Clinical Image

Journal Of Clinical & Medical Images Case Reports

Open Access, Volume - 3

Xanthogranulomatous pyelonephritis: An inconspicuous cause of growth impairment

Derakhshan D*; Safari M, Farahangiz M; Mohammadzadeh S

Shiraz Nephro-Urology Research Center, Shiraz University of Medical Sciences, Shiraz, Iran.

Received Date : November 22, 2023
Accepted Date : December 08, 2023
Published Date : December 15, 2023
Archived : www.jcmimagescasereports.org

***Corresponding Author:** Dorna Derakhshan, Shiraz Nephro-Urology Research Center, Shiraz University of Medical Sciences, Shiraz, Iran.
Email: Derakhshad@sums.ac.ir

Abstract

Xanthogranulomatous pyelonephritis (XGP) is a rare and severe form of kidney infection primarily affecting adults with low incidence among children. Most children are symptomatic with fever, recurrent urinary tract infections and obstructive uropathy. We present an incidental finding of XGP in an apparently healthy 5-years-old boy without noticeable clinical findings who was incidentally diagnosed during evaluation for growth impairment.

Introduction

Xanthogranulomatous pyelonephritis (XGP) is a rare and severe form of kidney infection that primarily affects adults. Its occurrence in children is extremely uncommon, and less than 300 pediatric cases have been reported. XGP is characterized by the presence of yellowish nodules or abscesses in the renal parenchyma. The exact cause of XPG in children is not fully understood; however, it is believed to be a result of chronic urinary tract infections and obstructive uropathies. The clinical manifestations of XPG are nonspecific, including abdominal pain, unexplained fever, weight loss, or a palpable renal mass. In this article, we report a pediatric case of XPG who had no specific symptoms and was diagnosed in an incidental abdominal sonography.

Clinical-description

An apparently healthy five years old boy was being investigated for retarded growth. His weight and Height were 13.5 kg and 97 cm, respectively. His physical examinations were all normal. He was afebrile with a normal abdominal examination. In the primary lab tests, he had microcytic anemia (hemoglobin level:9.6 mg. dl, MCV: 62.3 fl, RBC count: 5.17 million/mm3), mild leukocytosis (WBC: 12700, Neutrophil: 9700) and normal platelet count. Biochemistries were all within normal limits (serum creatinine: 0.5 mg/dl). Urinalysis showed pyuria (WBC 20-25), and urine culture was negative. The Erythrocyte sedimentation rate (ESR) was significantly elevated at 110. Due to the abnormal urinalysis, he underwent an abdominal sonography, which reported an abnormal shape of the left kidney with varying size stones up to 14 mm and varying size cysts and pyonephrosis. The right kidney and the rest of the findings of sonography were normal. Consequently, a contrast enhanced abdominal CT scan revealed significant dilatation of left kidney calyces with multiple cysts and varying sized stones in favor of XGP. In delayed films, the left kidney had no excretory function. (Figure 1).

Broad spectrum IV antibiotic therapy was started, and he underwent a left nephrectomy. Figure 1 reveals the gross and microscopic appearance of the kidney. The patient proceeded well post operatively and was kept on a seven-day course of antibiotic therapy with meropenem and vancomycin and was later discharged from the hospital. Subsequent follow-up visits up to 6 months were uneventful.

Discussion

XGP compromises less than 1 % of chronic pyelonephritis cases and is extremely rare in children. The disease course is characterized by the destruction and replacement of kidney parenchymal tissue with lipid-laden macrophages [1, 2]. Most of the cases are associated with urinary tract infections or obstructive uropathies. Patients generally present with several nonspecific symptoms, including fever, flank pain, flank mass, weight loss, and symptoms of urinary tract symptoms [3]. The distinct point of our case was that he had no history of previous urinary tract infections, unexplained fever, or renal colic. The only clinical point that persuaded the parents to seek medical help was inadequate weight gain. All other previously reported cases had significant symptoms [4, 2, 1, 5, 3].

The exact pathogenesis of the disease is still undetermined, but reported predisposing factors include recurrent urinary tract infection, urinary calculi, and obstructive uropathies [2],

Citation: Dorna Derakhshan. Xanthogranulomatous pyelonephritis: An inconspicuous cause of growth impairment. J Clin Med Img Case Rep. 2023; 3(6): 1601.

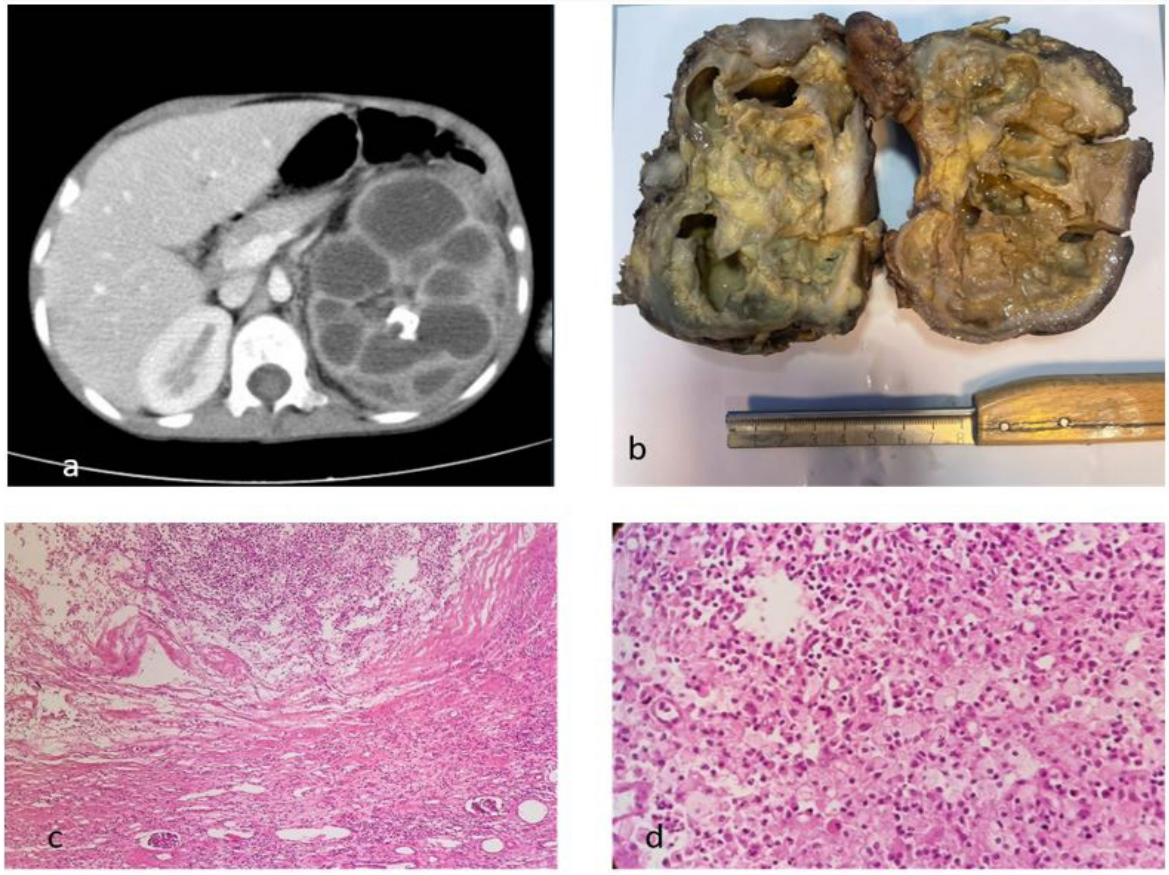

Figure 1: *Radiologic, gross and pathologic findings. contrast enhanced abdominal CT scan revealed significant dilatation of left kidney calyces with multiple cysts (bear's paw sign) and varying sized stones in favor of XGP (a), The dilated renal pelvis is filled with a large amount of pus material and necrotic debris. The walls of dilated calyces and renal pelvis are thickened with multiple yellow nodules. The renal cortex is largely atrophic. (b) Sections through the yellow nodules show replacement of renal parenchyma with foamy histiocytes, occasional multinucleated giant cells, and inflammatory cells. The inflammatory cells are composed of lymphocytes, plasma cells, and neutrophils. Lymphoid aggregates are also noted. (c : 40x stain, d: 400x H&E stain).*

but our patient had neither past history of urinary tract infections nor experienced renal colic.

None of the laboratory tests and radiological features are neither sensitive nor specific for the diagnosis of XGP [2]. The presence of leukocytosis, anemia, elevated ESR, CRP, and pyuria are the most abundant findings. A urine culture may be positive, and the most common cultured organisms include Proteus and E.coli [4, 2, 3] among the mentioned investigations. A CT scan has the utmost modality for identifying XPS and is essential for the evaluation of disease extension. The findings include an enlarged kidney with poor or absent function with concomitant urinary calculi [2] but sometimes may be misdiagnosed due to displaying similar neoplastic features with Wilms tumor, renal cell carcinoma, or renal tuberculosis. The confounding radiologic results lead to misdiagnosis and delay in treatment. Accordingly, some of the previously reported pediatric cases were suspected to be tumoral processes consistent with their radiologic findings [2,1] The definite diagnosis of XGP is only achieved by histopathologic examination [2, 3].

The mainstay of treatment includes antibiotic therapy, open radical nephrectomy, and resection of affected neighboring structures, as most cases are discovered in the late stages of the disease [1, 3]. XGP has seldom occurred in children and is easily misdiagnosed. Early diagnosis and prompt treatment play a crucial role in minimizing mortality and morbidity [2, 3]. The challenge of our case was being mostly asymptomatic and afebrile, which led to the extensive progression of the infection.

Source of funding: The work did not receive any funds from any source.

Ethics approval: The study was approved by ethical committee of shiraz university of medical sciences.

Consent: written informed consent was obtained from patients' parents for publication of this case report and accompanying images.

Authors contribution: DD conceptualized the study, DD, AD and MS planned and performed diagnosis and management and collected medical records. MF and SM performed histopathologic analysis and prepared microscopic images. All authors read and approved the final manuscript.

Declaration of competing interest: All authors have declared

that no competing interest exist.

Acknowledgement: the authors would like to thank patient's parents for allowing to use his medical information for medical purpose.

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