

## Endourology

# A rare presentation of xanthogranulomatous pyelonephritis and emphysematous pyelonephritis complicated with necrotizing fasciitis of scrotum and perineum (fournier gangrene)

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## ABSTRACT

We report a case of cooccurrence of xanthogranulomatous and emphysematous pyelonephritis along with necrotizing fasciitis (NF) of scrotum and premium in a patient was nondiabetic and the history of kidney stones. This case a unique presentation of NF due to two pyelonephritis causing fournier gangrene. Surgical management and antibiotics provide an effective management of the disease.

## Introduction

Necrotizing fasciitis (NF) is a rare soft tissue infection that causes the destruction of tissues due to necrosis and life-threatening sepsis. Malignancy, hepatic and renal dysfunction, alcohol abuse, diabetes, leukemia, vascular disease and drug abuse can increase the risk of NF. It can also be a result of skin trauma and surgical wounds in the perineum and lower limbs. Primarily, it is characterized by localized pain, abnormal vital signs and hemorrhagic bullae. Immediate management and surgical intervention is required, with antibiotics.<sup>1</sup> The inflammatory process can affect the soft tissues of the pelvis and abdomen, organs and other areas, and is difficult to be detected in the early stages.<sup>2</sup> Xanthogranulomatous pyelonephritis is a chronic kidney infection that usually results from obstruction of the renal system. Rarely, emphysematous pyelonephritis can develop as a complication of xanthogranulomatous pyelonephritis where the diagnosis can be challenging.

We report a case of cooccurrence of xanthogranulomatous and emphysematous pyelonephritis along with necrotizing fasciitis (NF) of scrotum and premium in a patient was nondiabetic and the history of kidney stones. This case a unique presentation of NF due to two pyelonephritis causing fournier gangrene. Surgical management and antibiotics provide an effective management of the disease.

## Case presentation

A 30-year-old man was reported to our center with lower abdomen pain and flank pain on the right side. He did not have any history of diabetes or use of immunosuppressive drugs or narcotics. He had a history of kidney stones and his vitals were unstable: respiratory rate 30/min, pulse rate: 160/min, blood pressure: 80/100 and temperature = 39 °C.

He underwent generalized abdominal examination with skin crytography from RUQ (right upper quadrant) to RLQ (right lower quadrant), LLQ (left lower quadrant), and extensive gangrene of the scrotum and perineum (Fig. 1).

The patient was hospitalized and treated with broad-spectrum antibiotics, serum therapy, and necrotic tissue debridement was performed. His labs showed:

White blood cells = 32100, platelet = 465000, blood urea nitrogen (BUN) = 116, creatinine (Cr) = 4.2, erythrocyte sedimentation rate = 124, Na = 125, K = 5.6, Blood sugar = 105, Hb = 9.

Due to the high BUN and Cr, the patient underwent CT scan of the abdomen and pelvis without venous contrast (Fig. 2). The findings from CT scan showed the right kidney containing several large stones along with air bubbles inside the renal parenchyma and the tissue around the kidney. There was a collection of fluid containing air around the right kidney, which spread to the subcutaneous tissue of the right flank. Based on this evidence, the diagnosis of xanthogranulomatous pyelonephritis

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**Fig. 1.** Extensive gangrene of the scrotum and perineum.



**Fig. 2.** CT scan of the abdomen and pelvis without venous contrast.

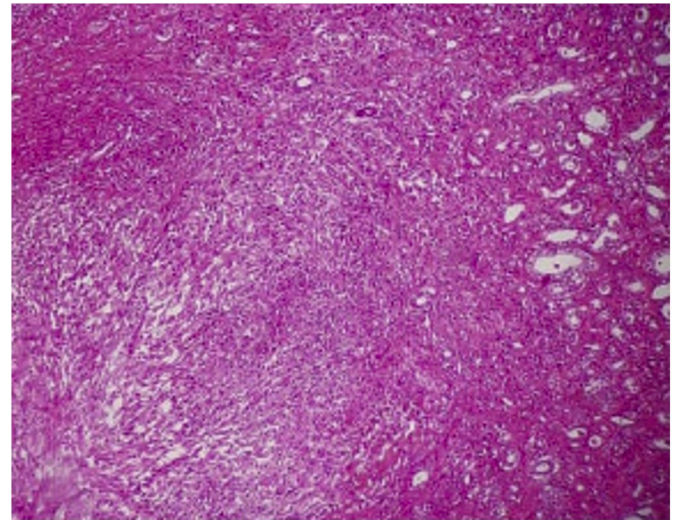
with necrotizing fasciitis caused by retroperitoneal abscess was proposed.

First, necrotic tissue debridement of the flank, scrotum, and penis were performed, and retroperitoneal abscess drainage and then right nephrectomy were performed. The patient was discharged after three weeks in good general condition.

The pathology findings indicated xanthogranulomatosis, which was characterized by destruction of kidney tissue, granulomas, abscesses, and accumulation of macrophages containing lipids (Fig. 3).

## Discussion

This case reported is a presentation of NF of scrotum and perineum (fournier gangrene) as a result of xanthogranulomatous and



**Fig. 3.** The pathology findings indicated all xanthogranulomatosis.

emphysematous pyelonephritis in a patient with kidney stones. Xanthogranulomatous pyelonephritis is an uncommon form of chronic infectious pyelonephritis (incidence 1 in 4100000) characterized by the presence of granulomas and abscesses along with the accumulation of fats in macrophages. It is more common in females and is characterized by flank pain, fever, palpable mass, urinary tract dysfunction, hematuria and weight loss.<sup>3</sup> Infection of renal parenchyma with gas accumulation is known as emphysematous pyelonephritis. Xanthogranulomatous pyelonephritis and emphysematous pyelonephritis are rarely presented with cooccurrence.<sup>4</sup>

NF is a rare skin infection that is diagnosed in CT as unsymmetrical fascia thickening with gas bubble. Diabetic and immunosuppressed patients presented with these findings should be suspected with NF. Ishigami et al.<sup>5</sup> presented a case report of NF with Xanthogranulomatous and emphysematous pyelonephritis of the left flank in inferior lumbar triangle in an immunosuppressed patient.

## Conclusion

However, our patient had no such history. CT showed the two pyelonephritis along with gas bubbles. This case signifies the rare complication of the occurrence of two pyelonephritis in nondiabetic and non-immunosuppressed patients at a new anatomical site.

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## Ethical approval and consent to participate

All procedures performed in this study involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

## Consent for publication

Not applicable.

## Availability of data and material

Data sharing is not applicable to this article as no datasets were generated or analyzed during the current study.

### Declaration of competing interest

The authors deny any conflict of interest in any terms or by any means during the study. All the fees provided by research center fund and deployed accordingly.

### CRediT authorship contribution statement

**Leila Haji Maghsoudi:** Conceptualization, Writing - original draft, Writing - review & editing. **Mojdeh Navab:** Writing - original draft, Writing - review & editing, Data curation, Supervision. **Haleh Pak:** Data curation, Writing - original draft, Writing - review & editing.

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