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Xanthogranulomatous Pyelonephritis Revealing a Pyelo-ureteral Duplication in a Child

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ABSRACT

Xanthogranulomatous pyelonephritis (PXG) is a severe and atypical form of chronic renal suppuration. It is characterized by the destruction of the renal parenchyma which is then replaced by a granulomatous tissue made up of a xanthomatous proliferation of foamy histiocytes. PXG can be diffuse or localized at the pole of the kidney. It is rare in children than adults. We report the case of a 3-year-old girl admitted with a urinary tract infection with an abdominal mass on ultrasound to find a pyonephrosis of a ureteral duplicity during the operation. An upper polar nephrectomy was performed. The pathological examination confirmed the diagnosis of xanthogranulomatous pyelonephritis in a double urinary system, which is an extremely rare association in childhood.

KEYWORDS

Pyelonephritis; Xanthogranulomatous; Suppuration; Chronic

INTRODUCTION

Xanthogranulomatous pyelonephritis (PXG) is a severe and atypical form of chronic renal suppuration. It is characterized by destruction of the renal parenchyma [1]. PXG can be diffuse or localized to a pole of the kidney. It is rare in children. The etiopathogenesis mechanisms remain obscure despite the many hypotheses put forward. PXG often remains a histological surprise for the surgeon [2]. An even rarer association is the presence of PXG on a double urinary system.

CASE

A 3-year-old girl is hospitalized for a right lumbar arch associated with a fever of 39˚C. Examination of the abdomen finds swelling of the right lumbar fosse that is fluctuating, painful with lumbar contact. The biological assessment shows a hyper leukocytosis at 12 elements/mm^3 - 13 elements/mm^3, a hemoglobin level at 7.7 g/dl, a C-Reactive Protein (CRP) at 144 µg/l. The ECBU notes a leucocyturia at 80 elements/mm^3 with a negative culture. On the renal ultrasound, there is pyelocalyx dilation, presence of a heterogeneous echogenic formation. The diagnosis of right pyonephrosis with perinephretic abscess on a kidney malformation is retained on the CT scan.

An upper right nephro-polar is performed. The pathological examination concludes with alithiasis diffuse right PXG on pyelo-ureteral duplicity complicated by a

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perirenal abscess. Antibiotic therapy is continued for three weeks. The final evolution is favorable. No contralateral recurrence was observed. The follow-up is currently of 18 months. This is shown in the Figure.

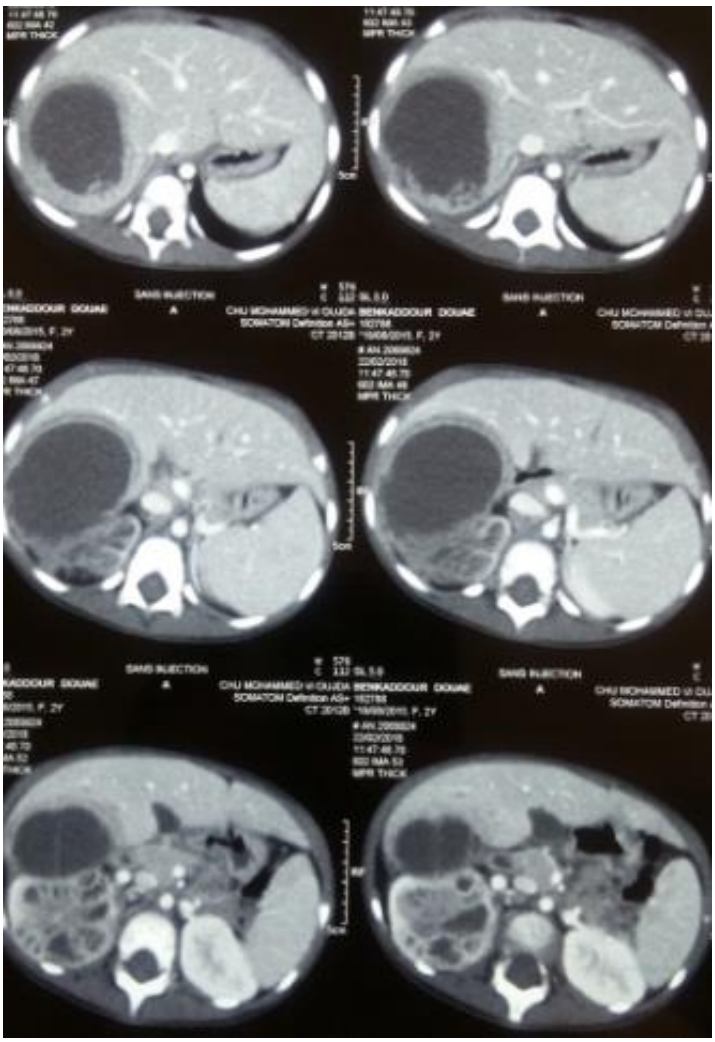

Figure: Pyelo-ureteral duplication in a child.

DISCUSSION

PXG is a special form of chronic kidney infection, characterized by the destruction of the renal parenchyma and its replacement by lipid-laden macrophages called xanthomatous cells [1]. It is most often diffuse, but can remain localized and take on a deceptive pseudo-tumor appearance posing a diagnostic problem with other renal masses, in particular nephroblastoma [3]. It affects both sexes with a large female predominance. Unilateral damage is the rule in children. It occurs in 60% to 75% of cases before the age of 5 years. Malformatif uropathies are responsible for obstruction in about 10 to 35% of cases after renal lithiasis [4]. Namely Pyeloureteral duplication which is defined as a complete or incomplete duplication of the upper excretory pole with two ureters which meet at a variable level [5] and constitutes a main factor favoring PXG in children according to Loffroy R, et al. as our clinically patient the urinary signs are variable and the incidence of urinary tract infection is 52% to 100% of the cases described in the literature [6], this is the case of our patient, Inflammatory anemia and hyperleukocytosis are found in most series and renal function is generally preserved. PXG has several radiological aspects, on ultrasound it is a large kidney with multiple hypoechoic areas with foci of necrosis, Computed tomography usually shows a reshaped mass of low density, with a characteristic “bear's paw” appearance [7]. The

histopathology of PXG lesions always shows inflammatory lesions of an acute or chronic character and huge macrophages loaded with lipids appearing as vacuoles [5]. Due to the rarity of this association of pyelo-ureteral duplicity and PXG, the diagnosis has only been made intraoperatively. Therefore it is important to suggest the diagnosis of PXG in any child who presents a renal mass or a renal abscess or peri - renal. The treatment of PXG is both medical and surgical [7]. In the diffuse form, total nephrectomy, sometimes enlarged to the peri - renal tissues, constitutes the treatment of choice. Indeed, the diffuse and irreversible nature of the lesions makes partial excision impossible and always leads to a total nephrectomy, in our case we left for an upper polar nephrectomy given the pyelo-ureteral duplicity of the patient. The evolution of the PXG after total or partial nephrectomy is excellent [6]. No case of recurrence on the contralateral kidney or the remaining renal stump has been described; the prognosis of PXG is excellent, healing is the rule [7].

CONCLUSION

Xanthogranulomatous pyelonephritis is a rare form of chronic pyelonephritis. Its symptomatology is little different from other chronic kidney diseases. However, only the histopathological analysis of the operative piece can make the diagnosis certain.

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