folia medica

Folia Medica 67(3)
DOI: 10.3897/folmed.67.e139995

Case Report

Xanthogranulomatous pyelonephritis caused by an unusual pathogen *Raoultella ornithinolytica*: a case report

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Received: 22 October 2024 ♦ **Accepted:** 11 December 2024 ♦ **Published:** 16 June 2025

Citation: Kutlu A, Erbin A, Ozdemir T, Sagir AE, Gurbuz BC, Canat HL. Xanthogranulomatous pyelonephritis caused by an unusual pathogen *Raoultella ornithinolytica*: a case report. Folia Med (Plovdiv) 2025;67(3):e139995. doi: 10.3897/folmed.67.e139995.

Abstract

Xanthogranulomatous pyelonephritis (XGP) is a rare and severe chronic inflammatory condition of the kidney. XGP's exact etiology remains unclear, although it is frequently associated with chronic urinary obstruction and infections within the urinary tract. Superinfections such as *Escherichia coli*, *Proteus mirabilis*, and occasionally *Pseudomonas* species typically cause XGP. Documented cases of *Raoultella ornithinolytica* (*R. ornithinolytica*) infections in humans, particularly within the urinary tract, are limited. However, advancements in diagnostic techniques are expected to increase the detection of such rare pathogens. To the best of our knowledge, currently, there is no evidence in the literature indicating that this pathogen is responsible for XGP. We present a case of XGP in which we isolated *R. ornithinolytica* and discuss its treatment in the current literature.

Keywords

gram-negative bacteria, kidney disease, xanthogranulomatous pyelonephritis, *Raoultella ornithinolytica*, rare pathogens, urinary tract infections

Introduction

Xanthogranulomatous pyelonephritis (XGP) is a chronic inflammatory condition characterized by destructive granulomatous inflammation within the renal parenchyma. It is defined as the substitution of normal renal or perirenal tissue with granulomatous tissue, which contains lipid-laden macrophages. The most common clinical symptoms in XGP patients are flank pain, fever, weight loss, macroscopic hematuria, palpable mass, and urinary tract symptoms.[1] Various risk factors have been identified, including obstructive uropathy, diabetes, obesity, recurrent urinary tract infections, and immunosuppression.[2] Xanthogranulomatous pyelonephritis is often confused with a true neoplasm, most commonly renal cell carcinoma, due to its similar clinical and radiographic characteristics and ability to involve adjacent structures or organs. Therefore, early identification and treatment are paramount to decreasing the associated morbidity and mortality rates. Although antibiotics can be given for acute infection, the usual treatment for XGP is a partial or total radical nephrectomy.[3] The organisms most commonly associated with XGP are *Escherichia coli* and *Proteus mirabilis*, followed by *Pseudomonas*, *Enterococcus faecalis*, and *Klebsiella*.[4] We present an XPG case in which we isolated the bacteria *R. ornithinolytica* and discussed it in light of current literature.

Case presentation

An 81-year-old woman patient presented to our department with worsening general condition, fever, and left lower back pain. In the past history, she had anemia, and insulin injections that controlled for diabetes mellitus. Her physical exam revealed only discomfort in the left costo-vertebral angle. Her fever endured for 5 to 6 days; subsequently, she exhibited no further symptoms of fever. The laboratory tests showed that the white blood cell (WBC) count was 5340/µL, the C-reactive protein level was 45.6 mg/L (the normal range is 0.5–5 mg/L), the procalcitonin level was 0.12 ng/mL (the normal range is 0–0.05 ng/mL), and the creatinine level was 1.12 mg/dL (the normal range is 0.5–0.9 mg/dL). Computed tomography (CT) revealed the presence of several air-density collection sites in the posterior medial region of the left kidney, located within the psoas compartment, with the largest measuring 6×2 cm (possibly an abscess). No evidence of stones was present ***(Fig. 1)****. The right kidney was evaluated as normal in terms of its position, size, parenchymal density, thickness, and collecting system.*

Based on these findings, the patient was diagnosed with stage 3 xanthogranulomatous pyelonephritis. We started with intravenous meropenem 3×1 g. We inserted a drainage catheter for the abscess, and approximately 30 cc of purulent fluid flowed from it every day. The urine culture revealed urogenital flora, while the abscess culture revealed Raoultella ornitolyticha, susceptible to meropenem, gentamicin, and tazobactam, and resistant to ampicillin, ciprofloxacin, levofloxacin, cefepime, and several other cephalosporines ***(Table 1)****.*

Table 1. Antibiogram of the isolated *Raoutella ornithinolytica*

Antibiotics	MIC value	Antimicrobial sensitivity
Gentamicin	≤2	Sensitive
Meropenem	≤2	Sensitive
Ampicillin/sulbactam	4/8	Sensitive
Piperacillin/tazobactam	≤4/4	Sensitive
Trimethoprim/sulfamethoxazole	>8/152	Resistant
Ciprofloxacin	1	Resistant
Levofloxacin	1	Resistant
Ceftriaxone	>4	Resistant
Cefepime	>8	Resistant
Ceftazidime	>8	Resistant
Cefuroxim	>16	Resistant
Ampicillin	>16	Resistant

MIC: minimum inhibitory concentration

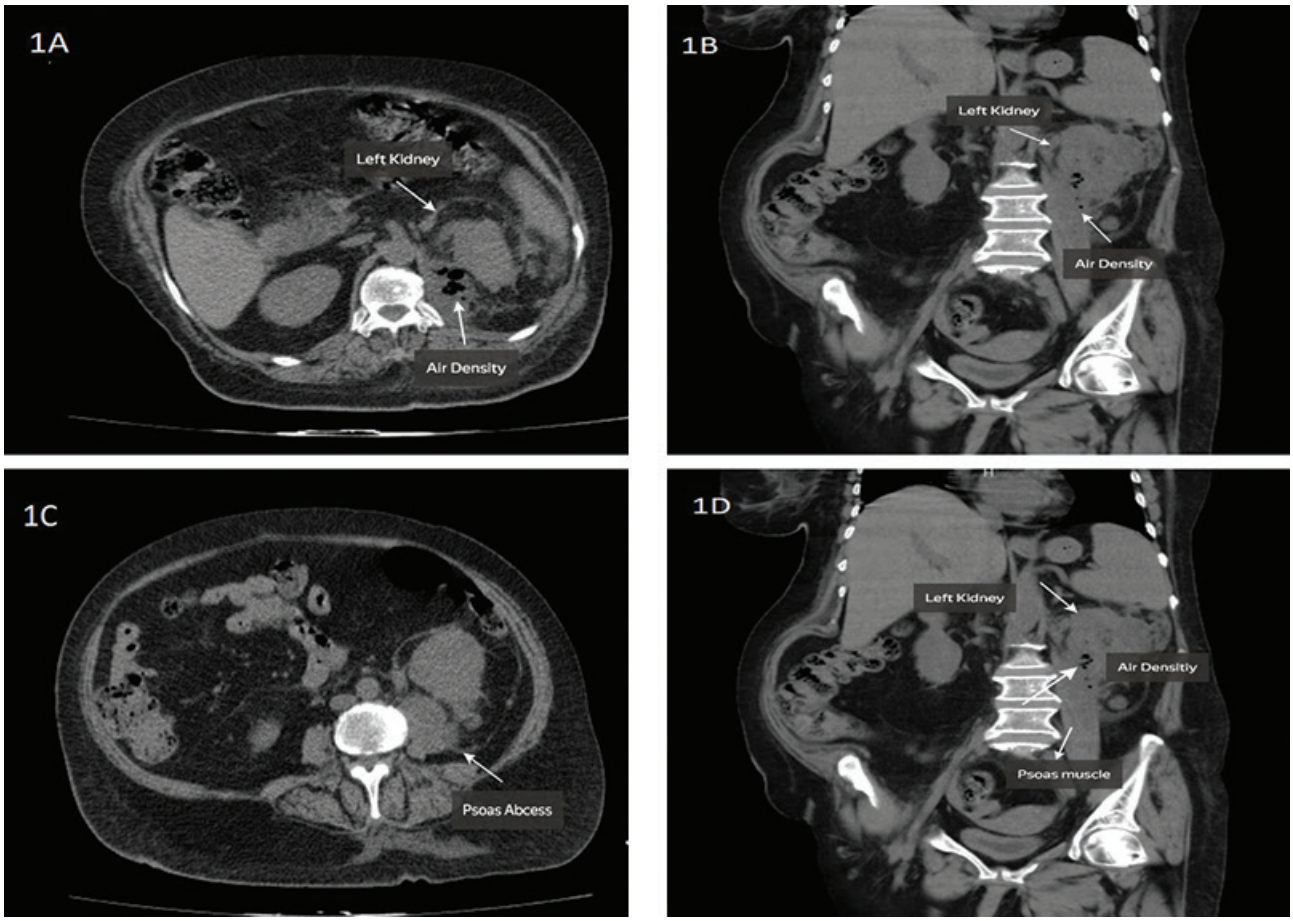

Figure 1. CT scanning images. The transvers sections (**1A, 1C**); coronal sections (**1B, 1D**).

At 14 days of treatment, the patient's symptoms largely resolved, with CRP levels decreasing by 35.6 mg/L, WBC at 7140/μL, and creatinine at 0.78 mg/dL. The patient was discharged for outpatient scintigraphy evaluations and surgical planning.

The dimercaptosuccinic acid (DMSA) scan indicated that the left kidney exhibited diminished and hypoplastic cortical function with diffuse low uptake, while the right kidney displayed a hypoactive region in the lower pole, with functional capacities of 24.7% for the left kidney and 75.3% for the right kidney ***(Fig. 2)****.*

The patient underwent a transperitoneal laparoscopic nephrectomy. The case revealed advanced adhesions surrounding the psoas muscle abscess at the lower pole level of both the kidney and the colon. The procedure was completed laparoscopically without any complications (operation time: 3 hours). At the end of the procedure, the Jackson-Pratt (JP) drainage catheter was placed ***(Fig. 3)****.*

The postoperative phase was uneventful, and the patient was discharged on the fifth postoperative day. The pathological examination revealed results compatible with XGP ***(Fig. 4)****.*

Discussion

Raoultella ornithinolytica is recognized as an environmental pathogen that infrequently causes human infections; however, its significance as a pathogen has increased in recent years. Research conducted by Seng et al.[5] indicated that *R. ornithinolytica* is implicated in several infection types: 36% of urinary tract infections, 14% of gastrointestinal infections, 13% of wound and skin infections, and 5% of bacteremia cases. Furthermore, it is associated with uncommon conditions including osteomyelitis, meningitis, brain abscesses, mediastinitis, pericarditis, conjunctivitis, and otitis. In the urogenital system, *R. ornithinolytica* has been associated with cystitis, prostatitis, pyelonephritis, and renal cyst infections.[5] No documented report regarding XPG exists to date. This case can be regarded as the first case in the literature.

Malek and Elder[6] have classified XGP into three distinct stages: stage 1: nephric, indicates only renal involvement; stage 2: perinephric, characterized by the involvement of perirenal fat; and stage 3: paranephric, characterized by significant involvement in the retroperitoneal region.[6] Stage

Figure 2. DMSA scanning images. Posterior view, small left kidney with decreased uptake **(2A);** split functions between left and right kidney; 24.7% and 75.3%, respectively **(2B)**.

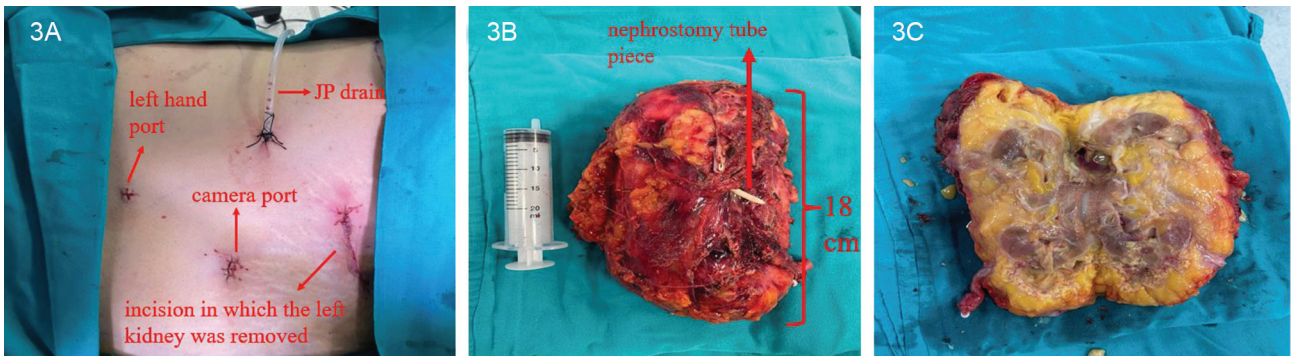

Figure 3. The operation site **(3A)**; exterior view of removed kidney **(3B)**; interior view of removed kidney **(3C)**.

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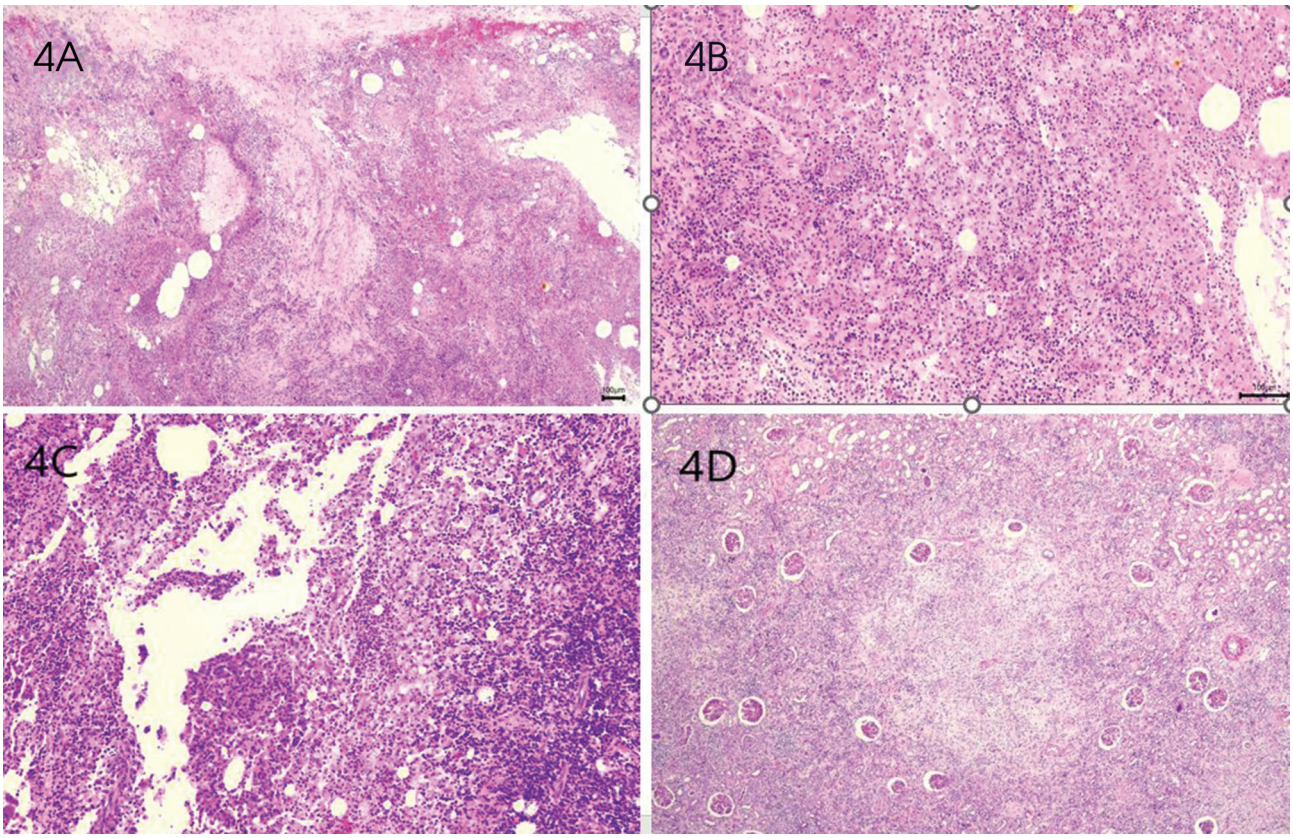

Figure 4. Inflammatory infiltration consisting of histiocytes and lymphocytes **(4A)**; Xanthomatous proliferation composed of foamy cytoplasms, accompanied by lymphocytes **(4B)**; Xanthomatous proliferation composed of foamy cytoplasms with areas of abscess formation, accompanied by lymphocytes **(4C)**; Interstitial fibrosis in the renal parenchyma with chronic inflammatory cells **(4D)**.

3 is associated with a significantly elevated requirement for nephrectomy. A recent systematic review involving over 1000 patients indicated that while standard treatment remains antibiotics and open radical nephrectomy, minimally invasive laparoscopic nephrectomy was performed in 34% of cases, and partial nephrectomy was conducted in 2% of cases. Nevertheless, more than 10% of these cases necessitated a conversion to open surgery.[7] During the laparoscopic nephrectomy procedure, significant adhesions and fibrosis in the surrounding tissues were encountered, complicating dissection. We successfully completed the procedure using a laparoscopic approach. In these instances, the necessity for open surgery and associated complications may be diminished when conducted by a specialist who has experience in advanced laparoscopic surgery, as demonstrated in our case (the procedure was performed by Dr. A.E.). A recent study indicated that anemia and the presence of abscesses are significant perioperative factors predicting complications during the perioperative period.[8] Our patient had both risk factors.

Conclusion

In conclusion, *R. ornithinolytica* is capable of inducing severe kidney infections and abscesses in the perirenal region, particularly in individuals with systemic risk factors. XPG cases caused by *R. ornithinolytica* can be managed with laparoscopic surgery performed by skilled practitioners, in conjunction with suitable antibiotics. Additional research is necessary to enhance the understanding of the implications of these infections and their management strategies.

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