Case report

Xanthogranulomatous prostatitis presenting as Pseudomonas aeruginosa prostatic abscesses: An uncommon complication after kidney transplantation

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ABSTRACT

Xanthogranulomatous (XG) prostatitis is a rare form of granulomatous prostatitis characterized by a benign inflammatory process of non-specific etiology that clinically may mimic carcinoma. Few cases have been reported in the English language medical literature, with only four reported cases presenting as prostatic abscesses. A 70-year-old male with type 2 diabetes mellitus and two previous kidney transplants presented with septic shock secondary to Pseudomonas aeruginosa bacteremia 4 days after undergoing a cystoscopy. Despite appropriate antimicrobial therapy, P. aeruginosa persisted in the blood for a total of 7 days. There were no indwelling prosthetic devices, no complicated pyleonephritis, and no endovascular sources of infection. Upon repeat clinical assessment, the patient reported pelvic pain. A digital rectal examination revealed prostatic tenderness and an endorectal ultrasound confirmed multiple prostatic abscesses. An ultrasound-guided transrectal needle aspirate drained scant purulent fluid and cultures grew the same phenotypic strain of P. aeruginosa. For definitive source control, the patient underwent transurethral resection of the prostate with unroofing of prostatic abscesses. The pathological findings were diagnostic of XG prostatitis. Given the rather acute presentation of this case, our hypothesis is that the prior urological instrumentation likely facilitated bacterial translocation and created the ideal environment for the development of pseudomonal prostatic abscesses resulting in XG inflammation and necrosis. XG prostatitis is a rare entity of uncertain etiology that can result in prostatic abscesses, and surgery is required for definitive diagnosis and management.

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Introduction

Xanthogranulomatous (XG) prostatitis is a rare form of granulomatous prostatitis characterized by a benign inflammatory process of non-specific etiology that clinically may mimic carcinoma [1]. Few cases have been reported in the English literature, with only four reported cases presenting as prostatic abscesses [2–5]. Infectious etiologies of granulomatous prostatitis include bacteria, fungi, viruses, and parasites [5]. Mycobacterium tuberculosis and fungi, particularly blastomycesis, coccidioidomycosis, and cryptococcosis constitute the most common infectious causes [6]. The Epstein and Hutchins classification of granulomatous prostatitis is controversial but commonly used; it divides it into four types: nonspecific (idiopathic), specific (infectious), iatrogenic, and secondary to systemic granulomatous diseases [7]. XG prostatitis is usually considered a rare form of nonspecific prostatitis [6,8].

Case report

A 70-year-old male with type 2 diabetes mellitus, and previous kidney transplantation in 2007 and 2017 for end-stage renal disease secondary to focal segmental glomerulosclerosis and chronic antibody-mediated rejected respectively, presented to the intensive care unit with distributive shock requiring vasopressor support. He was empirically started on piperacillin–tazobactam and 19 h later his blood cultures bottles grew a non-lactose fermenting gram-negative bacillus. Of note, the patient had undergone a cystoscopy for microscopic hematuria 4 days prior to this presentation, and did not recall
having received peri-procedural antimicrobials. Cystoscopy findings were the presence of bilobar prostatic hypertrophy but no significant macroscopic abnormalities in the bladder.

The blood culture isolate was later identified as *Pseudomonas aeruginosa*, susceptible to all anti-pseudomonal antimicrobials, and piperacillin-tazobactam dosing was appropriate. The patient rapidly defervesced and normalized his hemodynamic parameters and was later transferred to the ward where he was transitioned to oral ciprofloxacin. Despite clinical improvement, he had persistent *P. aeruginosa* bacteremia and thus high-dose ceftazidime was added to the antimicrobial regimen. Nevertheless, the patient remained persistently bacteremic for a total of 7 days. There were no indwelling catheters or prothetic devices, no perinephric abscess or hydronephrosis, no clinical or radiographic evidence of pneumonia, and no known cardiac valvulopathy or arterial aneurysms. The patient had ongoing delirium and upon repeat clinical assessment he reported rectal pain. A digital rectal examination revealed an enlarged and tender prostate which was followed up by an endorectal ultrasound that confirmed prostatomegaly, increased vascularity and multiple prostatic abscesses, the largest measuring 3.4 cm of greatest diameter (Fig. 1). An ultrasound-guided transrectal needle aspirate of the prostate drained only small amounts of very thick purulent fluid and cultures grew the same phenotypic strain of *P. aeruginosa*.

For definitive source control, the patient underwent transurethral resection of the prostate gland with unroofing of prostatic abscess. The surgical pathology specimen revealed sheets of foamy histiocytes, abundant necrotic debris and rare hemosiderin deposition (Fig. 2A–B). Auramine-rhodamine and Grocott-Gomori’s methamine silver stains were negative for acid-fast bacilli and fungi, respectively. The histiocytes did not express cytokeratin AE1/AE3 by immunohistochemistry, excluding the possibility of an epithelial neoplasm. The pathologic findings were ultimately diagnostic of XG prostatitis.

After surgery the patient completed 10 days of oral ciprofloxacin but shortly thereafter, surveillance urine cultures grew *Pseudomonas aeruginosa* and the patient reported new lower urinary tract symptoms. A decision was made to pursue 6 additional weeks of ciprofloxacin, which the patient was able to complete without developing significant adverse events. In a follow-up assessment one month after discontinuing the antimicrobials, the patient was clinically doing very well with no infectious or urinary tract symptoms.

**Discussion**

Herein, we report an unusual case of XG prostatitis with multiple abscesses as the source of persistent *P. aeruginosa* bacteremia. As the patient was initially critically ill with fluctuating mental status, it only became apparent that he had prostatic tenderness 7 days into his persistent bloodstream infection. Endovascular sources were appropriately sought for but prostatic abscesses were not immediately considered given the lack of localized symptoms due to the patient’s delirium. This case is rather unique as it occurred in a transplant recipient presenting with a common bacterial infection secondary to an uncommon disease. XG pathologic findings are well described in the kidney and gallbladder but the prostate is a rare site for this entity [1]. Most described cases corresponded to adult patients in the sixth decade of life [8].

On histopathology, XG prostatitis exhibits a large number of pale-looking lipid-laden macrophages, so-called foamy histiocytes, and accumulation of inflammatory cells including lymphocyte, plasma cells and occasionally polymorphonuclear cells and eosinophils.
A possible pathophysiologic mechanism for XG prostatitis involves blockage of prostatic ducts and stasis of secretions resulting in an intense localized inflammatory response [6,9]. The incidence of prostatic abscesses has been reported as low in the antimicrobial era [10]. Risk factors include diabetes mellitus, chronic kidney disease, and immunosuppression [11], all of which were present in our patient. XG pyelonephritis is known to rarely occur in renal allografts [12], but little is known about the pathophysiology of XG prostatitis in this population.

Given the rather acute presentation of our case, we hypothesize that the prior urological instrumentation may have facilitated bacterial invasion and created the ideal environment for the development of pseudomonal prostatic abscesses resulting in XG inflammation and necrosis. Alternatively, previous microscopic hematuria could have resulted from XG prostatitis due to chronic urinary tract infections and immune dysregulation with bacterial sepsis triggered by cystoscopy.

In the kidney, XG pyelonephritis is thought to result from abnormal inflammatory response to bacterial infection [12]. There has been at least 8 reported cases of XG pyelonephritis in renal transplant recipients [12], of which the most common identified pathogen was Escherichia coli followed by Klebsiella spp. However, this is the first reported case of XG prostatitis in a transplant recipient. We postulate that the chronic rejection resulting in chronic allograft dysfunction predisposed to recurrent or chronic subclinical infection which may have triggered an abnormal immune response.

Presently, the occurrence of prostatic abscesses is relatively rare but should be considered in the differential diagnosis of persistent P. aeruginosa or other gram-negative bacilli bloodstream infections. To our knowledge, only 4 cases of XG prostatitis presenting with prostate abscesses have been published to date (Table 1). All published cases required prostate resection. Although XG prostatitis is an uncommon entity of uncertain etiology, its differential diagnosis includes a number of potential infectious causes.
Table 1
Summary of reported cases of XG prostatitis presenting with prostatic abscesses.

| Case | Age | Presenting symptoms | Predisposing risk factors | Pathogen | Antibiotic therapy | Surgery | Year of publication |
|------|-----|---------------------|---------------------------|----------|--------------------|---------|---------------------|
| 1    | 82  | Gross hematuria, voiding difficulty | None | Gram-positive cocci | Yes | TURP | 2011 |
| 2    | 65  | Sepsis of unknown origin | DM, immunosuppression (anti-TNFα) | Staphylococcus aureus (methicillin-sensitive) | Yes (> 4 weeks) | TURP | 2013 |
| 3    | 52  | High-grade fever and acute urinary retention | None | Unknown | Yes | TURP | 2012 |
| 4    | 59  | LUTS, gross hematuria, fever | DM | Unknown | Unknown | TURP | 2018 |
| 5    | 70  | Septic shock | DM, immunosuppression (kidney transplant), CKD | Pseudomonas aeruginosa | Yes (ciprofloxacin > 6 weeks) | TURP | 2019 |

TURP: Transurethral resection of the prostate; DM: Diabetes mellitus; LUTS: Lower urinary tract symptoms; CKD: Chronic kidney disease.

Conclusion

XG prostatitis is a rare entity of unknown etiology that can mimic prostate adenocarcinoma. Our case constitutes the 5th published report of XG prostatitis with prostatic abscesses and the first case presenting with pseudomonal sepsis. The definitive therapeutic management of XG prostatitis requires surgical resection and the final diagnosis is made by histopathology.

Author contributions

The author contributions were as follows:

- Conceptualization: Sara Belga and Carlos Cervera.
- Supervision: Carlos Cervera.
- Writing – original draft: Sara Belga.
- Writing – review & editing: Will Chen, Gavin Low and Carlos Cervera.
- Pictures: Will Chen and Gaving Low.

All authors were involved in the patient’s care.

Conflicts of interest

None.

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