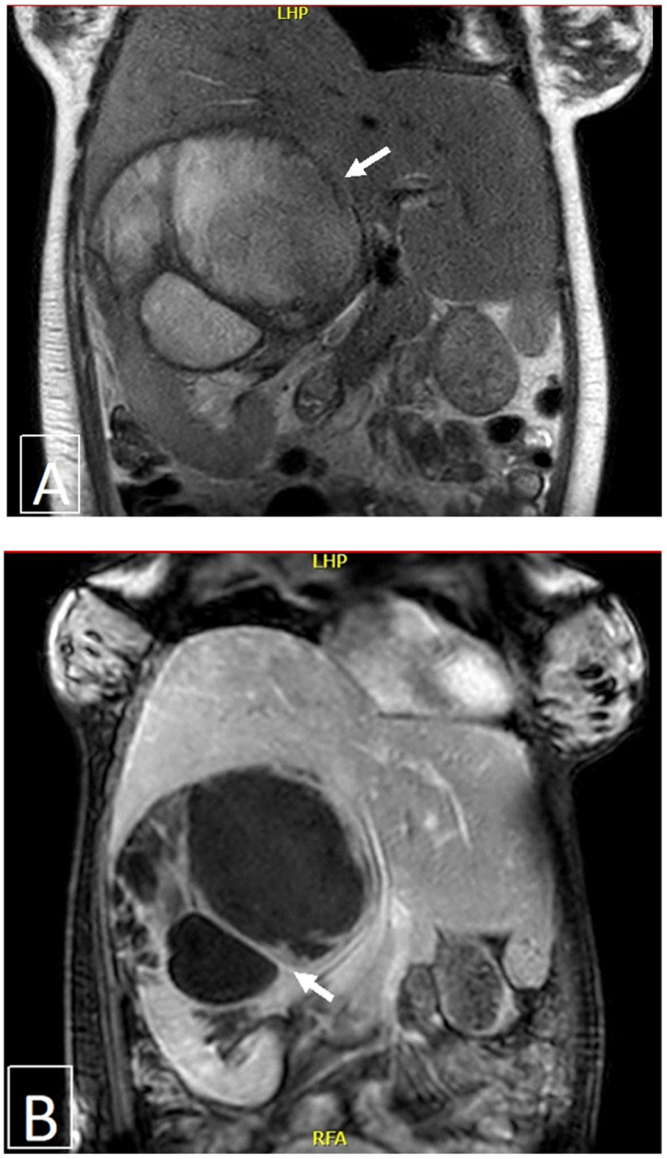

Fig. 2. Magnetic Resonance Image of the abdomen and pelvis with 1.5 T field strength post 20 ml of Dotarem administered intravenously. A: coronal T2W images of the abdomen and pelvis revealing a large multiloculated and multiseptated cystic mass arising from the upper pole of the right kidney, measuring 19 × 15.5 × 16.5 cm in its transverse, AP and craniocaudal dimensions, respectively. The mass is abutting the right hepatic lobe with no definite infiltration. It exerts mass effect on adjacent structures including the aorta, celiac trunk, portal confluence, gallbladder and the pancreas, which are deviated to the left side. There is right perinephric fat stranding and edema. B: T1W coronal image during venous phase showing a large multicystic lesion arising from the upper lobe of the right kidney with wall and septal enhancement post contrast administration. The cysts do not show any solid enhancement or component.

hemodynamics where her lowest mean arterial pressure reached 59 mmHg. It was then that the decision was made to proceed with surgical exploration.

Extended Chevron incision was performed. A huge right kidney was identified, with the ascending colon and duodenum medially retracted. The perinephric tissue was easily dissected anteriorly and caudally; however, it was extremely adherent posteriorly and superomedially. Attempts to elevate the kidney from the psoas fascia resulted in eruption of a large posterior cyst, and a massive amount of purulent foul-smelling grey-brown fluid spilled over the field. After thorough irrigation of the surgical field, nephrectomy was completed, and further cleaning of surrounding suspicious-looking fat tissues was performed. The postoperative period was uneventful. Her hemodynamics improved significantly after resection, and she was discharged 4 days postoperatively with heart rate and blood pressure completely normalized; she was afebrile at the time of discharge. Urine and pus culture, from ruptured intraoperative cysts, grew multi-sensitive *Escherichia coli*.

Grossly, the specimen weighted 1.26 kg consisting of a 17 × 16 × 10 cm kidney. The mass, arising from the upper pole, was tan yellow and soft with cystic areas and areas of necrosis. Histopathological examination of the specimen revealed sheets of lipid-laden macrophages, histiocytes, and multinucleated giant cells associated with acute and chronic inflammation (Fig. 3), involving the renal parenchyma and extending into the perinephric fat and adrenal gland. There was no evidence of hydatid cyst disease. GMS, PAS, and AFB stains were all negative for microorganisms. Such constellations of pathological findings were in accordance with xanthogranulomatous pyelonephritis (XGP). She was then seen 2 weeks after discharge; her vital signs were within normal range and her wound was clean with no evidence of infection.

3. Discussion

Xanthogranulomatous pyelonephritis is a rare inflammatory disease of the kidneys. It is generally associated with an obstructing calculus that leads to urinary stasis followed by a chronic infectious process in the form of a subacute inflammatory reaction. This drives the transformation of parenchymal cells into lipid-laden macrophages known as foamy cells [3]. The most commonly cultured organisms are *Proteus mirabilis* and *Escherichia coli* [4]. In many instances, XGP is closely associated with hepatic lesions. In more than 50% of cases, XGP presents with a marked elevation of liver enzymes [5].

Radiological findings on CT scan illustrate an enlarged kidney with several hypodense egg-shaped areas of fluid collection usually associated with renal calculi [6]. Radiological grading is dependent on the extent of inflammatory changes depicted on imaging [7]. Stage 1 is when inflammatory changes are confined to the renal parenchyma. Stage 2 is when those changes extend into the perinephric fat; while stage 3 is when the inflammation extends through the retroperitoneum surpassing Gerota's fascia [7]. Although radiological findings can be suggestive of XGP, the diagnosis is in essence a pathological diagnosis that comprises of an enlarged kidney size with several scarred areas and an

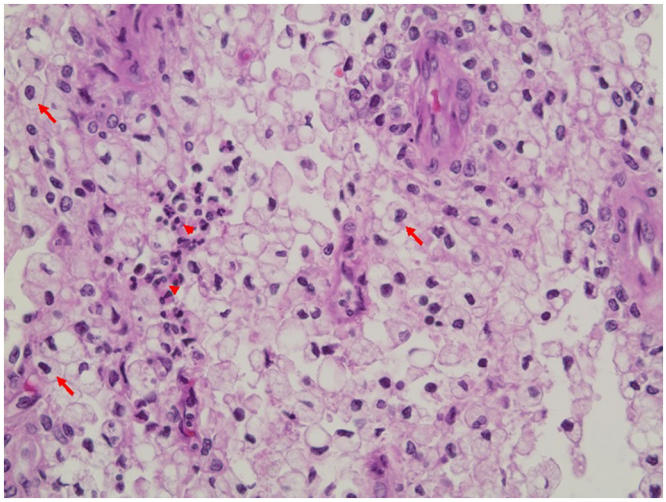

Fig. 3. Histological cross-section (×100) of the renal mass with hematoxylin and eosin staining. The lesion is composed of sheets of predominate histiocytes of abundant pale cytoplasm and larger euchromatic nuclei without prominent nucleoli (arrows), multinucleated giant cells (arrowheads), in a background of acute and chronic inflammation involving the renal parenchyma and extending into the perinephric fat. CD68 and Cytokeratin AE1/3 immunohistochemistry staining is respectively positive and negative; hence, the absence of malignant cells.

adherent renal capsule. Histological findings reveal the presence of lipid-laden macrophages diffusely scattered within the renal parenchyma [3].

Pregnancy by itself is known to have an immunomodulatory effect on the normal physiological functioning of the body which has been proven across different disciplines. One of the immunomodulations that occur during pregnancy is the response to bacterial infections [8]. As such, this may have resulted in the abnormal and amplified immunological response to the urinary pathogen, leading to this rapid septic deterioration. The association of XGP with pregnancy is a very rare yet lethal condition. Prompt investigation should be initiated upon a suggestive history of recurrent urinary tract infections (UTIs) or stone formation [9]. Typical treatment of XGP involves a partial or total nephrectomy depending on the parenchymal involvement as well as the surgeon's expertise [10]. With the added complexity of the situation caused by pregnancy, timing of the operation is of paramount importance due to the known implications of surgical intervention on fetal development and risk of premature delivery or fetal demise, particularly in the first and third trimesters, respectively. If diagnosed in the aforementioned trimesters, it is recommended to either place a nephrostomy tube, with or without antegrade double J ureteral stent insertion as reported by Figueroa et al. [11], or insertion of a ureteral stent in a retrograde fashion, as reported by Ferreira et al. [9]. Both are suitable alternatives awaiting definitive surgical treatment. Following nephrectomy of the involved kidney, biannual to yearly imaging of the contralateral kidney is proposed, depending on the clinical suspicion of urinary tract infections or nephrolithiasis, with early aggressive management to preserve a predisposed contralateral solitary kidney.

To our knowledge, this is the first case of XGP unrelated to a documented history of neither kidney stones nor recurrent UTIs diagnosed and managed immediately post-partum, in an otherwise previously healthy 26-year-old woman. As opposed to the previous peri-partum reports, our patient presented with acute clinical deterioration the night following her delivery. This is an atypical presentation of this disease. Hence, the authors recommend consideration of XGP as a differential diagnosis of a renal mass, especially in the presence of sepsis.

Contributors

Jose M. El-Asmar contributed to the literature review, and edited the final version of the manuscript.

Rayan Ghanem contributed to the literature review.

Rashed Ghandour contributed to the literature review.

Eliane Al-Halabi contributed to the literature review and writing of the initial manuscript.

Jad A. Degheili edited the final version of the manuscript.

All five authors contributed to the writing of the manuscript, and approved the final version prior to submission.

Conflict of Interest

The authors declare that they have no conflict of interest regarding the publication of this case report.

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Patient Consent

Written Informed consent was obtained from the patient for publication of this case report and the accompanying images.

Provenance and Peer Review

This case report was peer reviewed.

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