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CASE REPORT

Xanthogranulomatous Pyelonephritis: A Case Report and Review of the Literature

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Abstract

Xanthogranulomatous pyelonephritis (XGP) is a rare infectious urological emergency. It is a severe form of chronic pyelonephritis, often associated with urinary lithiasis and recurrent urinary tract infection. It can progress to progressive renal destruction. We report the case of a 23-year-old man, presenting for two years with left lumbar pain, associated with intermittent fever. Imaging revealed a 'Bear's Paw Sign' upstream of a left renal pelvic stone on Uro-Tomodensitometry and a non-functioning left kidney on renal scintigraphy. He underwent emergency percutaneous nephrostomy followed by extended nephrectomy one month later. Pathological analysis confirmed the diagnosis of XGP. In this case, we discuss the epidemiological, clinical, radiological and therapeutic aspects of this rare condition.

Keywords: Xanthogranulomatous pyelonephritis; renal pelvic stone; Chronic urinary tract infections; Nephrectomy

1. Introduction

Xanthogranulomatous pyelonephritis (XGP) is a particular form of chronic renal suppuration characterized by the destruction of renal parenchyma and its replacement by granulomatous inflammatory tissue rich in foamy macrophages or "xanthomatous cells" (1). First described in 1916 by Schlagenhaufer, XGP remains a rare entity. Its pathophysiology has not been fully elucidated, and it occurs when chronic and/or recurrent renal infections coexist with excretory tract obstruction, whether due to renal lithiasis (particularly coralliform calculi) or other obstructive uropathies (2). Diagnosis is often difficult, as the symptoms are insidious and unspecific: low back pain, prolonged fever, altered general condition.

We report a case of histologically documented XGP after enlarged nephrectomy in a 23-year-old adult at the Yalgado OUEDRAOGO University Hospital Center (CHU-YO).

2. Case Presentation

K. I., a 23-year-old patient with no known pathological history, consulted our department for febrile left low back pain that had been evolving intermittently for 02 years. On admission, his general condition was WHO stage II, temperature 38.7; blood pressure 114/77 mmHg, heart rate 80 beats per minute and respiratory rate 18 cycles per minute. Physical examination revealed a large kidney.

A Uro-Tomodensitometry revealed a swollen left kidney with hypodense cavities surrounded by partitions giving a "Bear's Paw Sign" appearance (figure 1) upstream of a 14 mm (800 HU) renal pelvic stone and multiple 9 ; 8 and 7 mm stasis calculi. This was suggestive of left-sided xanthogranulomatous

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pyelonephritis upstream of a renal pelvic stone. The contralateral kidney was morphologically normal.

The biology revealed a hyperleukocytosis of 17,860 elements/mm3, a microcytic hypochromic anemia and a hemoglobin level of 10.5 g/dl. Blood glucose was normal at 4.53 mmol/l and creatinine 115.35 umol/l; blood ionogram normal. Urine cytobacteriological examination (UCE) was normal.

He underwent an echo-guided left nephrostomy, which yielded 1300 ml of pus. A sample was taken for cytobacteriological study. Probabilistic bi-antibiotic therapy with Ceftriaxone + Amykacin was administered for 04 days, and subsequently adapted to the antibiogram of the cytobacteriological examination of the pus, which isolated Klebsiella pneumoniae. A renal scintigraphy showed renal function at 9.3% on the left and 90.7% on the right. The patient was admitted one month later for left nephrectomy.

Under general anaesthesia, we performed a left lumbotomy under the 12th rib; this was a difficult extended nephrectomy (figure 3) due to multiple peri-renal adhesions creating a veritable retroperitoneal shield. The postoperative course was straightforward, and the patient was discharged on the 5th postoperative day.

The anatomopathological study of the nephrectomy specimen confirmed the diagnosis of XGP by showing a renal parenchyma largely reworked by inflammation with areas of suppurative necrosis, a cortex comprising fibrosed glomeruli in the shape of "sealing bread" associated with tubules of pseudo-thyroid appearance. The interstitial tissue was the site of a severe polymorphic infiltrate consisting of lymphocytes, plasma cells, foamy histiocytes and polymorphonuclear cells, very often altered (Figure 4a and 4b).

The patient was seen again at 3 and 6 months, with no complaints.

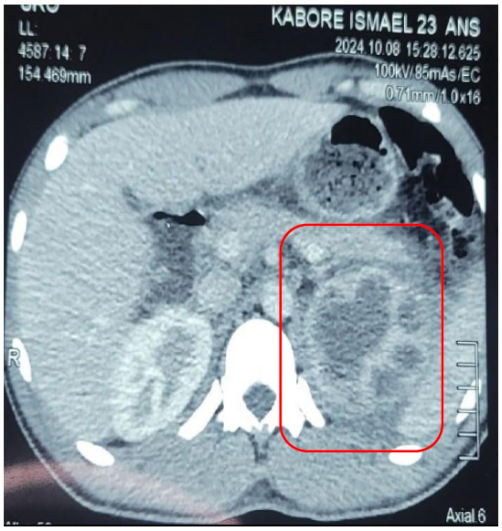

Figure 1. *Frontal section of injected CT scan showing 'bear's paw sign' in orange frame*

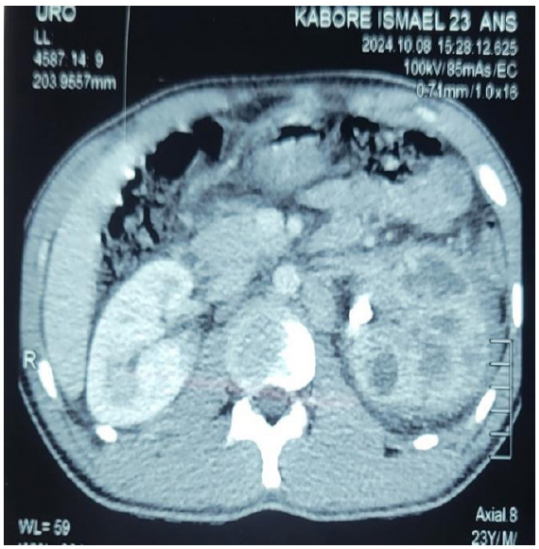

Figure 2. *Frontal section of injected CT scan showing a hypertrophied kidney with a downstream left renal pelvic stone.*

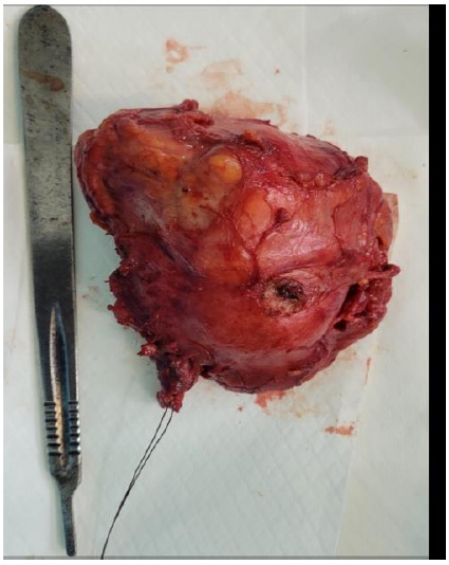

Figure 3. *Left total nephrectomy specimen.*

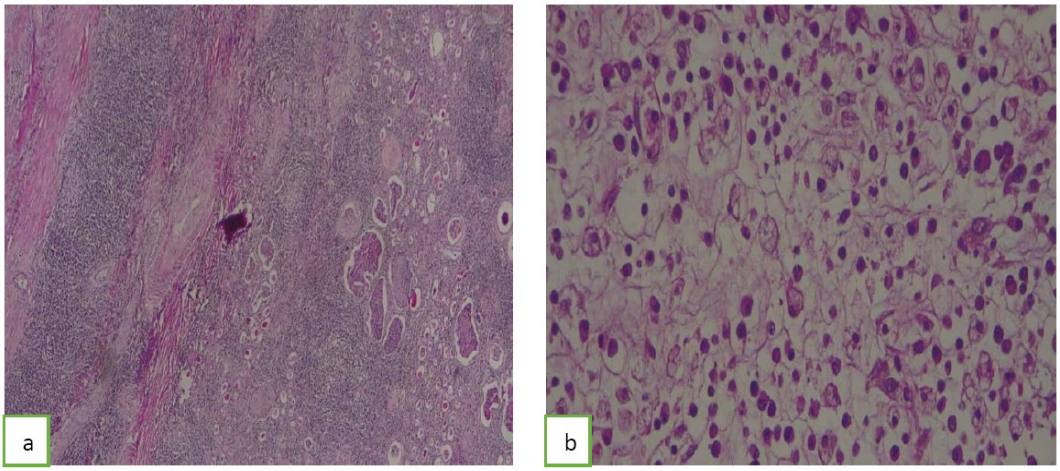

Figure 4. *Histology of the left nephrectomy specimen showing a XGP appearance (a: magnification 40; b: magnification 100).*

3. Discussion

Xanthogranulomatous pyelonephritis is a rare condition responsible for chronic suppuration of the kidney. Its incidence is estimated at 1.4/100,000 inhabitants, representing 0.6 to 1.4% of pyelonephritis diagnosed on imaging (2). It predominantly affects women between the ages of 40 and 60, often in a context of low socio-economic status or delayed management of urinary lithiasis (1,2). In our case, the patient was a young man of 23 years of age, which illustrates the possibility of involvement outside the usual epidemiological profile.

From a etiopathogenic point of view, several factors are involved in the initiation of the xanthogranulomatous process, the main one being chronic obstruction of the excretory tract, most often due to lithiasis, as in our patient's case. This obstruction may also result from ureteral stenosis of specific infectious origin, such as tuberculosis or bilharzia, or from stenosis of the pyeloureteral junction secondary to uropathic malformation, or even more rarely from a tumour of the excretory tract (3,4). In Tunisia, Hamza et al. found a lithiasis in 90% of 42 cases, the remainder being isolated cases of retroperitoneal fibrosis, pyeloureteral junction anomaly, ureteral stenosis and obstructive megaureter (1).

Diagnostically, the most frequent clinical signs are lumbar pain, fever, malaise and weight loss, usually evolving for more than six months. In our case, the course of the disease lasted 02 years, testifying to a frequent delay in consultation in our context, linked to the low socio-economic standard of living of our populations, recently aggravated by the security challenge the country has been facing for a decade.

XGP presents in two forms: a diffuse form, which is the most frequent and corresponds in fact to pyonephrosis, as was the case in our patient, and a focal pseudotumoral form, which raises the differential diagnosis of a renal mass (5). Radiologically, the preoperative diagnosis of XGP is made in only about half of cases, mainly on the basis of CT scan data (5). In our case, the preoperative diagnosis was

evoked by the long history of febrile low back pain, associated with a biological inflammatory syndrome (hyperleukocytosis, inflammatory anemia) and a uro-CT scan showing the "Bear paw sign": multiple hypodense cavities surrounded by partitions. This suggestive appearance has been described by several authors as highly characteristic of XGP (6-8).

Treatment must be aggressive, combining percutaneous drainage of the suppuration, antibiotic therapy and nephrectomy in the majority of cases. Bitherapy combining a third-generation cephalosporin and an aminoglycoside, in this case amykacin, is recommended as probabilistic treatment before being secondarily adapted to the antibiogram in accordance with the recommendations of the French-Speaking Society of Infectious Pathology (SPILF) on the management of severe pyelonephritis, 2018 update. Percutaneous drainage via nephrostomy is a key step in treatment, enabling purulent collections to be evacuated, helping to control infection and stabilize the patient before possible surgery (1). The standard treatment for XGP is nephrectomy, which can be performed openly or laparoscopically. Surgery can be difficult due to perirenal adhesions, making excision delicate as in our patient's case, and sometimes necessitating an open approach rather than laparoscopy (9). Recent studies, however, suggest that laparoscopic nephrectomy is a safe and effective alternative to open surgery for the treatment of XGP, offering advantages in terms of postoperative pain and recovery (10).

4. Conclusion

XGP is a rare but potentially serious pathology, the diagnosis of which should be raised in the setting of chronic pyelonephritis, particularly in the presence of obstructive lithiasis. CT scanning plays a key role in preoperative diagnosis. In most cases, treatment involves bi-antibiotic therapy + percutaneous drainage and nephrectomy. Histological confirmation is essential for a definitive diagnosis.

Conflict of Interest: The authors declare no conflict of interest.

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