CASE REPORT

Diffuse xanthogranulomatous pyelonephritis with psoas abscess in a pregnant woman

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We report the first case, to our knowledge, of xanthogranulomatous pyelonephritis (XGP) with psoas abscess occurring during pregnancy. A 37-year-old woman in the third trimester of pregnancy presented with low back pain and a fever. From sonographic features, a multidisciplinary team decided to perform computed tomography of the abdomen with contrast agent injection, which strongly suggested diffuse XGP of the left kidney with a psoas abscess. Cesarean section at 32 weeks was followed by extended nephrectomy. Pathological examination of the operative specimen confirmed the diagnosis. Outcomes were favorable in the mother and baby. The diagnosis and treatment of XGP during pregnancy are discussed.

Key Words: kidney, xanthogranulomatous pyelonephritis, computed tomography, pregnancy, nephrectomy

Introduction

Diffuse xanthogranulomatous pyelonephritis (XGP) is a distinctive form of chronic renal infection with phlegmon and destruction of the parenchyma, which is replaced by xanthomatous cells. The cause is unknown. The typical patient is an older woman with a history of chronic urinary symptoms. We are not aware of previous reports of diffuse XGP with local spread manifesting as psoas abscess in a pregnant woman. We report a case at the third trimester of pregnancy. The contribution of imaging studies to the diagnosis is discussed, as well as the treatment.

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Case report

A 37-year-old woman at week 30 of her sixth pregnancy was admitted for lower back pain and fever. She had no significant history of disease or surgery. She reported a severe decline in general health over the last weeks with marked asthenia and weight loss of 6 kg during the pregnancy. For the last few days she had been experiencing dysuria with a burning sensation on micturition. Findings were unremarkable from the gynecological examination. The left flank was tender to palpation. Urine microbiology disclosed *Escherichia coli* infection. Blood tests showed leukocytosis and severe inflammation (erythrocyte sedimentation rate, 80 mm/h; and C-reactive protein level, 100 mg/L). Blood cultures were negative and renal function tests were normal. Appropriate antibiotics were given. By ultrasonography the left kidney was enlarged and contained multiple atypical cyst-like images filled

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with thick heterogeneous hypoechoic material and separated by fleshy septa. The images suggested hydropyonephrosis, but no obstacle was seen. Superinfection of a renal tumor was considered the most likely diagnosis. A multidisciplinary team decided that the severity of the clinical and ultrasound features warranted computed tomography (CT) centered on the kidneys before and after injection of an iodinated contrast agent. The left kidney was enlarged and remodeled, with intraparenchymatous pseudocysts containing thick grainy material. The pseudocysts were about 20 HU in density, without postcontrast enhancement, and contained free-floating stones. Peripheral contrast enhancement was noted, as well as dilation of the renal cavities and infiltration of the perinephric fat. The necrotic loculations extended to the left psoas muscle, indicating spread to adjacent tissue, Figure 1. The features of renal phlegmon suggested diffuse XGP rather than a tumor, although this last possibility was not confidently ruled out. Needle aspiration of the psoas muscle under ultrasound guidance retrieved purulent fluid. Smears and cultures were negative for organisms, suggesting an effect of the antibiotics. No foam cells were found. The risk of maternal and fetal complications prompted cesarean section at 32 gestational weeks followed by extended left nephrectomy. A large perirenal phlegmon was removed and the psoas abscess was drained.

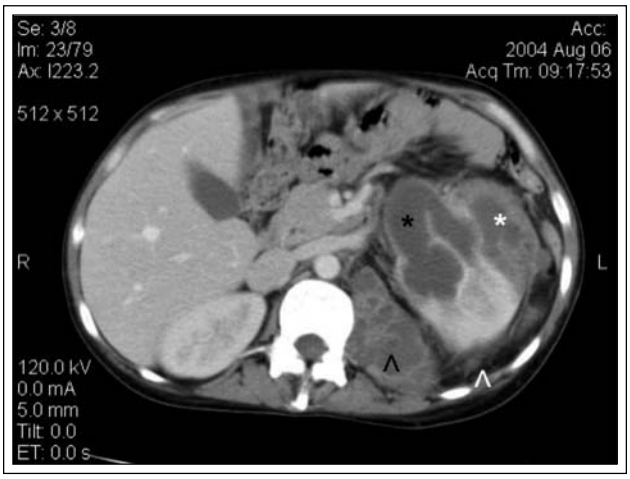

Figure 1. Computed tomography of the kidney after contrast agent injection: enlarged left kidney with hydropyonephrosis (black asterisk), a multilocular intraparenchymatous pseudocyst (white asterisk), infiltration of the perirenal fat (white arrow), and an abscess in the adjacent ipsilateral psoas muscle (black arrow).

Pathological studies showed pyelonephritis with perirenal phlegmon and psoas abscess complicating chronic XGP. Histology disclosed granulomatous tissue composed of lymphocytes, plasma cells, polymorphonuclear cells, and clusters of lipid-laden foamy macrophages. The subsequent course was uneventful in the mother and baby. Clinical follow-up was 2 years at the time of this writing.

Discussion

Xanthogranulomatous pyelonephritis was first described by Schlagenhaufer in 1916. This rare form of low-grade chronic renal infection is characterized by progressive destruction of the renal parenchyma. The cause is unknown. Women are predominantly affected (in a 3:1 ratio), usually in mid-life (50-60 years) and after a long history of recurrent urinary tract infection or urinary stones.[1] Involvement of both kidneys is exceedingly rare.[2] XGP occurs as two variants, a focal form that mimics a tumor and a diffuse form that contributes 90% of cases. Gross examination shows an enlarged kidney with an irregular surface. The histological hallmark is replacement of the renal parenchyma by lipid-laden foamy macrophages in combination with an inflammatory granuloma and a lymphoplasmacytic infiltrate. Investigations may point to inflammation and infection, but these findings are not specific. Cholestasis without jaundice may help to suggest the diagnosis.[1] In patients with urinary tract infection, *E. coli* and *Proteus mirabilis* are the most common organisms.[1] The clinical symptoms are often inconspicuous and nonspecific, consisting of a fever, a decline in general health, and insidious lower back pain. In advanced forms, renocolic fistula, pyelocutaneous fistula, or a psoas abscess may be the presenting manifestation.[3-5] However, development of a psoas abscess related to XGP is exceedingly rare: thus, we are aware of only ten cases with imaging data reported to date, five in adults and five in children.[5-7] One of the adult cases was diagnosed several years after nephrectomy. Psoas spasm was the presenting symptom in half the cases. Only three cases of simple XGP during pregnancy have been reported.[3,8,9] Termination of pregnancy was required before nephrectomy in one patient. In another patient, nephrectomy at 16 gestational weeks was followed by favorable long-term outcomes in the mother and infant. The remaining patient had previously donated a kidney and underwent partial nephrectomy during the second trimester of pregnancy. Her renal function remained normal and she was able to carry the

pregnancy to term. We are not aware of previous reports of XGP with psoas abscess during pregnancy. Our patient was successfully treated by extended nephrectomy after cesarean section at 32 gestational weeks. Outcomes were favorable in both the mother and the baby.

Xanthogranulomatous pyelonephritis is difficult to diagnose. Ultrasonography findings are non specific. CT, in contrast, strongly suggests the diagnosis and provides information on local and regional spread, as illustrated by our case.[6] When performed with caution, CT is safe after 14 gestational weeks. The neonate should be screened for hypothyroidism related to iodine exposure. Although the best time for surgery during pregnancy remains controversial, there is widespread agreement that the risk to the fetus is lowest during the second trimester. Surgery is a stressful event and may therefore induce birth defects during the first trimester and premature labor during the third trimester. However, the disease poses a risk to the mother and fetus, most notably via the infectious process, which cannot be controlled by pharmacotherapy alone. In addition, although CT findings are highly suggestive, they may simulate renal clear-cell carcinoma, which also may be present.[10] Histology is needed to establish the definitive diagnosis. Postponing the histological diagnosis until the end of the pregnancy may put the mother at undue risk for tumor progression. Although needle aspiration (as in our patient) or percutaneous needle biopsy can be performed during pregnancy, these techniques are helpful only when they retrieve typical lipid-laden foamy macrophages. Management decisions should be made by a multidisciplinary team including radiologists, urologists, and obstetricians. Based on the age of the fetus and the wishes of the mother, the risk to the mother and fetus associated with waiting until delivery occurs must be weighed against the risk to the baby associated with premature delivery via cesarean section before nephrectomy. The second option produced favorable outcomes in the mother and baby in our case.

Conclusion

Diffuse XGP is an exceedingly rare pattern of chronic pyelonephritis, and spread to the psoas is even less common. We report the first case of this combination occurring during pregnancy. Management by a multidisciplinary team is crucial in this situation. Histology of the nephrectomy specimen is the only means of establishing the definitive diagnosis. □

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